Health Technology Assessment. Polysomnographie et monitoring à domicile des nourrissons en prévention de la mort subite

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Préface

Chaque année, on estime que neuf enfants sur dix mille meurent de façon inexpliquée durant leur sommeil avant leur premier anniversaire. C'est à la fois peu et beaucoup trop, tant il est vrai que le décès d'un enfant reste inacceptable.

La polysomnographie, soit un test du sommeil, et les monitorings cardio respiratoires à domicile qui peuvent s'ensuivre ont été, pendant des années, pratiqués à une très large échelle en Belgique afin de détecter et suivre les nourrissons à risque de mort subite.

La question qui se pose est de savoir pour quelle part la polysomnographie a contribué à la réduction de 60% de la mortalité infantile des moins d'un an observée dans les années 90. Ce succès semble plutôt attribuable aux règles de prévention assez simples comme coucher l'enfant sur le dos, offrir un environnement de sommeil adéquat et l'arrêt éventuel du tabagisme des mères.

L'usage de polysomnographie a diminué au cours des dernières années, sauf pour des enfants de moins d'un an présentant des indications très spécifiques, mais demeure plus élevé qu'à l'étranger et la grande variabilité de son utilisation en Belgique reste sans explication.

Doit-on applaudir cette diminution progressive des polysomnographies en Belgique ou au contraire être attentif quant à une sous-utilisation de ce test ? Après tout, cet examen peut-il être utilisé comme 'test de la mort subite' et l'information aux parents quant aux avantages et inconvénients du test est-elle adéquate ?

Ces questions ne sont pas anodines et sont fortement chargées d'un point de vue émotionnel. Le Centre d'expertise a choisi d'y répondre sous la forme d'un Health Technology Assessment, approche pluridisciplinaire dans laquelle sont évoquées aussi bien les aspects médicaux et économiques que le point de vue du patient (ici les parents). Nous formons le vœux que tous les acteurs concernés y trouvent les réponses aux questions qu'ils se posent : décideurs au plus haut niveau, parents, médecins, paramédicaux et institutions en charge du bien-être et de l'accueil de la petite enfance.

Jean-Pierre Closon
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Directeur général

Introduction

La polysomnographie (PSG) est un examen du sommeil réalisé durant la nuit qui mesure certains paramètres physiologiques, en particulier cardiorespiratoires. Si des anomalies sont identifiées chez le nourrisson examiné, celui-ci sera placé sous la surveillance d'un moniteur cardio-respiratoire (CR) à son domicile. En 2004, 43.634 polysomnographies ont été réalisées dans des hôpitaux belges dont 19.335 (44%) chez des enfants de moins d'un an. Pour l'année 2003 nous avons estimé les coûts des PSG financés par des fonds publics à près de 10.000.000€ pour ce groupe (environ 500€ par PSG). Le coût du monitoring a domicile atteint plus de 4.000.000 €.

En Belgique, une PSG est en général réalisée chez les enfants de moins d'un an pour les indications suivantes :

- Pour identifier les enfants à risque d'incidents cardio-respiratoires ; partant du principe que ces incidents sont détectés par une PSG, qu'ils indiquent un risque d'événements CR récurrents et que ces derniers peuvent compromettre le développement psychomoteur du nourrisson. Grâce à une surveillance par moniteur à domicile, ces incidents peuvent être écourtés et leur impact dommageable limité ;
- Comme partie d'une investigation générale afin d'établir un diagnostic, comme dans le cas de malaise manifestement mortellement grave de type Apparent Life Threatening Event (ALTE) ;
- Pour identifier les enfants à risque de décès inopiné partant du principe que la PSG peut identifier des anomalies qui indiqueraient un risque de mort subite du nourrisson (MSN) accru. Les enfants qui présentent une PSG anormale seront alors placés sous surveillance d'un moniteur à domicile en prévention de la MSN.

Une ou plusieurs des indications mentionnées ci avant peuvent s'appliquer à différents groupes de nourrissons : les nourrissons nés prématurés (avant 37 semaines gestationnelles) ou dysmatures (présentant un poids de naissance trop petit pour leur âge gestationnel), les nourrissons qui ont présenté un malaise de type ALTE, les enfants souffrant de problèmes médicaux spécifiques (syndrome Pierre-Robin, affections neuromusculaires, etc.), les frères ou soeurs d'enfants décédés de MSN et les enfants en bonne santé, qui passent le test sans indication précise.

	Indications actuelles pour l'utilisation de la PSG en Belgique chez des nourrissons		
Groupes de nourrissons	**Incidents cardio-respiratoires**	**Recherche de diagnostic**	**Identification du risque de décès inopiné**
Prématurés et dysmatures	X		X
ALTE	X	X	X
Problèmes médicaux spécifiques	X	X	
Frères/sœurs d'enfants décédés de MSN			X
En bonne santé			X

Cent décès attribués au syndrome de la mort subite ont eu lieu en Belgique en 1997, soit 9 cas/10.000 naissances vivantes. La MSN est 'le décès inopiné et inattendu d'un enfant âgé de mois d'un an qui reste inexpliqué après un examen post-mortem approfondi, incluant une autopsie complète, un examen du lieu du décès et un revue de l'histoire médicale du nourrisson'. Malgré un déclin de plus de 50% dans plusieurs pays (déclin attribuable en grande partie aux campagnes d'éducation préconisant que le nourrisson soit placé sur le dos pour dormir), la MSN reste la première cause de décès entre l'âge de 28 jours et d'1 an, contribuant à 25% des décès dans ce groupe d'âge.

Objectifs et méthodes

Les objectifs de cet Health Technology Assessment sont :

(1) Evaluer la valeur des polysomnographies nocturnes et le recours au moniteur à domicile chez les enfants de moins d'un an, particulièrement en prévention de la MSN. Cette partie a été réalisée au travers d'une analyse critique de la littérature scientifique clinique et économique relative à ce sujet.

(2) Etudier l'utilisation des PSG et des moniteurs à domicile chez les enfants de moins d'un an en Belgique et leur coût pour la sécurité sociale. Nous avons analysé les données relevantes dans les bases de données disponibles (séjours hospitaliers, codes de facturation, données des mutuelles) et nous avons mené une étude qualitative exploratoire auprès de professionnels de la santé et de parents d'enfants qui ont passé une PSG.

Effectivité clinique de la PSG et du moniteur à domicile pour les enfants de moins d'un an

Efficacité technique

Il ressort de la littérature que tant pour la PSG chez les jeunes enfants que pour le monitoring à domicile, des données standard validées font défaut. La validation technique de ce type d'appareil fait également défaut. A cela s'ajoute le manque de garanties suffisantes quant à la qualité de l'interprétation des résultats. Ces questions se posent encore plus précisément pour les moniteurs qui sont proposés à l'achat ou à la location dans le circuit commercial.

Utilisation de la PSG et du moniteur à domicile pour identifier les incidents cardio-respiratoires

Nous n'avons pas trouvé dans notre analyse critique de la littérature de données probantes pour répondre aux questions suivantes :

(a) Les incidents cardio-respiratoires récurrents chez des enfants à risque de tels incidents sont-ils dommageables – en particulier pour leur développement psychomoteur ultérieur ?

(b) Y a-t-il une corrélation entre les incidents cardio-respiratoires identifiés par une PSG chez des prématurés ou des nourrissons dysmatures et la survenue d'incidents cardio-respiratoires récurrents dans les mois qui suivent ?

(c) La surveillance à domicile par moniteur d'incidents cardio-respiratoires peut-elle améliorer le développement psychomoteur des patients identifiés comme à risque pour ces incidents par une PSG ?

En ce qui concerne l'utilité de la PSG pour clarifier un diagnostic sous-jacent pour un nourrisson qui a présenté un ALTE, nous n'avons trouvé aucune données permettant d'identifier les cas où une PSG peut aider à poser un tel diagnostic. Nous n'avons trouvé aucune recommandation ou guideline basés sur des données probantes reprenant quels examens techniques doivent être réalisés chez un enfant ayant présenté un ALTE, le diagnostic de ce dernier étant du reste (par définition) basé souvent sur l'interprétation subjective des parents.

L'utilité de la PSG chez les nourrissons souffrant d'affections médicales autres (par exemple problèmes neuromusculaires, etc.) chez lesquels on constate des symptômes cliniques d'insuffisance (cardio-) respiratoire, est communément acceptée dans la littérature, bien que peu de données probantes soient disponibles. L'indication médicale de surveillance attentive de ces enfants à domicile est évidente, seule demeure la question de savoir si le recours à un moniteur à domicile offre des garanties suffisantes. L'oxymétrie, malgré la sensibilité aux fausses alarmes et dans certains cas précis, peut constituer une alternative. Un raisonnement similaire peut être tenu quant à l'usage de moniteur à domicile pour les enfants de mère toxicomane.

La PSG et le moniteur à domicile comme outil de dépistage du risque de MSN

Parce que la MSN est un événement rare et pour des raisons éthiques évidentes, il n'existe pas d'essai contrôlés randomisés (RCT) quant à la valeur prédictive de la PSG dans l'évaluation du risque de MSN chez des enfants en bonne santé ou chez des enfants présentant un facteur de risque autre pré-établi (comme la prématurité). La littérature disponible indique sur base d'études de cohorte convaincante que la PSG ne prévient pas la survenue de MSN.

L'impact de la surveillance par moniteur à domicile sur l'incidence de MSN ne peut non plus être établi à l'aide de RCT. Des études de cohortes observationnelles à grande échelle n'ont pas détecté une incidence de MSN moindre attribuable à l'introduction d'une surveillance à domicile par moniteur.

Coût-efficacité de la PSG et du moniteur à domicile

Peu d'évaluations économiques sur le rapport coût-efficacité de la PSG et du moniteur à domicile en prévention de la mort (subite) sont rapportées dans la littérature. La littérature existante est particulièrement de mauvaise qualité. Il existe des indications comme quoi l'utilisation de moniteurs à mémoire (appareils qui enregistrent les paramètres cardio-respiratoires pendant une phase d'alarme et qui les stockent dans une mémoire électronique pour être analysés ultérieurement) serait une stratégie à coût moindre en comparaison de ceux sans fonction de mémoire pour lesquels le médecin qui fera des analyses plus approfondies ne dispose que des observations des parents.

PSG et moniteur à domicile chez les nourrissons en Belgique : description quantitative

En 2004, 19.335 PSG ont été réalisées dans des hôpitaux belges pour des enfants de moins d'un an. Le nombre total de PSG pédiatriques (c à d les PSG réalisées chez les patient de moins de 16 ans) a diminué de 31.236 en 1995 à 20.637 en 2004, soit une diminution de 34%. Etant donné la part importante que représentent les PSG réalisées chez des patients de moins d'un an, ce groupe est principalement concerné par la diminution observée.

Le nombre de PSG pédiatriques varie fortement entre les hôpitaux avec une moyenne de séjours hospitaliers (déviation standard) de 205 (246) pour 66 hôpitaux différents en 2004.

De même, le taux d'admission pour PSG dans le groupe d'âge des 0-4 ans varie fortement entre les arrondissements, allant de 7 admissions hospitalières pour PSG par 1000 enfants jusqu'à 85 pour 1000, avec une moyenne nationale de 36, une médiane de 33 et une déviation standard de 16.

Comme le révèlent les données des APR-DRG (All Patient Refined Diagnosis Related Groups), 73% de ces séjours rapportés pour PSG en 2004 concernent l'indication « autres fonctions qui influencent l'états de santé » qui est particulièrement vague, amenant à soupçonner que ces séjours concernent principalement des enfants en bonne santé.

Sur base des données des mutualités de près de 77.000 bébés nés en 2004, nous avons calculé que 15% des enfants en dessous d'un an en Belgique avaient passé au moins une PSG en milieu hospitalier. Ce nombre paraît considérablement élevé lorsqu'on le compare aux données internationales, comme nos analyses sur les données australiennes le confirment.

Au coût d'environ 500€ par séjour hospitalier pour PSG (partie financée par l'état), augmenté de 75€ pour le patient (parfois couvert par une assurance privée complémentaire) la pratique répandue de ce test chez les nourrissons en Belgique se révèle onéreuse (en n'estimant que les coûts financiers directs). Nous estimons le coût total pour la sécurité sociale à 9.632.067€ en 2003.

Les données disponibles relatives au moniteur à domicile indiquent une tendance à l'utilisation croissante d'appareil à mémoire au détriment d'appareils sans mémoire. Sur base de données des mutualités de 46.465 bébés nés en 2004, nous avons calculé une durée de surveillance moyenne pour les appareils avec et sans mémoire de 169 et 161 jours respectivement. Ces résultats vont à

l'encontre de ce à quoi on pouvait s'attendre en regard de la littérature scientifique qui rapporte que l'utilisation de moniteurs à mémoire s'accompagne d'un nombre de jour sous monitoring plus bas. Cela serait dû au moindre nombre de fausses alarmes occasionnées par ce type d'appareil. La durée plus longue du monitoring en Belgique peut refléter en partie l'application de protocoles d'utilisation sous-jacents différents (dans lesquels les patients plus sévères sont préférablement équipés de moniteur à mémoire).

Les coûts remboursés par la sécurité sociale pour le monitoring d'un enfant (qui commencent avant l'âge de 2 ans) étaient de 4.747.110€ en 2003, ce qui amène à un montant total de 14.379.177€ pour les tests PSG et le monitoring à domicile en 2003 chez les patients de moins d'un an (soit environ 0.1% des frais médicaux remboursés par des fonds publics cette année).

Perspective parentale : approche qualitative

En prenant comme point de départ la perspective parentale, nous avons cherché à éclaircir la question de savoir pourquoi tant d'enfants sont testé en Belgique.

Le corpus de nos analyses comprend des entretiens avec des représentants d'organisations de pédiatres, d'institutions publiques en charge de la santé et du bien-être de l'enfant, et avec des parents ainsi que des outils de prévention (dépliants, livrets, sites web) ainsi que des fora de discussion sur Internet.

Les différentes campagnes de prévention adressées aux parents depuis le début des années nonante ne mentionnent pas l'utilisation d'un test PSG.

Les professionnels de santé belges que nous avons interrogés disent ne pas préconiser le recours au test pour dépister le risque de MSN en général. Mais qu'il peut faire partie d'un examen plus large du patient. Nous avons relevé à ce propos l'incongruité de l'utilisation du terme de 'wiegendoodtest' dans certains documents pour désigner une PSG chez le nourrisson. Ce terme signifie littéralement 'test de la mort subite' en néerlandais. De plus, nous avons pointé dans le site web de Kind & Gezin le fait que l'inquiétude parentale était explicitement mentionnée comme raison légitime de faire passer une PSG sans autre indication médicale.

Nous avons interrogé une série de parents (de 11 enfants dans notre échantillon final) qui avaient fait testé leur enfant sans raison médicale claire. Nous avons supposé ainsi que cela nous permettrait d'identifier un éventail de motivations possibles qui inciteraient les parents à tester leur enfant par une PSG. Nous présentons nos résultats sous forme d'hypothèses nécessitant des recherches plus approfondies:

- La PSG est erronément perçue par certains parents comme un test de dépistage pour chaque enfant de sorte que ce test devrait être envisagé par chaque parent ;
- Les parents qui ont déjà fait testé un enfant aîné font également tester le(s) cadet(s) ;
- Les médecins prescrivent trop facilement une PSG, parfois sans aucune explication quant au but spécifique du test, ce qu'il mesure précisément et le suivi potentiel.

Il ressort de la littérature scientifique que le monitoring à domicile peut avoir un impact sur le bien-être émotionnel et social des parents.

Conclusions

Bien que le nombre de PSG ait déjà clairement diminué au cours des dernières années, le testing polysomnographique de nourrissons est une pratique largement établie en Belgique avec plus de 19.000 tests par an en milieu hospitalier pour des patients âgés de moins d'un an. L'utilisation de la polysomnographie peut être justifiée pour certains groupes, comme les prématurés/dysmatures, les enfants qui ont fait un ALTE et les enfants qui souffrent de quelques affections très spécifiques. Il ressort de 6 grandes études épidémiologiques que la polysomnographie ne contribue pas au dépistage et la prévention de la MSN.

Une variation considérable des pratiques est observée en Belgique selon le lieu de résidence du patient ou entre les 66 hôpitaux concernés. Pour la majorité des patients, l'indication médicale est très vague, laissant suspecter que la plupart des enfants testés sont en bonne santé. Une comparaison avec des données internationales confirme l'utilisation élevée des PSG chez les nourrissons en Belgique.

Etant donné le nombre annuel élevé d'enfants testés par PSG, il n'est pas surprenant qu'environ 40% des enfants surveillés par moniteur à domicile soient des enfants nés à terme et pour lesquels aucun malaise de type ALTE n'a été rapporté. On peut constater des incidents cardiorespiratoires fréquents grâce à la PSG chez bon nombre d'enfants. La survenue de tels incidents durant cet examen n'est certainement pas prédictive de MSN. Rassurer alors les parents dans ce cas n'est toutefois pas une sinécure et la première question à se poser dès lors est 'quelle est la nécessité d'un tel test ?'.

Le taux d'utilisation élevé d'utilisation de PSG est probablement aussi lié au taux plus élevé d'utilisation de moniteurs à domicile. Cependant en l'absence de paramètres diagnostics clairs, nous ne pouvons pas quantifier le nombre précis de faux positifs.

Notre approche qualitative suggère que les professionnels de santé ne préconisent pas l'utilisation des PSG comme examen de dépistage universel. Cependant, des incohérences émergent dans la manière dont l'information est transmise aux parents. Ces derniers ne sont pas toujours contredits dans leur croyance que la PSG devrait être une mesure de sécurité à prendre par chaque parent.

Notre étude pointe un usage inapproprié pour une partie des PSG en Belgique qui implique des coûts supplémentaires significatifs pour l'examen et les admissions hospitalières, imputés aux parents et à l'assurance maladie.

Recommandations

Polysomnographies

Pour aboutir à un usage plus 'coût-efficace' des polysomnographies qui sont réalisées chez les nourrissons en Belgique, trois stratégies complémentaires et optionnelles peuvent être suivies :

- Améliorer l'information vers le grand public et les jeunes parents via les instances concernées telles que Kind en Gezin et l'ONE ainsi que les mutuelles:
    - L'utilité limitée de la polysomnographie pour la prévention de la MSN devrait être explicitement mentionnée dans les documents et sites web relatifs à ce sujet.
    - Les informations contradictoires devraient être supprimées. Ainsi le terme de 'wiegendood' ou 'test de la mort subite' ne doit plus être utilisé. De même que les parents ne doivent plus être avertis qu'ils peuvent faire passer une PSG parce qu'ils sont très inquiets.
- Renforcer le rôle des dispensateurs de soins et des accueillantes :
    - Les médecins devraient donner une information correcte quant à l'utilité de la PSG lorsque le sujet est abordé dans la conversation. Ils doivent avant tout rappeler les mesures de prévention de la MSN afin de rassurer les parents inquiets et souligner que leur utilité est bien prouvée.
    - Des instructions sur l'utilité de la PSG, basées sur des informations fondées, devraient être renforcées auprès des dispensateurs de soins (infirmières en maternité, kinésithérapeute qui pratique la kinésithérapie pré- et post-natale) et accueillant(e)s de nourrissons.
- La réforme de la règlementation actuelle :
    - En 2004, nous avons identifié 66 hôpitaux qui réalisent des polysomnographies chez des nourrissons. Actuellement 19 hôpitaux disposent d'un service de soins intensifs néonatals. Dix d'entre eux sont de surcroît reconnus comme centre de référence en charge des monitorings à domicile pour les nouveaux nés et les nourrissons à risque de mort subite. Au total, les centres agrées sont au nombre de 12. Cela signifie que le nombre d'hôpitaux avec un service de soins intensifs néonatal augmenté du nombre de centres agrées est de 21. Ces 21 hôpitaux semblent être les candidats les plus indiqués pour effectuer des PSG chez des nourrissons. Il pourrait donc être envisagé de donner uniquement l'autorisation d'effectuer des PSG chez les nourrissons à ces hôpitaux. A noter que cela pourrait avoir des répercussions importantes sur l'activité et le financement des services de pédiatrie des 45 hôpitaux qui perdraient leur agrément.
    - La qualité de l'interprétation des polysomnographies pourrait être améliorée, par exemple en requérant une formation spécifique de tous les membres de l'équipe du sommeil.
    - Une autre alternative possible serait que les parents dont l'enfant est candidat à une PSG soient référés à un pédiatre spécialisé dans l'évaluation du besoin en polysomnographie qui déciderait de la prescription finale.

Monitoring à domicile

La réglementation actuelle indiquant que seul les moniteurs qui mesurent le rythme cardiaque et la respiration entrent en compte pour un remboursement, concorde avec les connaissances scientifiques en la matière. La qualité technique de ces appareils pourrait éventuellement être améliorée en définissant des normes au niveau réglementaire.

Il est plus difficile de répondre à la question de savoir si seuls les moniteurs à mémoire doivent être remboursés. Il n'est pas clair jusqu'où doivent être analysées toutes les alarmes des moniteurs à mémoire. La valeur ajoutée de la répétition de PSG chez un même patient par rapport à l'analyse des rapports qui ressortent d'un monitoring avec mémoire chez le même patient n'est pas plus claire.

Une utilisation excessive de polysomnographie pourrait se traduire par un usage élevé de moniteurs à domicile. Si nos recommandations tendant à un usage approprié de la polysomnographie sont suivies, on peut s'attendre à ce que le nombre d'enfants sous surveillance d'un moniteur diminue proportionnellement.

Collecte de données et recherche future

Des questions relatives à l'environnement de sommeil des nourrissons avaient été posées au cours de la première enquête nationale de santé en Belgique (1997) mais pas dans la seconde (2001) ni dans la troisième (2004). Les futures éditions de cette enquête devraient prendre à nouveau cet aspect en compte, de sorte qu'on puisse collecter sur le long terme des données de comportement permettant d'évaluer l'impact des différentes campagnes de prévention.

Actuellement les 12 centres reconnus ne doivent pas communiquer à l'INAMI la durée moyenne de monitoring par type de moniteur (avec ou sans mémoire). Par contre, le nombre de patients sous monitoring au 1er janvier d'une année donnée ainsi qu'au 31 décembre de l'année qui précède doit l'être , ce qui semble ajouter peu de valeur. Dans les annexes de ce rapport, nous présentons une proposition de document de recueil destiné à optimaliser la collecte de données des centres agréés. Une option complémentaire de collecte de données auprès des 12 centres pourrait résider en un enregistrement séparé du monitoring d'enfants souffrant d'affections médicales spécifiques (par exemple, neuromusculaire). Ceci permettrait de séparer les données de monitoring pour une catégorie d'enfants qui souvent sont sous monitoring pour une période beaucoup plus longue que les autres catégories d'enfants.

Il manque dans la littérature scientifique des études de bonne qualité, particulièrement sur l'effet des incidents CR chez les nourrissons du point de vue du développement psychomoteur futur. Nous recommandons fortement la conduite de telles études.

Scientific summary

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LIST OF ABBREVIATIONS

5-HT	5-hydroxytryptamine
AHRQ	Agency for Healthcare Research and Quality (USA),
ALTE	Apparent Life Threatening Event
ANAES	Agence Nationale d'Accréditation et d'Evaluation en Santé(France)
APR-DRG	All Patient Refined Diagnosis Related Group
bpm	beats per minute
CCOHTA	Health Technology Assessment of the Canadian Coordinating Office
CDC	Centre for Disease Control (USA)
CEDIT	Comité d'Evaluation et de Diffusion des Innovations
CEP	Centre d'éducation du Patient
CHIME	Collaborative Home Monitoring Evaluation Group
CIA	Central Intelligence Agency
CM	Christelijke Mutualiteit
CMA	Canadian Medical Association
CNAMTS	Caisse Nationale d'Assurance Maladie des Travailleurs Salaries
CRD	Centre for Reviews and Dissemination
DARE	Database of Abstracts of Reviews of Effects
DRG	Diagnosis Related Group
DRG-DS	Diagnosis Related Group - Degree of Severity
ECG	electrocardiogram
EEG	electroencephalography
EMG	electromyography
EOG	electro-oculogram
ESPID	Document of the European Society for the Study and Prevention of Infant Death
GBPF	Groupement Belge des Pédiatres Francophones
HM	Home Monitoring
HSTAT	Health Services /Technology Assessment Texts (USA)
HTA	Health Technology Assessment
ICD-9	International Classification of Diseases (9th version)
ICES	Institute for Clinical Evaluative Sciences (Canada)
INAMI	Institut National d'Assurance Maladie Invalidité
INS	Institut National de Statistiques
IQ	Intelligence Quotient

K&G	Kind & Gezin
LCM	Landsbond Christelijke Mutualiteit
LOS	length of stay
MC	Mutualités Chrétiennes
MeSH	Medical Subjects headings
MFG	Minimale Financiële Gegevens
MKG	Minimale Klinische Gegevens
NeLH	National electronic Library for Health (UK)
NICE	National Institute for Clinical Excellence (UK)
NICU	Neonatal Intensive Care Unit
NIH	National Institutes of Health
NIS	Nationaal Instituut voor de Statistiek
NVSM	Nationaal Verbond van Socialistische Mutualiteiten
OLS	Ordinary Least Squares
ONE	Office de la Naissance et de l'Enfance
PG	Polygraphy
PMA	Postmenstrual age
PSG	Polysomnography
QTc	QT corrected
QT-interval	Measure of time between the start of the Q wave and the end of the T wave in the heart's electrical cycle.
RCM	Résumé Clinique Minimum
RIZIV	Rijksinstituut voor Ziekte- en Invaliditeitsverzekering
SES	Socio-Economic Status
SID	Sudden Infant Death
SIDS	Sudden Infant Death Syndrome
SIGN	Scottish Intercollegiate Guidelines Network (UK)
SpO2	Percutaneous oxygen saturation
SUD	Sudden Unexpected Death
TCT	Technische Cel / Cellule Technique
TV	Television
UK	United Kingdom
ULB	Université Libre de Bruxelles
US	United States
USA	United States of America
VVK	Vlaamse Vereniging voor Kindergeneeskunde

1 INTRODUCTION

1.1 SUDDEN INFANT DEATH SYNDROME: EPIDEMIOLOGY

1.1.1 Definition

Sudden Infant Death Syndrome (SIDS) is defined as the sudden death of an infant under 1 year old that is unexpected by history and unexplained after a thorough post-mortem examination, including a complete autopsy, investigation of the scene of death, and review of the medical history[1]. Despite declines of more than 50% in Canada, the United States, and many other countries (declines attributable in large part to educational campaigns advocating that infants be placed on their back to sleep), SIDS continues to be the leading cause of post-neonatal infant deaths (deaths between 28 days and one year of age), accounting for about 25% of deaths during that period[2, 3].

1.1.2 Incidence of SIDS and infant mortality

A 'diagnosis' of SIDS is unique in that the definition is reached by exclusion: by failing to show an 'adequate' cause of death. This is inevitably subjective and imprecise[4]. Figures are influenced by varying definitions of what constitutes a sufficient explanation of death[5], the proportion of death scenes investigated, the proportion of infant deaths autopsied etc. Time-trends analyses are also influenced by possible diagnostic transfers, or coding shifts. For instance in the United States, the overall post-neonatal mortality remained stable during the years 1999-2001 despite declining SIDS rates; in fact up to 90% in the decrease in SIDS rate could be attributable to changes in classification[6].

SIDS rates figures, international comparisons, and trends analyses therefore need to be interpreted with caution, and in parallel with trends in post-neonatal mortality. The SIDS rates in Canada and the US went down from 0.8 and 1.3 per 1000 live births in 1990 to 0.3 and 0.6 in 2002[2, 3]. The most recent data available for Belgium[i] date from 1997 (table 1). It should be stressed that under provision of law[ii] (March 2003), the unexplained death of a child below 18 months in Belgium will be followed by a mandatory autopsy unless at least one of the parents has an objection. This is likely to have a downward impact on the reported SIDS incidence.

[i] Separate data for Flanders are available up to 2004 quoting 25 SIDS cases (or 4 per 10.000 live births)[7].
[ii] External experts indicated this Legal provision hadn't come to its full effect yet (date of reference: Nov. 24th 2006).

Table 1. Infant mortality and Sudden Infant Death Syndrome in Belgium, 1993-1997

	1993	1994	1995	1996	1997
Total live births	122.210	117.020	116.122	116.442	116.213
Number of deaths					
Neonatal* (%)	504 (51%)	481 (53%)	398 (57%)	387 (63%)	414 (64%)
Post-neonatal* (%)	485 (49%)	422 (47%)	296 (43%)	228 (37%)	233 (36%)
Total (100%)	989	903	694	615	647
Mortality rates (per 1000 live births)					
Neonatal	4.1	4.1	3.4	3.3	3.6
Post neonatal	4.0	3.6	2.5	2.0	2.0
Total infant mortality	8.1	7.7	6.0	5.3	5.6
Sudden Infant Death Syndrome					
N deaths	247	209	139	99	100
Specific mortality rate (per 1000 live births)					
	2.0	1.8	1.2	0.9	0.9
Deaths during neonatal period/total				95/99	95/100
Post neonatal SIDS deaths / all post neonatal deaths				95/228 (42%)	95/233 (41%)

Source: Scientific Institute for Public Health. 1993-1995 data:[8]

From these figures it can be computed that over these 5 years, neo-natal mortality declined by 12%, post-neonatal mortality by 50%, and SIDS by 50%; 55% in the decrease in post-neonatal mortality is explained by the decrease in SIDS.

The following tables show international trends in the decrease in infant (0-1 year) and post-neonatal mortality (28 days-1 year). In relative terms, post-neonatal mortality has decreased more than neo-natal mortality, and this is at least partly explained by the decline in SIDS.

Figure 1. Infant mortality rate 1990-2003.

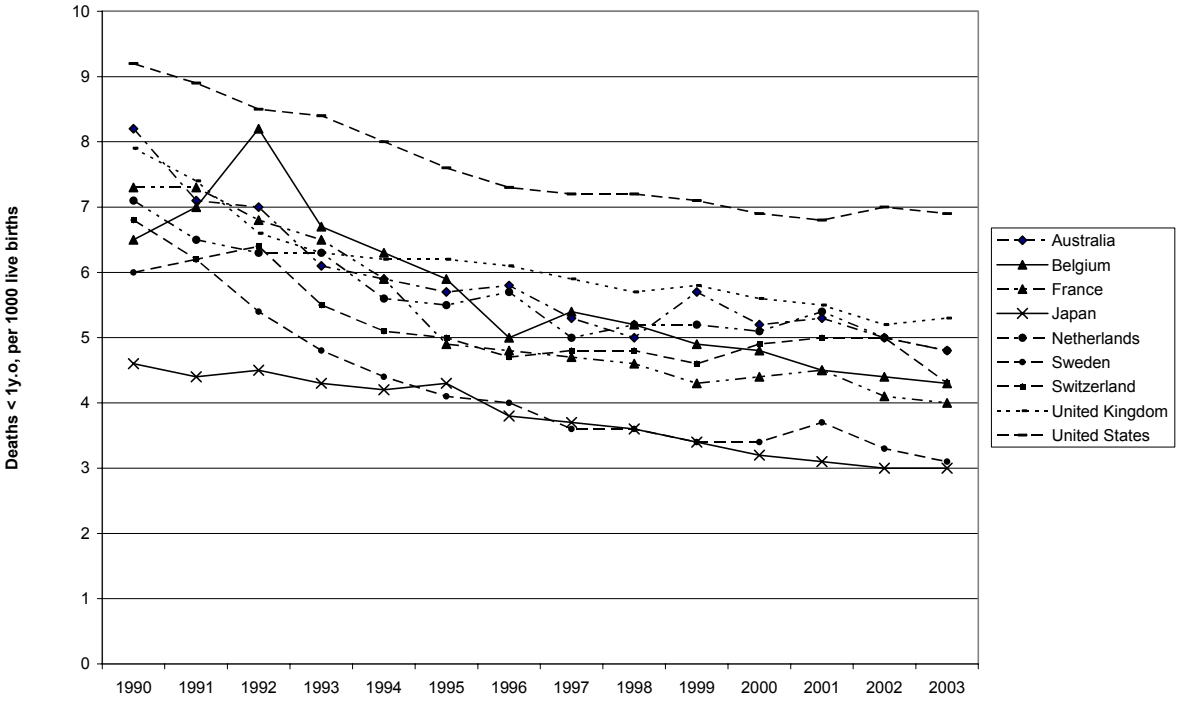


Figure 2. Post-neonatal infant mortality rate 1990-2003.

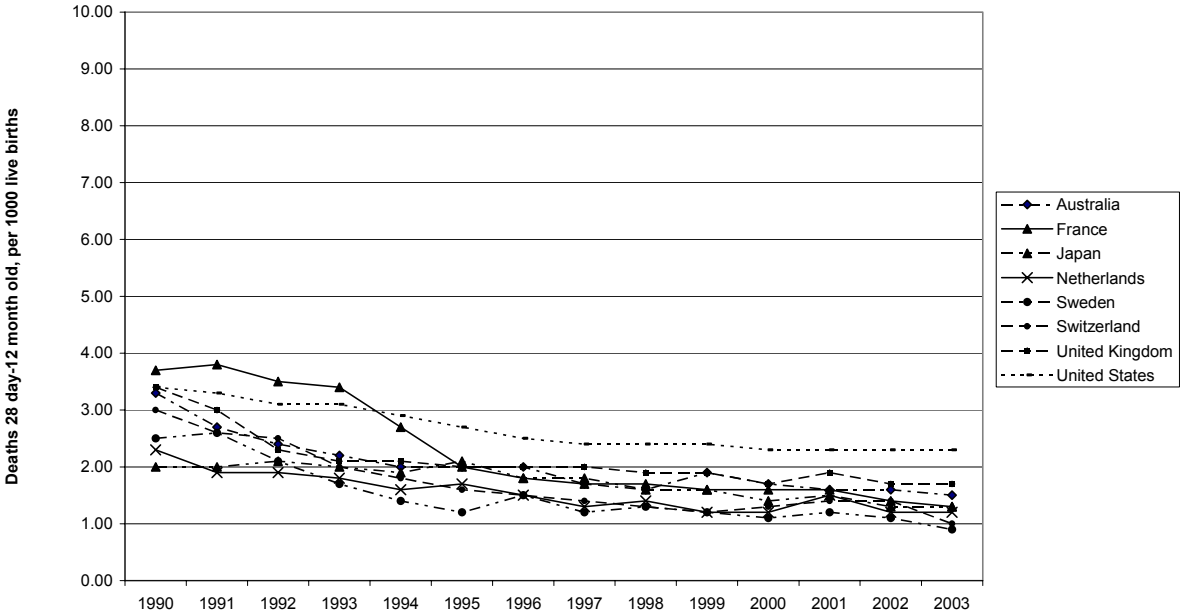
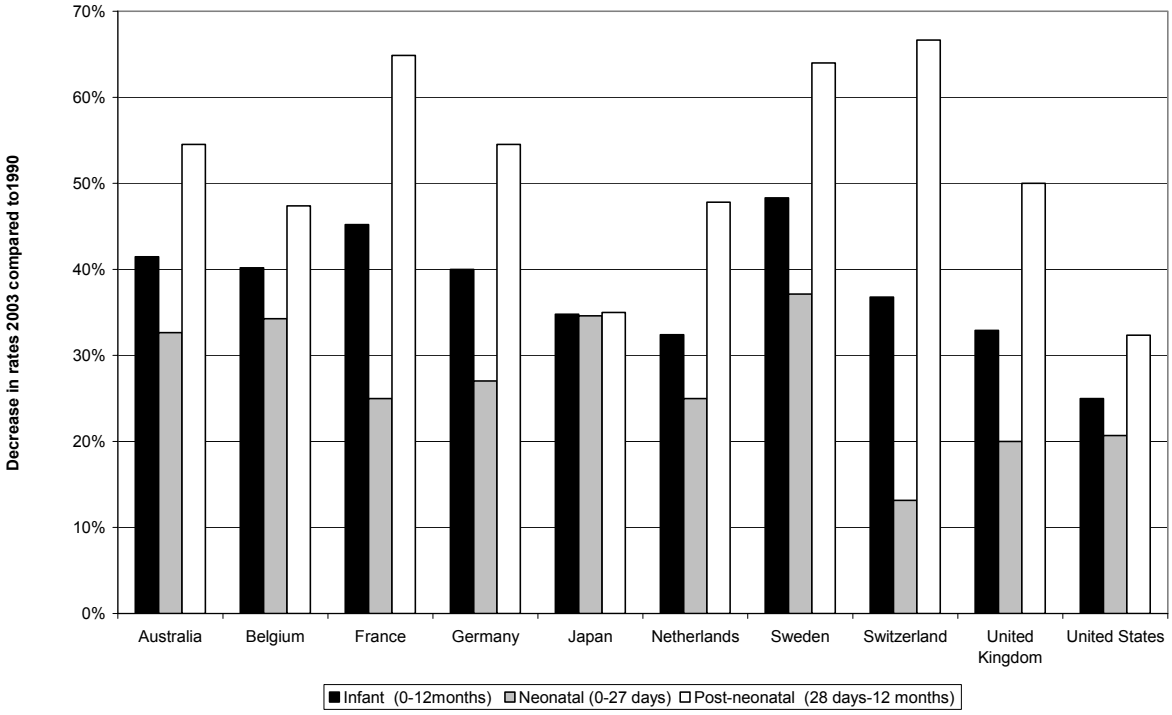


Figure 3. Changes in infant mortality rates 1990-2003.



Source for these 3 figures : OECD health data except for Belgium 1990 (National Institute of Statistics)

1.1.3 Risk factors FOR SIDS AND CLINICAL STRATEGIES

SIDS is more frequent among male infants; the age peak is around 2-4 months.

1.1.3.1 Causality

(adapted from[3])

The actual risk of SIDS in individual infants is determined by complex interactions, not yet fully understood, between genetic and environmental risk factors, some of them modifiable, some not.

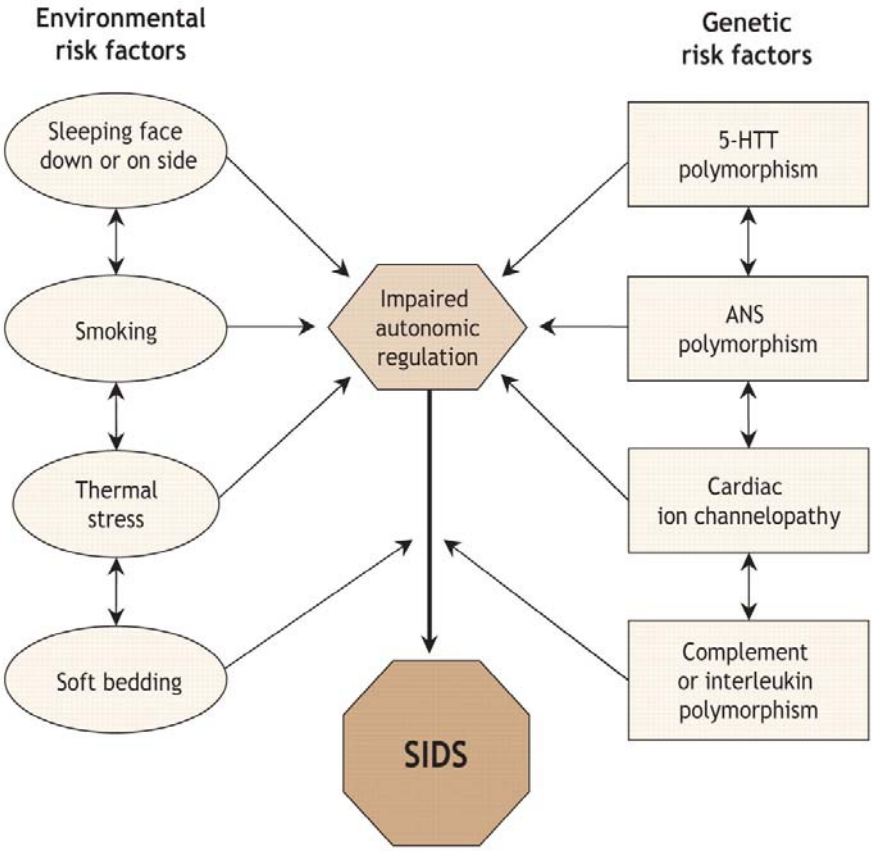
Up to 5 genes have been identified for which the distribution of polymorphisms differed between infants who died of SIDS and control infants; for instance sodium and potassium channels polymorphisms, or polymorphism of the serotonergic (5-hydroxytryptamine [5-HT]) transporter gene[9]. No specific clinical abnormality or phenotype has been delineated (except for cardiac ion channelopathies); however abnormalities one might expect from the identified polymorphisms are consistent with autopsy findings and are indicative of deficient brainstem autonomic neuroregulation. Other theories point to prolonged QT interval, metabolic disorders and/or apneas.

A deficit in arousal responsiveness maybe a prerequisite for SIDS to occur but may be insufficient to cause SIDS in the absence of other genetic, metabolic, morphologic or environmental risk factors.

There appears to be interactions between modifiable environmental risk factors such as prone body position, overheating, smoking environment and genetic risk

factors. For instance face-down sleeping does occasionally occur in prone-sleeping infants, and can result in episode of airway obstruction, but no-risk infants will arouse before such episodes become life-threatening, whereas infants with insufficient arousal responsiveness to asphyxia would be at risk for SIDS.

Figure 4. Schematic summary of potential interactions between environmental and genetic risk factors for sudden unexpected death in infancy and SIDS (reproduced from[3].



1.1.3.2 Clinical strategies

Clinical strategies can be classified into:

- strategies to reduce SIDS risk through modifiable (causal) risk factors and
- strategies aimed at identifying infants at higher risk for SIDS. Identifying high-risk infants can be the basis for targeted enhanced educational interventions to maximize adherence to recommendations for reducing the risk of SIDS[3].

Reducing the risk of SIDS

SIDS being a rare condition, the evidence to reduce risk comes from findings from observational studies, rather than randomized controlled trials. However epidemiological evidence and interventions based on this research (in particular

the promotion of prone sleeping position) have helped to reduce the evidence of SIDS. These recommendations are based on sound data, and absence of any evidence of harm.

The latest recommendations of the American Academy of Pediatrics to reduce the individual risk of SIDS include[6]:

- all infants should be placed on their back to sleep
- infants should sleep on their own crib (no bed sharing with the parents)
- infants should be placed on a firm mattress (no soft bedding)
- soft materials in the infant sleep environment should be avoided
- overheating and overbundling should be avoided
- mothers should not smoke during pregnancy and infants should not be exposed to second-hand smoke
- consider the use of pacifiers at bedtime, and naptime

Recommendations concerning the use of pacifiers (which could have a preventive effect) are still controversial[3].

Identifying infants at high risk for SIDS

No practical way currently exists to screen for any of the genetic polymorphisms and metabolic disorders in early infancy, but it is possible to identify infants at high risk for SIDS based on combinations of established risk factors[3].

Environmental factors found to be associated with an increased risk of SIDS are[3]:

- Maternal and antenatal risk factors:
  - low socio-economic status, low age, low level of education, single marital status, inadequate prenatal care
  - smoking
  - increased parity, short interval between pregnancies
  - intra-uterine hypoxia, fetal growth retardation
- Infant risk factors
  - prematurity
  - infants who had an apparent life-threatening event
  - siblings of prior SIDS infants
  - recent febrile illness

Key points

- **Despite considerable declines in incidence, SIDS still continues to be the leading cause of deaths between 28 days and one year of age.**
- **Due to its definition, by "default" of a clear cause of death, SIDS data need to be analyzed with due caution.**
- **The risk of SIDS in individual infants is determined by complex interactions, not yet fully understood, between genetic and environmental risk factors, some of them modifiable, some not.**
- **Clinical strategies in prevention of SIDS can target either the reduction of SIDS risk through modifiable (causal) risk factors or the identification of high risk infants.**

1.2 POLYSOMNOGRAPHY AND HOME MONITORING

1.2.1 polysomnography

1.2.1.1 Definition

By "polysomnography" (PSG) we refer to a centre-based patient monitoring process for the purpose of recording physiologic patient parameters during sleep. In keeps with general reimbursement criteria applying to Belgian residents (see also the appendix to this chapter) the specific definition of polysomnographic examinations in our report will cover polysomnographic examinations with a duration of minimally 6 hours, including protocol and print-outs of polysomnographic readings regarding the continuous and simultaneous recording for at least:

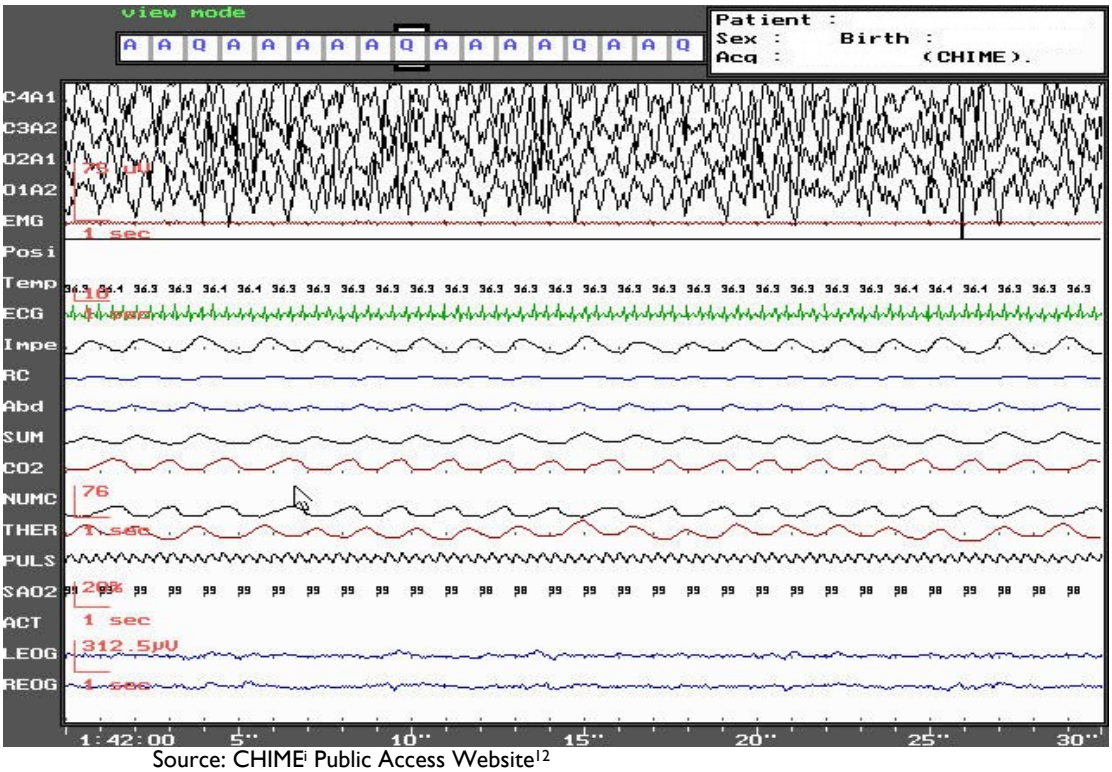
- one electroencephalography for assessing the global neural activity (E.E.G),
- one electro-oculogram for evaluating eye movements (E.O.G.),
- one electrocardiogram for registering heart rhythm (E.C.G.).
- continuous oximetry and 2 further parameters for gauging respiration (e.g. nasal flow, thoracal, abdominal efforts, etc.)

As indicated above, this enumeration is not limitative and additional physiologic parameters can be included in the polysomnographic examination such as the measurement of oesophageal pH for assessing regurgitation or an electromyography for gauging limb movements (E.M.G.)[10].

1.2.1.2 Technology

A full polysomnographic set-up will include sensor leads (electrodes, piezo crystal effort sensors, pulse oximeters, etc.) feeding physiologic measurements into a computer[11]. The resulting output is a series of waveform tracings, allowing for further analysis of observations, usually following a pre-defined fixed time interval called "epoch" (see figure 5).

Figure 5. 30 second epoch of Alice3 Polysomnogram with no cardiorespiratory events



Source: CHIME[i] Public Access Website[12]

Automated data analysis of polysomnographic readings is available through various software applications. To date, most computerized analytic methods aspire to emulate visual inspection by a clinical expert. As such, they mostly rely on rule-based logics referring to preset physiologic threshold values[13].

1.2.2 Home monitoring

1.2.2.1 Definition

By "home monitoring device" we refer to devices equipped to monitor (at least) both cardiologic (bradycardia's) and respiratory events (central apnea's) in the patient's home environment. In agreement with reimbursement criteria applying to Belgian residents (see appendix 2) these monitors can be equipped either with or without an event recording function.

1.2.2.2 Technology

Monitor devices qualifying for reimbursement minimally enable follow-up of heart rate and respiration, often also allowing for pulse oximetry[ii] monitoring. As is also the case for inpatient polysomnographic examinations, electrodes are attached to the patient's body and feed electronic readings through lead-cables into the monitoring device.

Alarm settings can be altered to allow for various threshold values with regard to heart rate, respiration and oxygen saturation. Physiologic alarms (presumably

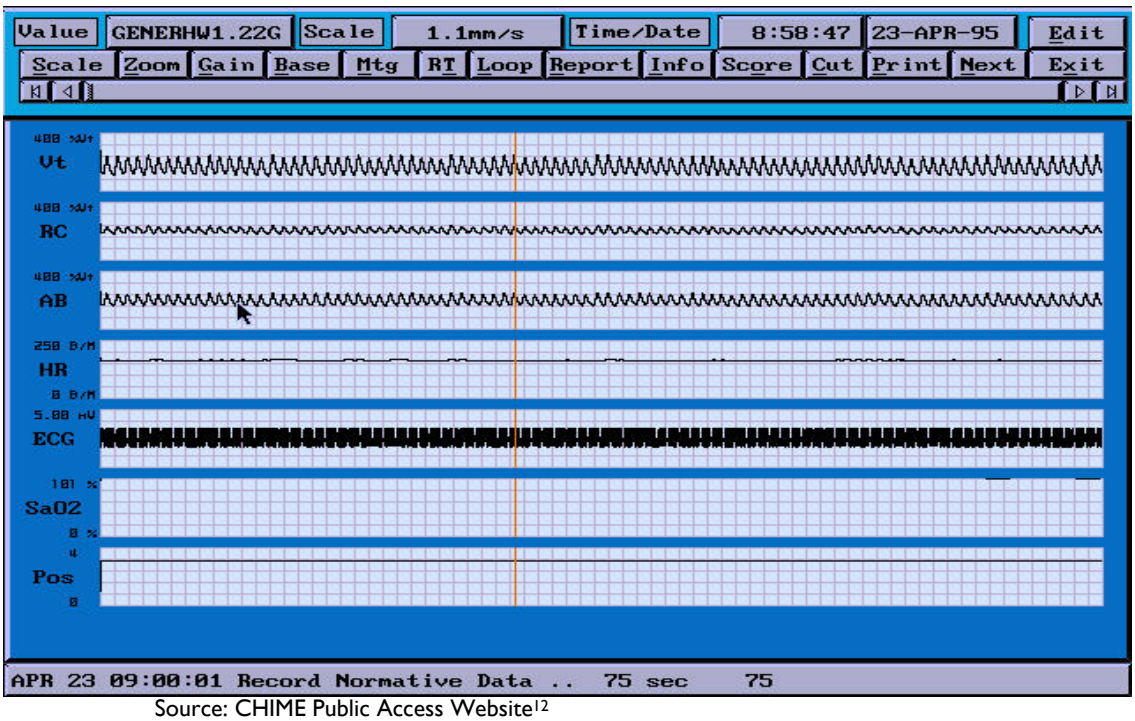
[i] Collaborative Home Monitoring Evaluation Group (CHIME)

[ii] External Belgian experts indicated this feature is not used in everyday practice as oximetry probes do not tend to remain attached to an infant's finger (or ear lobe) due to frequent nocturnal body movements.

stemming from anomalous physiologic readings) involve both audible and visual indicators. Some devices have the capacity of setting off equipment alarms due to technical malfunctions. More sophisticated devices include an event-recording memory, storing data in wavelet form (see figure 6) on patient events and alarms for further inspection by clinical experts.

Moreover, compliance information with respect to the practical use of devices by patients (e.g. average usage time per night, etc.) can be stored away to the internal memory.

Figure 6: Routinely collected 3 minute non-event record



Source: CHIME Public Access Website[12]

1.2.3 Belgian Context

1.2.3.1 Standard settings applying to Belgium

In accordance with Belgian reimbursement criteria applying to patients qualifying for home monitoring follow-up based on preceding polysomnographic readings, unless indicated otherwise, we will define a anomalous (or positive) reading (applying to inpatient polysomnographies) as a reading indicating:

- either one or several central apnea's lasting over more than 20 seconds, accompanied by desaturation (Sa_{O2} < 88%) or a bradycardia below 60 bpm,
- either indicating more than three obstructive apnea's, each lasting over more than 3 seconds.

1.2.3.2 Manufacturers/Distributors

Six Belgian manufacturers and distributors of polsyomnographic and home monitoring devices were contacted with the request to transmit information with regard to (unpublished) research, technical product specifications and market data. One distributor replied favourably by returning general product

information and price information. Confidentiality, however, was requested with regard to the latter information.

Through further contacts we identified three companies as principal players in the market for sleep monitoring devices:

- Medatec: producing polysomnographic equipment (Morpheus®) and home monitoring devices (Pamela®) for the Belgian (and foreign) market: reported to account for around 80% of the Belgian market.
- Omega Hospital: distributing polysomnographic devices (Alice® series) for foreign producer Respironics, servicing about 10% of the Belgian market .
- Air Liquide: acting as a distributor for the foreign produced Embla® polysomnographic devices (previously Medcare®) and holding a 10% stake in the Belgian market.

The Alice® and Ebmla® devices are regarded as the two world leading brands in polysomnographic devices, whereas Medatec should be considered more as a local player.

Key points

- **By "polysomnography" (PSG) we refer to centre-based patient monitoring of physiologic parameters during sleep.**
- **By "home monitoring devices" we refer to devices equipped to monitor (at least) both cardiologic (bradycardia's) and respiratory events (central apnea's) in the patient's home environment.**
- **We identified three prominent players on the Belgian market: Medatec, Omega Hospital and Air Liquide.**

2 GENERAL METHODOLOGY

This research follows a Health Technology Assessment (HTA) approach. The purpose is to support the process of decision making in health care at policy level by providing reliable information. A HTA collects and analyzes evidence in a systematic and reproducible way (and organizes it in an accessible and usable way for the decision makers). The principles of gathering, analyzing and using information are identical to the principles of Evidence Based Medicine (EBM) and Clinical Practice guidelines (GCP), but the purpose is different. EBM and GCP aim to support decision making at individual clinical or patient group level. In contrast, HTA aims to support decision making at policy level, leading to a different kind of recommendations and answers.

2.1 OBJECTIVES AND METHODS

The objectives of this health technology assessment were:

(1) To assess the value of overnight polysomnographies and home monitoring in under-one infants, particularly in prevention of SIDS. This was done through a critical appraisal of the clinical and economic scientific literature on the subject.

(2) To study the utilisation of PSG and home monitoring for under-one infants in Belgium and their cost to the social security. We analysed relevant data in available databases (hospital stays, billing codes, health insurer data); and conducted an exploratory qualitative study among health professionals and parents of infants undergoing a PSG.

2.2 FURTHER STRUCTURE OF THE REPORT

The next chapters are structured as follows:

- Chapter III contains a critical appraisal of the clinical scientific literature.
- Chapter IV contains a critical appraisal of the economic scientific literature.
- Chapter V offers a quantitative description of the current Belgian situation.
- Chapter VI describes the content, evolution and impact of various SIDS prevention campaigns.
- Chapter VII offer a qualitative exploration of issues at stake among the principal Belgian stake holders.
- Chapter VIII contains a general discussion.
- Chapter IX contains policy recommendations.
- Chapter X summarizes the comments made by the scientific validation team and describes how these have been accounted for in the report.

3 CLINICAL EFFECTIVENESS.

Following the report's scope as set out in the introductory chapters, our clinical literature search will focus on overnight polysomnographies for inpatients below 1 year as well as the use of (cardio-respiratory) home monitoring devices for this population.

3.1 CURRENT USE OF POLYSOMNOGRAPHIES AND HOME MONITORING IN THE INFANT POPULATION IN WESTERN COUNTRIES

3.1.1.1 Apnea and SIDS

The use of a polysomnography (PSG) as a screening tool to recognize those infants at increased risk of dying by SIDS, goes back to the publication of Steinschneider in 1972[14]. He postulated a role for apnea as pathophysiologic precursor to SIDS, based on his observation of apnea on hospital cardiorespiratory monitoring in 2 infants, both siblings of 3 other children that had died of SIDS. Both siblings subsequently died unexpectedly. In 1975, Stein et al. described the pediatric pneumogram as a new, simple method for detecting apnea in infants, e.g. at home[15]. More than two decades later, however, evidence of infanticide for all 5 infants of the family described by Steinschneider became known ("The Hoyt Case"[16]). Nevertheless, the suggestion of a possible relationship between infantile apnea and SIDS had fostered widespread use of laboratory polysomnographies and home monitoring. Since also premature infants, even if healthy, are predisposed to apnea, the so-called "apnea of prematurity", they were included in the group to be evaluated and eventually monitored at home. Unfortunately, the use of the PSG and home monitoring as a "standard practice" was established before its efficacy was proven.

3.1.1.2 ALTE (Apparent Life Threatening Event)

Apart from infants being screened or monitored within the scope of SIDS-prevention, PSG and home monitoring is also often used in infants that experienced an Apparent Life Threatening Event (ALTE). These infants previously were categorized as "Near-Missed-SIDS", but this terminology has been abandoned since it is not clear at all how exactly to define the relationship between ALTE and SIDS (see further). The definition of ALTE still mostly used[17, 18, 19]is the definition by the National Institutes of Health Consensus Development Conference on Infantile Apnea and Home Monitoring[20]: "an episode that is frightening to the observer and is characterized by some combination of apnea (central or occasionally obstructive), color change (usually cyanotic or pallid but occasionally erythematous or plethoric), marked change in muscular tone (usually marked limpness), choking, or gagging". As for SIDS, this definition makes broad or imprecise interpretations possible. Some authors prefer not to include choking or gagging since this might point to gastro-intestinal or upper airway problems[21]. Others have suggested limiting the term to infants who required vigorous stimulation for resuscitation or to patients without obvious abnormalities on physical examination[22, 23]. Sometimes infants with altered mental status are explicitly included in the definition of ALTE[24]. Up to 23%-50% of ALTE remain unexplained after thorough investigation and are classified as "idiopathic"[17, 25]. The severity of ALTE relies only on the history provided by the witness and on the first medical examination after the event, which can already turn out completely normal.

The relationship between ALTE and SIDS is not straightforward; most likely, ALTE and SIDS can be considered to be 2 separate entities (see further). In order to establish a diagnosis or to prevent new life-threatening events, but also due to the frightening character of ALTE, PSG and/or home monitoring is frequently used in these children.

3.1.1.3 Other medical conditions

A third group of infants or children for whom PSG as well as home monitoring are used in an attempt to prevent sudden death, are those children considered at risk because of a certain medical condition, e.g. infants with neuromuscular disorders, bronchopulmonary dysplasia, facial dysmorphia like Pierre-Robin syndrome etc. Also infants of drug dependent mothers sometimes are considered as being at an increased risk of SIDS.

This chapter aims at describing the scientific background and proven evidence for the use of PSG and home monitoring in infants below one year of age, based on a systematic review of the literature. The three groups described above (SIDS, ALTE, other medical conditions) will be considered separately.

3.2 METHODS

A literature search for the available evidence was undertaken between April and May 2006, searching the following databases and websites:

- Cochrane Reviews database, DARE, ACP Journal Club, Centre for Reviews and Dissemination (CRD) databases (University of York, UK),
- National Guideline Clearing House (USA), NeLH Guidelines Finder (UK), ANAES (France), CMA Infobase (Canada), CDC Recommends (Centre for Disease Control, USA), Health Services /Technology Assessment Texts (HSTAT, USA), ICES (Institute for Clinical Evaluative Sciences, Canada), New Zealand Guidelines Group, Scottish Intercollegiate Guidelines Network (SIGN - UK), National Institute for Clinical Excellence (NICE - UK), Agency for Healthcare Research and Quality (AHRQ - USA),
- Medline through PubMed
- Embase (this database was searched systematically until Januari 2004 in the Health Technology Assessment of the Canadian Coordinating Office (CCOHTA) "Review of Guidelines for Referral of Patients to Sleep Laboratories". In this study, an additional search from Januari 2004 onwards was performed.)

The search strategy, developed by two researchers, is described in detail in the appendix of this chapter (database, Mesh and/or "free terms") in order to be easily replicated for further validation (Appendix to chapter 3, see "Search Strategy description and number of results": 2.1 databases for Guidelines, 2.2 databases for Systematic Reviews, 2.3 Medline, 2.4 Embase). The literature search was performed by one researcher.

From the retrieved publications, articles in English, French or German published between 1981-2006 (25 years) were kept. Since the first report on the possible link between SIDS and pneumocardiography was only published in 1972 by Steinschneider, and since currently available technology is very different compared to the early pneumocardiographs, this was considered to be

appropriate. The search was completed by additional reference tracking. Reference selection was performed by one researcher on the basis of titles and abstracts. Studies concerning diagnostic value of polysomnography (PSG) or diagnostic/preventive value of home monitoring in Sudden Infant Death syndrome (SIDS), Apparent Life Threatening Event (ALTE) or other infants (below 1 year) were retained. Studies concerning technical efficacy were also included. Excluded were articles concerning preventive conservative measures for SIDS since this was not the scope of the review. Also articles describing mere hypotheses (e.g. "High mean apnea duration was correlated with low substance P level in the first months of age in SIDS risk infants selected anamnestically. This may reflect a delayed maturation of respiratory control mechanisms."[26]) were not taken into account. Finally, editorials or narrative reviews, case-reports and publications focusing on normal values for age were excluded. Especially for the search in PubMed and Embase, an older article recently followed by an article of much higher quality was not retained. On the whole, 2052 references were retained by the developed search strategy and were read by title/abstract. After reading of title/abstract, 68 references were kept, which were used for the efficacy and clinical effectiveness study of polysomnography and home monitoring in infants (chapter 3.5 to 3.9).

The quality of observational studies, cohort studies or case-control studies was evaluated using the appropriate form of the Dutch Cochrane Collaboration[27].

Based on this appraisal, a level of evidence (good, moderate or weak) was assigned to each of the included studies. The evidence of case-series was considered as being weak. The appraisal of each of the included references is indicated in the evidence tables (Appendix to chapter 3: see 2.5 Guidelines and Systematic Reviews-Analysis of Results, and 2.6 Medline and Embase-Analysis of Results).

Finally, the value of polysomnography (PSG) as a means of detecting infants at increased risk of SIDS was evaluated according to the model of "Hierarchy of diagnostic efficacy", as described in 1991 by Fryback and Thornbury[28]. In this model, 6 levels are discerned in the evaluation of efficacy of diagnostic tests: technical efficacy, diagnostic accuracy, diagnostic thinking, therapeutic impact, patient outcome and cost-effectiveness analysis. If a test performs poorly at one level, it is unlikely to perform well at a further level. Also for home monitoring, the levels of efficacy according to Fryback and Thornbury were applied; which might seem contradictory since this is no "diagnostic test". However, the "diagnostic process" of recognition of cardiorespiratory events is the first step in the use of home-monitoring (before an alarm can be given), and therefore it was considered appropriate to use the above mentioned "Hierarchy of diagnostic efficacy".

3.3 RESULTS (I): MAIN PHYSIOLOGICAL PARAMETERS CONSIDERED IN THE LITERATURE

3.3.1 Apnea

Apnea or a cessation of respiratory airflow, is analyzed by type (central, obstructive, mixed, periodic breathing), duration (short, prolonged) and frequency[29]. It is considered to be central if there is cessation of both airflow and inspiratory effort. An "obstructive apnea" is defined as cessation of airflow at the nose and the mouth with concomitant increased inspiratory effort. A "mixed apnea" is defined as a combination of central and obstructive apnea within a single episode of airflow cessation. Periodic breathing, which is also

sometimes considered in young infants, is usually defined as three respiratory pauses of at least 3 seconds with intervening periods of respiration lasting less than 20 seconds[30, 31], but other definitions are also used.

Especially the duration of an apnea is important when considering its pathological nature. In agreement with the American Academy of Pediatrics Task Force on prolonged apnea in 1978[32] , a short central apnea is defined as an apnea lasting from 2 to 10 seconds in duration which can be normal at all ages, whereas a prolonged apnea is determined by most investigators as a cessation of breathing for 15 to 20 seconds or longer. Briefer episodes associated with bradycardia, cyanosis, or pallor may be considered to be significant. The basis for this definition was the finding of Hoppenbrouwers et al[33] in 1980, that an apnea of 10-20 sec is common among healthy infants whereas pauses of 20 sec or longer are very rare. The Policy Statement in 2003 of the American Academy of Pediatrics on Apnea, SIDS and Home Monitoring[20] also uses this definition: "Apnea of infancy is defined as an unexplained episode of cessation of breathing for 20 seconds or longer, or a shorter respiratory pause associated with bradycardia, cyanosis, pallor, and/or marked hypotonia. The term "apnea of infancy" generally refers to infants with gestational age of 37 weeks or more at the onset of apnea. Apnea of prematurity is defined as sudden cessation of breathing that lasts for at least 20 seconds or is accompanied by bradycardia or oxygen desaturation (cyanosis) in an infant younger than 37 weeks' gestational age. It usually ceases by 37 weeks' postmenstrual age but may persist for several weeks beyond term, especially in infants born before 28 weeks' gestation. The most recent data indicate that extreme episodes usually cease at approximately 43 weeks' postconceptional age."[20]

Although most authors adhere to the above mentioned definition of pathological apnea, several articles were found in which other definitions were used (see Evidence Tables in Appendix of this chapter).

It should be noted that some authors focus especially on obstructive apnea's when studying the problem of SIDS. These obstructive apnea's can only be detected when evaluated in the sleep laboratory since the currently used home monitors for infants register chest or abdominal movements as well as heart beats but don't allow registration of the nasal and oral airflow. (For more information on the registration of chest and abdominal movements in infants, see De Groote et al. in 2002[34]). For obstructive apnea's, these authors consider a duration of more than 2 seconds as being abnormal[35, 36, 37]. This value is based on 1) the observation by Kahn et al[36] that in 75 infants obstructive apnea's induce a larger decline of transcutaneous oxygen saturation as compared to central apnea's of the same duration and 2) the observation by the same author that in 110 normal infants, the 95th percentile for central apnea duration lies on 15 seconds whereas only 5% of obstructive apnea's in this study group lasted longer than 2 seconds[36, 37]. To our knowledge, no data from other authors are available concerning transcutaneous oxygen saturation during obstructive apnea's. Only one study reports on obstructive apnea's during home monitoring[38]. The authors studied normal infants and preterms, ALTE, SIDS-siblings by means of inductance plethysmography (a method not commonly used in home monitoring, allowing to detect not only central but also obstructive apnea's); is was noted that among apnea's of more than 30 (thirty) seconds, 70% included at least 3 obstructed breaths. The authors point to the frequency of obstructive breaths of long duration, without commenting on significance of these events (see further).

3.3.2 Bradycardia

In young infants, heart rate and heart rate variability vary according to age. Therefore, bradycardia should be defined based on the age of the infant and the duration of the bradycardia. Nevertheless, even among authors defining bradycardia's in relationship to age of the infant, the cut-off values for certain ages vary[39, 29, 40]. From many studies it is clear that not only apnea, but also bradycardia with or without apnea can occur in healthy infants, preterms or ALTE and as a consequence often the terminology "cardiorespiratory events" is used. Whereas in the seventies the main focus was on apnea as cause of the SIDS or of unexpected life threatening events, it is now clear that bradycardia as well can be the single and/or first sign[41, 42, 38].

3.3.3 Desaturation

Episodes of oxyhaemoglobin desaturation are familiar in preterm infants and can also be demonstrated in healthy term infants.

Based on studies of large groups of normal infants, a sudden decrease of oxygen saturation < or = 80% for > or = 4s; or a baseline baseline SpO2 below 90% is mostly considered to be abnormal[43-45].

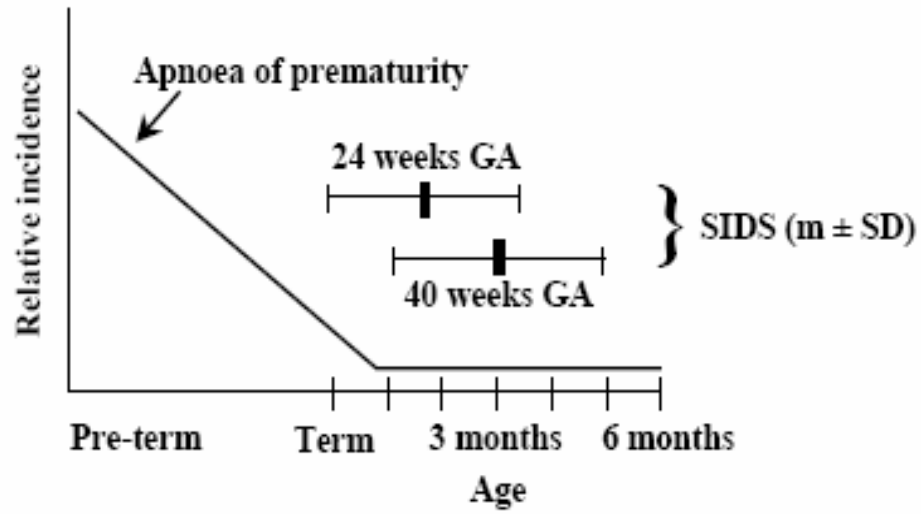
3.4 RESULTS (II): EPIDEMIOLOGICAL DATA OF INFANT GROUPS CONSIDERED TO BE AT INCREASED RISK FOR SIDS

Epidemiological studies indeed revealed an increased risk of SIDS for certain groups of infants.

3.4.1 Prematures/dysmatures

Infants of short gestation or low birthweight are at an increased risk for SIDS (as well as they are at an increased risk of sudden but explained deaths). To some degree, an overlap exists between the epidemiological factors predisposing to SIDS and to preterm delivery or low birthweight (low socioeconomic class, maternal smoking, etc.). The SIDS risk is estimated to be 3 times higher compared to normal control infants for babies born at a gestational age of 24-28 weeks, and declines thereafter gradually with increasing pregnancy duration. Also small-for-gestational age or "dysmatures" are at an increased risk, as well as infants from multiple births (twins, triplets, etc.[46-49]. While the Back-to-Sleep campaign induced a significant decrease in the incidence of SIDS, the relative risk for preterms/dysmatures seems to increase over time. As a consequence, their relative proportion in the total group of SIDS-deaths increases[48, 50]. It should be noted that the peak incidence of SIDS in preterms, as expressed in weeks of post-term age, comes a few weeks earlier than the peak incidence in full term babies[51, 52].

Figure 7. Relationship between the timing of peak incidence of SIDS (sudden infant death syndrome) to the (relative) incidence of apnoea in infants with apnoea of prematurity. GA, gestational age at birth; m, mean; SD, standard deviation.



Source: Baird TM, Semin Neonatol 2004;9(3):205-11.

Conclusion: there is a proven increased risk for SIDS in preterms/dysmatures. This risk increases with decreasing pregnancy duration. It should be noted that there exists a certain overlap between the epidemiological factors predisposing to SIDS and to preterm delivery or low birthweight.

3.4.2 SIDS siblings

After a critical review, Bentele[53] retains a relative risk for SIDS among brothers and sisters of SIDS-victims ("SIDS siblings") of 3.6 to 9.5. However, the quality of the included studies is questionable (use of questionnaire, no autopsy…). Hunt[54] finds the same range in additional studies of better quality (large population-based studies) and notes that this number corresponds to the recurrence risk of infant mortality in non-SIDS cases and in surviving twins after SIDS in the first twin-member (thus questioning the role of genetic factors). The main criticism of these epidemiological mortality studies is that they don't take into account epidemiological factors associated with SIDS, with one exception[46]. Many of the factors underlying SIDS are predicting factors for infants dying suddenly of expected causes as well, except for the age distribution at death, the role of maternal prenatal smoking which is more important in SIDS, and the frequency of congenital abnormalities in (by definition) non-SIDS infant deaths[46]. This makes clear that there remains a place to consider SIDS as a distinct entity. Hunt CE concludes that probably a complex interaction between genetic and environmental factors determines the risk to develop SIDS.

Conclusion: there is a proven increased relative risk for SIDS in SIDS siblings, but this increase is comparable to the recurrence risk of infant mortality in non-SIDS cases. Many socio-economic and environmental factors predisposing to SIDS are the same for general infant mortality. Nevertheless, there are arguments that there remains a place to consider SIDS as a distinct entity, that is probably caused by a complex interplay between genetic and environmental factors.

3.4.3 ALTE

In a systematic review on ALTE by McGovern[25] (8 studies, 643 infants) the incidence of ALTE has been described as 0.6-0.8% of all emergency visits of infants under 1 year, and 0.6/1000 live births; recurrences varied between 0 and 24%. Death rate in the ALTE group was 8/1000, but some of these infants suffered from congenital metabolic disorders. Oren[55] defined a subgroup of infants more likely to die: 13% of infants with ALTE at night and terminated only after mouth to mouth resuscitation; however, the quality of this study is low. Kiechl-Kohlendorfer[56] found in a prospective population-based study a higher incidence for ALTE: 2.46/1000 live births (identified on the basis of hospital admission) of whom half are idiopathic; whereas a New Zealand study even reported an annual hospital admission rate for ALTE of 9.4/1000 live births[57]. The subjective nature of the definition might be an explanation for this large variance in incidence. It should be noted that in the large prospective study of Ramanathan[38], also in 43% of healthy term infants at least one apnea of 20 seconds or more during a 3 month period was registered. 23% to 50% of ALTE remain unexplained after thorough investigation and are classified as "idiopathic"[17, 25].

Concerning the relationship between SIDS and ALTE, Bentele[53] gives a survey of the available literature at that time. He finds an increased risk for SIDS after ALTE, but points to the difficulties in the retrieved studies: small groups, confounders like epilepsy considered as ALTE etc. Hoffman[58] in a large population based study, including 10% of all USA live births during the study period and 757 SIDS victims, found a higher frequency of maternal reported ALTE (6%) in SIDS-victims compared to control infants. Likewise, Platt et al[47] in a large population-based study including 363 SIDS, noted a significant higher incidence of ALTE in SIDS-victims as compared to normal controls (OR 2.55 (1.02-6.41); however, the same was true for sudden explained infants deaths as well. According to the Consensus Document of the European Society for the Study and Prevention of Infant Death (ESPID) in 2004, and based on a literature review including 3 other large population based studies, a relationship between ALTE and SIDS could only involve a fraction of all SIDS cases since less than 10% of future SIDS victims had presented an ALTE at some time before death[17].

In the literature review of Kahn[17], infants with an ALTE were a few weeks younger than SIDS victims and enjoyed more favorable circumstances at the time of the event, such as supine sleeping or being found during day time; and the incidence remained the same after the "Back-to-Sleep" campaign. These characteristics separate ALTE from SIDS, and make it acceptable to consider them as two separate entities.

In conclusion: due to the subjective nature of the definition of ALTE, the incidence range is wide. Mortality incidence of ALTE is only reported in few studies. There is evidence that SIDS victims before death more often present with ALTE compared to normal controls, although this is true only for less than 10% of SIDS victims. Most likely, ALTE and SIDS can be considered to be 2 separate entities.

3.4.4 Other medical conditions at increased risk for SIDS

Among the infants considered to be at increased risk for SIDS secondary not to certain environmental or socioeconomic factors but secondary to medical conditions, infants of drug dependent mothers are sometimes taken into account as well. The literature on incidence of SIDS in this group is described here. Bentele[53] reviews available studies on SIDS among infants from drug

dependent mothers: in studies on a population of at least 100 infants, the risk amounted from 14 till 25/1000 infants. This figure is high, but many risk factors concur in this group of infants (low socioeconomic class, young unmarried mothers, smoking,etc.). In these studies no information on the performance of autopsy is available and hence their quality can be discussed. A later meta-analysis Fares[59] found a lower SIDS-incidence for cocaine- or polydrug-use during pregnancy (5.2/1000; 95% confidence interval: 4 to 7/1000), a number equalled (5-13/1000) by the population-based study of Ward[60]. Another case-control study in a tertiary-referral centre found only a significant increase in SIDS among babies exposed to polydrugs during prenatal life if they were low-birth weight (below 2500 grams)[61].

Key points

- **Infants born at 24-28 weeks of pregnancy have a 3 times higher risk for SIDS; the risk decreases with increasing pregnancy duration.**
- **Brothers and sisters ("siblings") of SIDS-victims are at an increased risk for SIDS, probably due to a complex interplay of genetic and environmental factors.**
- **Due to the subjective nature of the definition of ALTE ("Apparent Life Threatening Event"), the incidence range is wide. There is evidence that SIDS victims before death more often present with ALTE, although this is true only for less than 10% of SIDS victims. Most likely, ALTE and SIDS can be considered to be 2 separate entities.**
- **Infants of drug dependent mothers are at increased risk for SIDS.**

3.5 RESULTS (III): TECHNICAL EQUIPMENT EFFICACY: LITERATURE REVIEW

3.5.1 Technical Equipment

Detailed evidence tables, including the retrieved literature on technical efficacy, are included in the appendix to this chapter. For further background we also refer to the first chapter of our report.

3.5.1.1 Polysomnography

Whole night polygraphic sleep studies are conducted to assess respiratory, cardiac or neurological abnormalities during sleep. EEG, EOG and chin (submental) EMG recordings contribute to the scoring of sleep or wake state; thoracic and abdominal movement recordings as well as nasal and oral airflow detection are used for the analysis of breathing and apnea characteristics; heart rate is recorded from ECG electrodes; oxygen sensors inform on oxygen saturation values during sleep and following apnea; gross body movements are scored by actigrams or surface EMG, as well as by artifacts on the other leads. Although at first instance aimed to describe conditions to perform PSG in older children (and not in infants), the American Thoracic Society[62] in 1996 describes consensus recommendations on the registration techniques to be used.

3.5.1.2 Home Monitoring

Home monitoring can be conducted by pneumographic or by cardiorespiratory monitoring without or with event-recording (the so called "memory" monitors).

In 1987, the NIH Consensus Conference on Infantile Apnea and Home Monitoring[31] recommended cardiorespiratory monitoring rather than pneumography-based monitoring ("apnea monitoring"), because it had been demonstrated in several reports that not only apnea but also bradycardia could be the first signal of an unexpected infant death. The latter has been confirmed in many studies since then.

Some literature is available on the advantages of event-recording (see further).

3.5.2 Literature: Technical Efficacy

3.5.2.1 Polysomnography

Bentele[53] mentioned the lack of standardization of pneumocardiographic techniques. The Technology Report of CCOHTA (Canadian Coordinating Office for Health Technology Assessment) in 2005[63] also points to the fact that PSG for application in children has been poorly standardized in its performance and interpretation. Some publications study the validity of PSG interpretation. Hunt in 1988 demonstrated that the definition of apnea-onset affects the result of the pneumogram (low quality study); Crowell[64] in two well conducted studies demonstrated that inter- and intra-rater reliability between PSG evaluators is only moderate although it can be improved by training.

3.5.2.2 Home Monitoring

The efficacy of home monitoring devices has been discussed already in 1986 by the NIH Consensus Conference on Infantile Apnea and Home Monitoring but their recommendations were not accompanied by references. The Conference recognized that transthoracic electrical impedance registration of breathing movements, the mostly used technical approach, was susceptible to artifacts and did not register obstructive but only central apneas. It was recommended to develop a set of minimal standards for infant home monitors, and also to discourage the marketing of "over the counter" monitors since these devices don't guarantee any quality and moreover its use is not supervised by professionals as should be the rule. The effectiveness of home monitoring, according to this Conference, had never been addressed by scientifically designed studies, nor for the prevention of SIDS, nor for ALTE, nor in SIDS siblings; but the difficulty to conduct such studies was recognized. It was noted that the incidence of SIDS had not significantly declined since the introduction of home monitors.

The superiority of inductance plethysmography over transthoracic impedance registration of breathing movements has been confirmed in later well-conducted studies[38]. Nevertheless, most currently available monitors still use impedance registration and don't register obstructive apnea's.

The CEDIT-study[65] (moderate quality) in 1999 compared polysomnography and monitoring (impedance) in preterms born before 34 weeks postmenstrual age, and found that the average apnea frequency was twice as high on polysomnography compared to monitoring; both modalities showed two thirds of false alarms (movement artifacts…). The same was reported by de Nardi[66] (moderate quality) and by Rowland[67] (weak quality). By use of the same

equipment in the hospital and at home, Keens[68] found no difference between the different environments of recording (weak quality).

Weese-Mayer[69] et al examined a group of infants and toddlers (interquartile range of 2-18 months) with hospital or home event-recording (impedance). 62% of events above threshold were due to movements or loose ends, 32% to false alarms (cardiogenic artefact, low-amplitude signal…) and only 6% was due to true events. It should be clear that in toddlers movement artefacts are more frequent compared to very young infants, but nevertheless the message confirms findings of other authors.

Event-recording, according to Weese-Mayer[69], made it possible to discontinue the monitoring in 50/54 infants since parents could be reassured about false alarms, spontaneous resumption of breathing, gradual decrease in frequency of long apnea's etc. By the use of event-recording (including oximetry), Poets[70, 71] could clarify 19 out of 34 unexplained ALTE and found hypoxemia without apnea/bradycardia in a few preterms. Côté et al[41] demonstrated by means of event-recording that parental event reporting did not correlate with significant events under recording: some clinical events were not significant (63% of correlation) whereas some significant events were not clinically reported (64%). Although the evidence for all of these 3 studies is only weak, they point to a preferential use of event-recording home monitoring devices.

Poets et al[70, 71] proposed inclusion of oximetry to the classical cardiorespiratory monitoring, since not all serious events are accompanied by prolonged apnea's and/or bradycardia's and since prolonged hypoxemia seemed to occur as the only abnormal finding in some infants. A major problem with home oximetry however, remains its sensitivity to motion artifacts.

Some references in the literature concern the proposal of a new type of device; none of these monitors were even tested out properly[72, 73]

Conclusion: PSG for application in infants is poorly standardized; minimal quality standards were not found in the literature. Few studies are available, demonstrating that inter- and intra-rater reliability between PSG evaluators is only moderate (good evidence), in one study the reliability was improved by training. Also for infant home cardiorespiratory monitoring references for minimal quality standards were not found; especially for "over-the-counter" monitors not supervised by professionals this might be a problem. Home cardiorespiratory monitoring is sensitive to false alarms (moderate evidence). It usually is performed by means of impedance registration which is not a reliable method in the detection of obstructive apnea's (good evidence). The available studies could highlight the possible advantages of event-recording at home, but clear comparative studies between the different systems are lacking. Home oximetry has been proposed to be associated with cardiorespiratory monitoring to improve early detection of severe events, but it remains sensitive to artifacts and its added value has not yet been studied well.

Key points

- **Polysomnography for application in infants has been poorly standardized in its performance and interpretation.**
- **For infant home cardiorespiratory monitoring, minimal quality standards were not found in the literature; especially for commercially available monitors this might be a problem**
- **Some studies highlight the possible advantages of cardio-respiratory event-recording at home, e.g. in earlier discontinuation of monitoring.**

3.6 RESULTS (IV): CLINICAL EFFECTIVENESS OF POLYSOMNOGRAPHY IN OTHERWISE HEALTHY INFANTS AND INFANTS CONSIDERED TO BE AT INCREASED RISK FOR SIDS

Are apnea's, bradycardia's or other abnormal PSG parameters predictive of later SIDS in otherwise healthy infants, including preterms/dysmatures and SIDS siblings?

Since the report by Steinschneider[14] in 1972, a lot of attention has been paid to the relationship between apnea's as detected by polysomnography and later SIDS. Although apnea's have been studied most extensively, already in early studies cardiac monitoring was performed at the same time ("pneumocardiography"), especially since the report of Schwartz et al[74] on the possible contribution of long QT-syndrome (cardiac arrhythmia) to the phenomenon of SIDS. Later on, the full polysomnography as described above, became established. Only a few studies address the contribution of EEG and oximetry to the prediction of SIDS. Generally, two categories of studies are found addressing this question: mortality studies and the so-called "physiological" studies. These studies are conducted in healthy infants on the one hand, and in infants with an increased risk for SIDS on the other hand (preterms/dysmatures, SIDS siblings). The role of PSG in infants presenting with ALTE will be considered separately.

3.6.1 Mortality Studies in healthy infants

A first type of studies is prospective mortality studies, in which a large group of infants that underwent PSG is taken in follow-up until they reach the age of one year. PSG of infants that became SIDS-victims during follow-up are then compared to matched controls from the same cohort.

Several authors found the PSG (or in older studies "pneumocardiography") unable to predict SIDS.

The CCOHTA-study[63] from the Canadian Coordinating Office for Health Technology Assessment retrieved the references of 2 large study centres, one from Great-Britain and one from Belgium; as well as 1 additional review. In Great-Britain, a large prospective multi-centre study of 6.914 full terms and 2.337 preterms yielded 29 infants who died of SIDS (diagnosis confirmed by autopsy): no SIDS-case had apnea's longer than 20 sec or signs of cardiac arrhythmia on his prospectively performed pneumocardiographic recording[75]. The preterms in this study had been reported already previously[76]. Sixteen out of the 29 SIDS from the Great-Britain prospective multi-centre study were

further evaluated in a separate study: compared to controls, SIDS had fewer instead of more respiratory pauses[77]. Fifteen SIDS-cases from the 1983 study that were full term born, with one additional case, were separately reported again by Southall in 1986[78]. In-depth further evaluation of the respiratory patterns of 10 SIDS cases from the prospective 1983 study (enlarged to 9856 infants), confirmed the previous findings[79]. Also, from the same 1983 cohort, the cardiorespiratory recordings of the first 10 full-term SIDS-cases compared to 100 control subjects were additionally analysed by spectral evaluations of heart rate and respiratory rate by a blinded evaluator; none of the studied parameters could distinguish SIDS from other infants[80]. Most of these studies are large prospective cohort studies of moderate quality; autopsy was performed in the studied SIDS-cases, but no information is available on blinding of the investigators, except for the study of Gordon[80].

The CCOHTA report[63] also refers to the Belgian group of Kahn, who reports 11 SIDS among a prospective cohort of 2.000 infants[81, 82]. This cohort has later been updated by the author and his co-workers, and has been finally summarized in 2002. In this review, Kahn et al describe 40 SIDS-cases and 607 controls out of a follow-up of more than 20. 000 infants undergoing PSG: the SIDS-victims had more obstructive and mixed apnea's but not exceeding a duration of more than 15 sec. For 18 of these SIDS-cases the cardiac sympathovagal balance was calculated, which was greater than for 36 controls. However, there is an absence of markers in some future SIDS-victims and an overlap in values between the SIDS and control groups. The authors conclude that the contributions of their findings for SIDS remain to be determined. The quality of the studies contributing to the review was moderate.

Included in the CCOHTA-reference list is another review by Bentele[53] which brings together research in this area up to the mid-1980s; in these studies different methods had been applied and it is concluded that the results of numerous studies were at variance and often controversial. Some of the studies of this review were mentioned as individual references in the CCOHTA-study.

Apart from these references, the literature review described above, yielded 3 other studies. One small prospective cohort study concludes that obstructive and central apnea's of more than 2 seconds are significantly more frequent in 7 future SIDS-victims compared to controls; but the quality of this study was judged to be low[83]. Another prospective study (moderate quality) in 401 asymptomatic infants found that all infants (8/401) who subsequently became SIDS-victims, had a normal pneumocardiography[84]. One study evaluating EEG-alteration found that cortical arousal on EEG in 16 future SIDS-victims was less frequent compared to controls[85]. The clinical significance of this finding remains to be determined.

It can be concluded that this first type of large scale, prospective cohort mortality studies of moderate evidence fail to demonstrate a relationship between apnea's and/or bradycardia's (or related parameters like spectra) on pneumocardiographies or PSGs, and later SIDS in otherwise healthy infants.

Cardiac channelopathies and SIDS

Recently, it has been proven by means of molecular genetic research that cardiac channelopathies (e.g. long QT-syndrome, giving rise to cardiac arrhythmia) in some cases are responsible for SIDS[86, 87]. As mentioned by Tester[86], Schwartz[88] draw attention to the prevalence of cardiac arrhythmia's in neonates and the relationship to SIDS, in one of the largest and longest prospective electrocardiographic studies ever conducted in neonates (more than 34.000 neonatal ECG's were performed). Nowadays, it is estimated that

about 5% of SIDS cases in Western countries can be explained by cardiac arrhythmia's. Probably in the next future, as new gene mutations underlying this disorder might be discovered, their prevalence might increase. As such, these cases of infant death by definition don't belong to the group of SIDS anymore. The same is true for specific metabolic disorders in the fatty acid oxidation pathway, which was also illustrated to be a possible mechanism to explain SIDS in about 4% of in SIDS cases[86].

Since the study of Schwartz[88], the debate has risen whether in every neonate an ECG should be performed. The large-scale study of this author led to the insight that on a population-level, abnormal ECGs in neonates are associated with SIDS. However, the positive predictive value of a prolonged QT-interval on ECG in the first week of life for one individual is extremely poor[86]z. Therefore, a systematic ECG performed in every neonate will not lead to a better prevention of SIDS. On the other hand, in every SIDS-case, the possibility of a cardiac channelopathy should be taken in mind. A thorough evaluation of the family history is mandatory since SIDS is only one expression of these channelopathies, some of which also can present with arrhythmia's or sudden death later in life. However, a mutation found in a SIDS-victim can be sporadic as well. Genetic counselling should be proposed to the family of the SIDS-victim, in order to clarify all these details.

3.6.2 Mortality studies in infants at increased risk for SIDS

Mortality cohort studies have also been conducted in infant groups that are healthy at first sight but are at increased risk for SIDS: preterms/dysmatures or SIDS-siblings. Babies born to drug-dependent mothers also are at increased risk for SIDS, but specific mortality cohort studies to our knowledge are not available. (For the discussion of the PSG in ALTE, see further).

Preterms/dysmatures

It has been documented extensively that preterm babies, due to immaturity, are prone to episodes of apnea's or periodic breathing, as well as bradycardia's or desaturation, be it clinically visible or not[89, 90]. The same is true for babies born small-for-gestational-age. These events at first sight can be frightening, and after the appearance of pneumocardiography (later PSG), prematures quickly became candidates for this screening. Moreover, prematures/dysmatures are at an increased risk for SIDS. In this paragraph, the link between apnea's of prematurity (eventually bradycardia's or desaturations) and SIDS and subsequently between SIDS and PSG will be discussed based on the mortality studies available in the literature.

The mortality-studies can be summarized as follows. Hoffman et al[58] performed a very large retrospective population-based study including about 10% of the total USA live-birth population during the inclusion period. He found 757 autopsy-confirmed SIDS-cases which is the largest series ever reported; compared to 1514 matched control subjects the SIDS-cases (preterms or dysmatures as well as term babies) did not show a higher prevalence of apnea's. The weakness of this study lies in its retrospective nature and in the methodology (maternal interview and review of medical records). PSGs were not performed in this study. The English large prospective multi-centre study of moderate quality[76] including 2337 preterms/dysmatures has already been mentioned: future SIDS-cases did not show apnea's longer than 20 sec or signs of cardiac arrhythmia on the prospectively performed pneumocardiographic recording. The review of Lequien et al[51], although of limited quality, confirms

the previous findings. Ravet et al[91] in their review admit that a PSG cannot predict the SIDS; nevertheless, based on expert opinion, they advice that infants with respiratory symptoms or apnea's on the NICU (Neonatal Intensive Care Unit), should have a PSG before they leave the hospital and go home with home monitoring; other infants with birthweight below 1700 grams should undergo PSG at 46-48 weeks PMA (postmenstrual age).

It should be noted that the original studies found[76, 58] are studies of at least 17 years old. This means that extreme preterms that only recently are considered for treatment (e.g. after 23-27 weeks of pregnancy) were not included in these studies. As a consequence, no conclusion can be drawn concerning this group of extreme preterms, and the relationship apnea-SIDS or the predictive value of PSG

In conclusion, SIDS cases before dying probably don't show a higher rate of apnea"s of prematurity than age-matched non-SIDS cases (moderate evidence); moreover, mortality studies show that PSG in preterms or dysmatures that later will die from SIDS doesn't show specific cardiorespiratory abnormalities compared to age-matched controls (moderate evidence). Apnea of prematurity probably can not be considered to be a precursor of the SIDS, and a PSG in preterms/dysmatures does not predict the SIDS (moderate evidence). It should be noted that extreme preterms (23-27 weeks pregnancy duration) were not included in these studies.

SIDS-siblings

Kahn et al[81] in a prospective cohort study of moderate quality, performed PSG tests on a large group of infants, among whom 923 SIDS-siblings. 4 SIDS-sibs died subsequently: 1 with and 3 without previous PSG-abnormalities. Rahilly et al[84] also found that in his prospective cohort study including 322 SIDS siblings all 7 babies who died subsequently had shown normal PSG readings. This study was of moderate quality. Monod[92] studied 650 SIDS siblings of whom 4 later died. PSGs for these infants showed no significant abnormalities compared to control infants; the study is of low quality. Oren et al[93] found that in a prospective cohort study of only weak quality, found that for 64 SIDS siblings (and 9 ALTE) PSG was not predictive of the subsequent death in 5 of them.

Consequently, we conclude that a PSG is not a reliable predictor of the SIDS in SIDS siblings.

Key points

- **There is moderate evidence in the literature (mortality studies) that apnea's and/or bradycardia's on a PSG do not predict the SIDS in healthy infants.**
- **Apnea's of prematurity can not be considered to be precursors of the SIDS, and a PSG in preterms/dysmatures does not predict the SIDS (moderate evidence). It should be noted that extreme preterms (23-27 weeks of pregnancy duration) were not included in these studies.**
- **In SIDS siblings, a PSG is not a reliable predictor of the SIDS.**

3.6.3 "Physiological" studies

A second type of studies, the so-called "physiological studies", tries to find a relationship between apnea's and/or bradycardia's and medical conditions considered as being related to the SIDS because of their increased risk of SIDS: prematures, SIDS-siblings and ALTE infants. It is assumed that these risk groups represent intermediate phenotypes relevant to the SIDS. So, if an increased frequency of apnea's and/or bradycardia's can be found in these conditions compared to normal controls, there might be some evidence to suppose that apnea's and/or bradycardia's are related to the SIDS.

Three points are to be taken into account when evaluating these studies:

- the incidence of SIDS in these subgroups of infants: are they really at increased risk for SIDS?;
- the scientific background to use these conditions as models to study the SIDS;
- the relevance of apnea's, bradycardia's or other "physiological" PSG parameters in predicting the SIDS for these conditions.

Epidemiological studies indeed revealed an increased risk of SIDS for certain groups of infants (see higher). In this paragraph, preterms/dysmatures and SIDS siblings will be considered, for the discussion on ALTE, see further. Can these risk groups be used as models to study the SIDS?

A sound scientific background to use preterms, or SIDS siblings as models to study the SIDS in otherwise healthy, term-born infants is lacking. Although preterms have an increased incidence of SIDS, it should be noted that the average age at the SIDS is different in this group compared to SIDS-victims that were born at term[51, 52]. Even if otherwise healthy, this makes clear that many physiological processes in preterms are different as compared to their term peers. Also, in studies about the SIDS in this group of infants, it is often not mentioned how many of them had other conditions secondary to the prematurity e.g. (stable) brain damage which makes them more vulnerable[94]. The special situation of infants suffering from bronchopulmonary dysplasia due to preterm birth will be discussed further. Studying SIDS-siblings seems to be interesting to get to know more about the SIDS, assuming that they have certain predisposing factors in common to their deceased brother or sister. It probably can be assumed that socio-economic factors mostly are the same for subsequent siblings. However, one can never be sure about the degree of concordance in genetic background between two siblings. Even when taking into account the increased risk for a subsequent sibling to develop the SIDS, taken many siblings together, only a small percentage eventually will die of the SIDS because of its very low incidence[54]. Since epidemiological differences (e.g. different timing of peak incidence) are found between the SIDS and ALTE, these two groups mostly are considered to be separate entities (see further). For all these reasons, there are probably too many confounders confusing results, when taking these 3 subgroups of infants as a model to study the SIDS.

Although their methodological basis can be criticized, many studies are available concerning the relevance of apnea's, bradycardia's or other PSG parameters in preterms/dysmatures, SIDS siblings or ALTE infants to predict the SIDS. Some authors clearly state that the purpose of their study indeed is not to find a relationship to the SIDS, but rather to contribute to the frequency and significance of apnea's, bradycardia's or desaturation in these risk groups.

Preterms/dysmatures

Very important is the well-conducted study of Ramanathan in 2001[38], reporting for the CHIME Study Group (Collaborative Home Monitoring Evaluation Group). They used continuous home monitoring (and not PSG) to document the frequency of apnea's and/or bradycardia's in risk-groups compared to normal control infants, and stated clearly that an increased frequency of apnea's and/or bradycardia's was not necessarily linked to an increased risk of SIDS; nor did they comment on the clinical significance of these events. Based on a cohort study in 1079 infants including preterms, SIDS siblings, ALTE infants and healthy term control infants, they concluded that cardiorespiratory events were quite common (41% of infants) also in healthy term infants, and were only statistically more frequent in preterms (and not in ALTE infants or SIDS-siblings). 75% of these events included apnea's whithout a bradycardia. "Extreme events" were only common in preterms until 3 weeks post-term age, and ceased before the age of peak incidence of the SIDS. Extreme events were defined as apnea's of at least 30 seconds or a heart rate during at least 10 seconds below 60 beats per minute (bpm), respectively 50 bpm for infants of less than respectively more than 4 weeks post-term age. Apnea's were measured by means of inductance plethysmography, a method not commonly used in home monitoring, allowing to detect not only central but also obstructive apnea's. For the extreme events including apnea's, 70% included at least 3 obstructed breaths; but obstructive breaths are not detected by an impedance pneumography used in conventional home monitoring. So, cardiorespiratory events are more frequent in preterms compared to normal control infants (but not in ALTE infants or SIDS-siblings); and apnea's without a bradycardia are the most frequent events, but are not well detected by conventional home monitoring methods.

Some other "physiologic studies" have been conducted in preterms. Hoppenbrouwers[95] reported in a small case-control study of moderate quality, that in otherwise healthy prematures with apnea's of prematurity of more than 20 sec. on a PSG between 32-36 weeks, a PSG at 4 weeks post-term did not differ from controls without apnea's. Poets et al[70] found similar results: abnormal values at time of NICU discharge for respiration or oxygen saturation normalized between 2-7 weeks post-term age. Barrington[96] in 1996 showed that 91% of very-low-birthweight infants (birth-weight below 1251gr) on PSG showed apnea's of 12 sec or more just before discharge from the NICU; these apnea's were mostly obstructive and were not predictive of a later ALTE as reported by the parents. The quality of the last 2 studies has been rated as good. Cote[41] in a group of 147 infants of which 29 preterms found no relationship between PSG results at discharge and later significant events during home monitoring for the preterms (apnea's more than 20 sec, apnea's less than 20 sec associated with a bradycardia, bradycardia's adjusted to age during more than 5 sec). As compared to the other groups (ALTE infants, SIDS siblings, parental anxiety infants), preterms had the highest incidence of significant events, they also had a 100% concordance between events reported by the parents and significant events on the recorder. For all groups, significant events were most common during the first month of monitoring. Due to its retrospective character, the evidence of this study is weak.[i]

[i] An article accepted for publication in Acta Paediatrica was provided by a member of the scientific validation committee of this study. This article (G. Naulaers et al. Cardiorespiratory events recorded on home monitors: the effect of prematurity on later serious events. Acta Paediatrica 2007) describes a cohort of 96 preterms with abnormal PSG at discharge that were sent home with event-recording home monitoring. The more preterm the infant had been at birth, the older he was (as expressed in post-conceptional or post-term age) before cardiorespiratory events ceased. This confirms that the group of extreme preterm infants (born before 28 weeks of pregnancy) are a vulnerable group that at least for some parameters should be considered apart from the

Only the review of Baird[94], although of limited quality since no methods on reference retrieving are reported, includes some references on developmental consequences of early apnea's in preterms. It should be noted that these references are confronted with the problem of separating the consequences of premature birth from the effects of apnea's. Infants born prematurely have a higher rate of central nervous system injury; these conditions contribute to poor neuro-developmental outcome and also cause apnea's. Also, studies assessing improvement in long-term outcome as a result of treating apnea's of prematurity are few and are confounded by the same issues. Of the available studies, some report an influence of early apnea's on neuro-developmental outcome, others don't. This author advises to respect an apnea-free observation period of 3-8 days before discharging preterms home (based on one reference); in the subset of preterms showing persistent cardiorespiratory events, home cardiorespiratory monitoring until 3-4 weeks post-term age (based on the same references as mentioned above) may offer an alternative to a prolonged hospital stay.

In another review also lacking methods on reference retrieving, Fleming[48] recommends monitoring for preterms (without bronchopulmonary dysplasia, see further) until a period of 8 days free of apnoeic spells is reached (based on Darnall[97], the only reference addressing this issue). After that period continuation of monitoring is not necessary since many authors (as already mentioned) prove that persistence of these episodes (until 2-3 months of post-term age according to Fleming[48]) are not associated with an increased risk of SIDS. The classical preventive recommendations for all young infants are also applicable to preterms or dysmatures; moreover, the potential benefits from following the measures are proportionally greater than for term infants.

Conclusion: apnea's of prematurity, bradycardia's and desaturations are frequently found on PSGs among otherwise healthy preterm infants; they tend to decrease and reach the same frequency compared to term infants around the age of 2-7 weeks post-term. Their peak is well before the peak incidence of SIDS, which is between 4 and 10 weeks of post-term age for ex-preterms. An increased frequency of apnea's and/or bradycardia's as documented by PSG or home monitoring does not allow to conclude about the risk of SIDS (see mortality studies).

The clinical significance of these events, especially on neuro-developmental outcome, is not clear yet.

It should be noted that none of these studies considered specifically the results for extreme preterms (23-27 weeks of pregnancy duration at birth).

SIDS siblings

In the large review of Hunt[54], 31 "physiological" studies in SIDS siblings are taken into account. Many studies show methodological shortcomings (e.g. only 19 studies comment on the performance of autopsy in the SIDS cases), and there are as many studies showing differences for SIDS siblings as there are studies showing no differences for a variance of parameters; sometimes results are conflicting (increased apnea versus decreased apnea). 4 other studies were not included in this review. Monod et al[92] performed PSG in 650 SIDS siblings and compared the results with 146 ALTE and 204 control infants. They found subtle differences between controls and others for very short central apnea's (2-5 sec), the clinical significance of these findings is not clear. In the study of Di

group of preterms born at a later stage of pregnancy. However, no firm conclusions can be made concerning the value of PSG as predictor of later cardiorespiratory events, since no control group (preterms that showed a normal PSG at discharge) was sent home with home monitoring.

Marco[98] the frequency of obstructive or mixed apnea's (more than 3 seconds at least 3 times occurring) was the same in 201 SIDS-siblings compared to the literature for the general population. Especially for the group of SIDS siblings, the study of Cote[41] was biased: only 39% of all SIDS siblings at follow up in the hospital participated in the study, whereas for the other groups this was 84% or more. The methodological quality of the study of Abreu e Silva[99] is very weak.

No conclusion can be drawn from this type of studies in SIDS siblings. Probably there are too many confounders precluding clear outcomes.

Key points

- **In "physiological studies", apnea's and/or bradycardia's are studied in medical conditions considered as being related to the SIDS (because of their increased risk of SIDS), assuming that these risk groups represent intermediate phenotypes relevant to the SIDS.**
- **An increased frequency of apnea's, bradycardia's and/or desaturation as documented by PSG or home monitoring is found in (otherwise healthy) preterms up to the age of 2-7 weeks post-term; the peak is well before the peak incidence of the SIDS.**
- **Extreme preterms (23-27 weeks of pregnancy duration) were not studied specifically and might be a different group.**

3.7 RESULTS (V): CLINICAL EFFECTIVENESS OF POLYSOMNOGRAPHIES IN ALTE

Are apnea's, bradycardia's or other abnormal PSG parameters predictive of later SIDS in ALTE infants?

3.7.1 Mortality studies

Three studies of weak quality could be found[100, 92, 101] in which it was shown that a normal PSG in ALTE infants does not exclude later SIDS.

3.7.2 Physiological studies

Some small abnormalities on PSGs during the 2nd and 4th month of age were found compared to control infants by Guilleminault[102] and Monod[92]. PSGs in ALTE infants showed more mixed and obstructive apnea's of more than 3 sec during sleep, respectively more central apnea's between 2-5 sec. The clinical significance of these results remains to be proven.

Although it concerns relatively small case-control studies, the conclusions of Daniels[42] and Côté[41] are interesting. They found a correlation between PSG abnormalities and the subsequent recurrence of ALTEs. These authors were strict in their definition of "abnormality" and considered apnea's only as being abnormal if the duration was at least 15 respectively 20 seconds; they also used strict criteria for bradycardia's or desaturation. Larger studies still have to confirm these results.

Three small studies comment on the psychomotor evolution in children who had experienced an ALTE. In the study of Baroni et al[103] (weak quality) the outcome at the age of 10 to 14 months was less favourable for psychomotor and mental development; but at long term (7 years and mid-puberty) results of IQ test and behaviour were comparable to typically developing children

(Kahn[104] good quality; Milioti[105] moderate quality). The latter author noticed some subtle neurological deficits like associated movements on neurological examination, but their relevance is unclear.

The use of a PSG for diagnostic purposes in ALTE infants

There is a long list of possible underlying diseases or abnormalities that can give rise to an ALTE. It is generally accepted that etiological exploration should be based on clinical interpretation of the circumstances and the presentation of the ALTE, and this was confirmed by Kahn et al[17]. There is little evidence about the question which examinations are indispensable in an etiological exploration of an ALTE, but some authors make recommendations based on their own experiences, some limited literature or own case series[19]. According to the literature reviews of Kahn[17] and McGovern[25], 23% to 50% of ALTEs remain unexplained after thorough investigation and are classified as "idiopathic". If there is no clinical clue or if other technical examinations are negative, a PSG can be considered to clarify the nature of the event. No studies are available on the percentage of PSGs that actually can clarify the diagnosis in ALTE infants. This seems logical, since no evidence-based diagnostic guidelines are available on which examinations to perform before concluding that "no diagnosis can be found".

Conclusions: a PSG is not a reliable predictor of subsequent death in infants that experienced an ALTE (3 studies of weak quality). There are conflicting results whether it might predict repeat ALTEs; further studies, preferably on large controlled groups, are necessary. A PSG might be useful as a tool to diagnose the etiology of the ALTE, but no literature is available on the precise number of cases that can be clarified by PSG. Further studies are recommended.

3.8 RESULTS (VI): CLINICAL EFFECTIVENESS OF POLYSOMNOGRAPHIES IN OTHER MEDICAL CONDITIONS

PSGs and home monitoring are used for other medical conditions in which infant death is more likely to occur, e.g. congenital abnormalities involving upper respiratory tract like Pierre-Robin syndrome, neuromuscular disorders in an advanced stage, Ondine's syndrome etc. Death in these infants (or older children) in fact is not due to the SIDS, since a medical aetiology can be pointed out.

Generally in the literature, the necessity of home monitoring and/or a PSG is not really questioned, although only a few studies yet addressed the added value of these technical possibilities in the above conditions.

The HTA-study by CCOHTA referred to several publications, all low quality references (case series or small case-control studies), but probably it is difficult to conduct large-scale studies in this group of (severely) handicapped infants suffering from relatively rare disorders.

Specifically BPD (bronchopulmonary dysplasia) secondary to complicated preterm birth, is also concerned in the review of Fleming[48]: it is proposed that these infants should have regular oximetry and should be discharged with home oxygen if their baseline shows basic hypoxia. The negative medical effects of continuous hypoxia (impaired growth, cardiac insufficiency…) in young infants have been demonstrated many times in the general paediatric literature.

Key points

- **PSG is not a reliable predictor of the SIDS in infants that experienced an Apparent Life Threatening event (ALTE).**
- **There are conflicting results whether it might predict repeat ALTE; further studies, preferably on large controlled groups, are necessary.**
- **Clinical guidelines on which examinations to perform in ALTE are available, based on medical experience and some limited literature. PSG can be a useful tool to diagnose the etiology in ALTE, but there is no evidence available on the precise number of cases that can be clarified by PSG. Further studies are recommended.**
- **The necessity of PSG in other medical conditions (like Pierre-Robin syndrome, bronchopulmonary dysplasia, neuromuscular disorders, etc.) is generally accepted.**

3.9 RESULTS (VII): CLINICAL EFFECTIVENESS OF HOME MONITORING

Often it is argued, that it is difficult to prove the value of home monitoring in the prevention of the SIDS, since alarms that led to timely intervention are not followed by medical intervention and hence pass by unnoticed. The only truly convincing evidence for effectiveness of home monitoring in preventing the SIDS would be a controlled clinical trial. Unfortunately, to design such a study is difficult due to the low incidence of the SIDS, and moreover, would raise many ethical questions. In the absence of a controlled study, less compelling evidence for effectiveness can come from epidemiological studies on the incidence of SIDS since the introduction of home monitors.

Two mortality studies of weak quality[93, 101] showed that the SIDS can occur despite home monitoring, in ALTE infants as well as in SIDS siblings. However, the quality of the monitor used was not well documented, and for some cases no autopsy was performed. Kahn[81] in a study of moderate quality found that of 153 infants considered at risk, 150 infants on home monitoring survived whereas of the 3 infants for whom home monitoring was refused (no SIDS siblings), 2 died but no autopsy was performed. Freed[106] in a weak retrospective cohort study found that 13 out of 14 infants died while off the monitor.

Fleming[48] refers to six large epidemiological studies on the incidence of the SIDS. Only after the well-known "Back-to-Sleep" campaign on preventive measures for the SIDS in the early nineties, the incidence of the SIDS declined significantly, whereas the use of home monitoring already had been introduced on large scale at the beginning of the eighties. Two retrospective studies of weak quality, pointing to the same phenomenon, were additionally found[107, 108]

As already mentioned previously, in the home monitoring study of Ramanathan in 2001[38] it was shown that up to 41% of infants experienced cardiorespiratory events exceeding conventional thresholds, also many healthy control infants. These events were only significantly more frequent among preterms up to 43 weeks post-conceptual age. This group was also most likely to experience extreme events. The authors comment that this study did not aim at delineating the pathologic nature of these events.

Some authors advise to use home monitoring in the diagnosis of an ALTE[41], but don't document the percentage of clarified ALTE cases. Others debate vividly on the necessity of home monitoring not only to prevent unexpected death, but to avoid developmental damage to the brain, e.g. in preterms[106] No evidence for this point of view is available.

Conclusion: Based on several large epidemiological studies, home monitoring (before the "Back-to-Sleep study) did not lead to a decreased incidence of the SIDS. For some other indications for which home monitoring is used (diagnosis of ALTEs, prevention of neurological damage in infants with severe cardiorespiratory events, etc.), no literature is available.

Key points

- **The efficacy of home monitoring in the prevention of the SIDS or sudden infants deaths due to other reasons is difficult to prove, since for obvious ethical reasons no randomised controlled trial can be performed.**
- **However, the introduction of home monitoring did not lead to a decreased incidence of the SIDS, as demonstrated in large epidemiological studies.**
- **The effect of home monitoring on psychomotor development by prevention of repetitive cardiorespiratory events has not clearly been demonstrated yet. Further studies are recommended.**
- **Some authors advise home monitoring to clarify the diagnosis of ALTE, but there is no evidence available on the precise number of cases that can be clarified.**

3.10 GENERAL CONCLUSIONS AND DISCUSSION

3.10.1 General conclusions PSG

- In the literature, no clear quality standards exist for PSGs in infants. Many authors, but not all, adopt the same definitions for the physiological parameters; there are concerns about the basic validity (inter-rater and intra-rater reliability etc.) of PSG interpretation which has not been studied very well. Note that the Belgian RIZIV/INAMI reimbursement is conditional on minimal PSG criteria (see also chapter I).
- A PSG is not a diagnostic tool in prediction of the SIDS. Moderate evidence comes from large scale mortality studies for healthy infants as well as subgroups at increased risk for the SIDS.
- There is good evidence that preterms/dysmatures are at an increased risk for the SIDS. It should be noted that many (but not all) of the underlying epidemiological factors associated with prematurity are also factors associated with SIDS. Preterms/dysmatures are also prone to cardiorespiratory events, often obstructive in nature and especially until some weeks post-term age when it stops spontaneously (good evidence). There is moderate-quality evidence that these events (especially the so-called "apnea's of prematurity") are no precursors of the SIDS

(notwithstanding the fact that preterms show an increased risk for SIDS). It should be noted that the group of extreme preterms (23-27 weeks of pregnancy duration at birth) has not yet been studied well and might be a different group.

- The fear is that repetitive cardiorespiratory events in preterms/dysmatures might compromise their outcome (especially their psychomotor development), however, the limited available evidence concerning this topic shows conflicting results. Further studies are recommended.
- The Belgian situation in which a PSG in preterms is reimbursed by the RIZIV/INAMI when performed at 8 weeks of post-term age, corresponds to the scientifically well established knowledge (good evidence) that at this age mostly apnea's of prematurity (or other cardiorespiratory events) spontaneously have ceased. However, literature on the correlation between cardiorespiratory events on a PSG and later recurring cardiorespiratory events in ex-preterms/dysmatures is sparse and of weak quality. Moreover, the pathologic nature of these events is not proven yet since literature concerning psychomotor outcome is scanty. Further studies are recommended.
- A PSG sometimes is used to clarify diagnostic difficulties in ALTE infants. No clear evidence is available in how many cases a PSG can lead to a diagnosis in ALTE; this is difficult to say since no evidence-based recommendations or guidelines are available on which technical examinations should be performed in ALTE and since the diagnosis of ALTE (by definition) often has to rely only on subjective interpretation of parents.
- When taken all ALTE cases together, there is one study of good quality indicating that the incidence of subsequent cardiorespiratory events is not significantly increased compared to normal control infants[38]. However, when separating the ALTE group based on the results of their first PSG, some authors find weak to moderate evidence that infants for which the first PSG was abnormal later on show more cardiorespiratory events (the pathologic nature of these events, again, has not been demonstrated). This evidence needs confirmation by larger, preferably controlled, studies.
- The necessity of a PSG in infants suffering from other medical conditions (e.g. neuromuscular disorders, etc.) in case of clinical symptoms of (cardio-)respiratory insufficiency, is usually accepted in the literature, although only limited evidence is available. It is obviously not easy to conduct good-quality studies in this group of often rare, disabling disorders.

Only one guideline recommends PSG for "obviously very anxious parents", based on "common sense"[109].

According to the levels of evidence for diagnostic tests of Fryback and Thornbury[28] (see methods), there is unclarity about technical efficacy (level 1); and the criteria for level 2 (diagnostic accuracy) are not fulfilled. The overall diagnostic value of a PSG in the prediction of the SIDS is very low.

3.10.2 General conclusions Home monitoring

No clear quality standards are available for monitors in infants, especially for "over-the-counter" monitors this might be problematic.

Cardiorespiratory monitoring is superior to (single) respiratory monitoring; advantages of event-recording are demonstrated in the literature (but without really comparing effectiveness).

Obstructive apnea's can not be detected with the currently available systems; there is one study of good quality that obstructive apnea's might be frequent in infants (without commenting on their pathologic nature or not).

Evidence of moderate quality demonstrates that current home monitors miss a lot of cardiorespiratory events in infants. Besides that, many false alarms are reported.

Secondary to all the above findings, it is concluded that quality assurance to recommend home monitoring as a reliable tool in detecting cardiorespiratory events in infants (without commenting on their pathological nature or not) is lacking.

Oximetry might be more effective in detecting cardiorespiratory events in infants, but there is little literature available concerning this topic. Moreover still many disadvantages (related to false alarms secondary to motion) are mentioned. Nowadays, oximetry is sometimes used at home in certain well-defined medical conditions.

Whether home monitoring is able to prevent SIDS is difficult to study due to the obvious ethical problems involved in such a study. Mortality studies but especially large scale epidemiological studies couldn't detect a decrease in SIDS-incidence attributable to the introduction of home monitoring. Home monitoring should not be advised to prevent SIDS.

As already pointed out, home monitoring should not be advised to prevent the SIDS (moderate evidence). This has been confirmed in preterms/dysmatures.. Extreme preterms (23-27 weeks pregnancy duration at birth) have not yet been studied well and might be a different group. Concerning the fear that repetitive cardiorespiratory events might compromise their psychomotor development, as already mentioned, the limited available evidence concerning this topic shows conflicting results. More studies are needed.

Some debate is going on in the literature whether or not a subset of preterms ready to go home, but still showing persistent cardiorespiratory events, should have home cardiorespiratory monitoring as an alternative to a prolonged hospital stay (which obviously is cost-effective). As already pointed out, there is no uniform evidence available concerning the effect of home monitoring on later psychomotor outcome in these infants and as such it is debatable whether monitoring is necessary. Concerning the apnea-free observation period to be respected before discharging otherwise healthy preterms home, most authors advise to respect a period of 8 days but this is based only on one reference and as such needs to be confirmed in order to obtain better quality of evidence.

The classical preventive recommendations for SIDS in all young infants are also applicable to preterms or dysmatures; moreover the potential benefits from following the measures are proportionally greater than for term infants.

Some authors advise home monitoring to clarify diagnostic difficulties in ALTE: again the disadvantages of the technical equipment have to be kept in mind.

Further, no clear evidence is available in how many cases monitoring can lead to a diagnosis in ALTE cases; which is understandably difficult to study since the diagnosis of ALTE allows (by definition) subjective interpretation and its incidence as well as mortality rates are not well documented.

The use of home monitoring in SIDS siblings is understandable, but again, the technical limitations of the device should be kept in mind: parents shouldn't be given false reassurance. The incidence of the SIDS in SIDS siblings is increased, to a rate comparable to the increase of general infant mortality after a first explained sudden infant death. Probably the recurrence of the SIDS in siblings, although in absolute numbers rare, is due to a complex interplay between genetic and epidemiological factors. This underlines the relative importance of the preventive measures, which have proven their efficacy in the SIDS.

The situation in infants with underlying medical conditions like neuromuscular disorders, bronchopulmonary dysplasia, etc. is different, since the medical indication to supervise them closely is evident; the only question is whether conventional home monitoring is the appropriate way (see higher: reliability of these devices). Mutatis mutandis the same is true for infants of drug-dependent mothers.

No clear evidence is available concerning the duration of monitoring. It should be kept in mind that in the well-conducted study of Ramanathan[38], no significant difference was found for the amount of cardiorespiratory events during home monitoring between normal infants and ALTE or SIDS siblings except for preterms. There is good evidence for spontaneous cessation of cardiorespiratory events in ex-preterms around the age of a few weeks post-term; this can give some information on when to cease eventual monitoring.

Some illustrations on the use of event-recorded monitoring in the cessation of home monitoring are available in the literature.

According to the levels of evidence for diagnostic tests of Fryback and Thornbury[28] (see methods), there are insufficient guarantees on the technical efficacy of home monitoring (level 1); and the criteria for level 2 (diagnostic accuracy) are not fulfilled. The overall value of home monitoring in the detection of cardiorespiratory events in infants is very low.

4 COST-EFFECTIVENESS

4.1 METHODS: FILTERS AND DATABASES

General search terms were identified in accordance to the research scope through related MeSH headings (tree numbers between brackets):

- Polysomnography (E01.370.520.625)
- (Sudden) Infant Death (C23.550.260.322.400)
- Monitoring, ambulatory (E01.370.520.500)

Particular search terms pertaining to the economic assessment of medical interventions were retrieved following relevant MeSH headings (tree numbers between brackets):

- Costs and cost analysis (N03.219.151)
- Technology Assessment, biomedical (N03.880)

Terms resulting from this search were combined by means of Boolean operators and wild-card characters. These filters were then applied to the Pubmed and Embase databases, (mapping selected MeSH headings to the corresponding EMTREE terminology). In addition, a query was entered into the CRD-database (York University), allowing for a more specific health economic search. A further overview of filters and databases is given in the appendix.

4.2 RESULTS

Abstracts for unique entries were analysed. Relevant publications were limited to research including a form of economic analysis. As a result, three publications were selected for further analysis. Two studies are based on previous prospective clinical research[110, 111]. One study included the analysis of primary data[112]. Detailed evidence sheets for these three publications are to be found in the appendix.

The first study, by Waite, compared the use of monitors versus weighing scales in the follow-up of SIDS siblings[112]. Two groups of families (each comprising 50 babies) were randomly assigned to these two protocols. Remarkably, this study presents parents as patients (and not the SIDS siblings). The targeted outcome is described as "satisfaction with their designated methods" and evaluated through means of a survey. The authors conclude that both groups of parents seemed equally satisfied, but a lower overall cost was observed for the scales-group. Several methodological flaws are present in this publication:

- A detailed cost analysis for the proposed cost items is absent.
- The main outcome, "parent satisfaction" is not clearly elucidated (nor quantified).
- Key elements regarding the type(s) of monitors used are insufficiently detailed.

The cost-utility analysis initially proposed by this paper therefore appears to take on a more descriptive form, emphasizing differences in health care utilisation among the two groups.

Steinschneider[110] presents a descriptive cost analysis, comparing home monitoring relying on parental monitoring versus event-recorded monitoring.

Implicitly, the targeted outcome is mentioned to be successful SIDS prevention. Parents were asked to keep an alarm log based on proper observations and simultaneously an event-recording monitor had been installed. The analysed population counted 155 SIDS siblings and was based on previous prospective research[113]. The authors found that event-recorded monitoring could lower patient treatment costs as fewer diagnostic tests (polysomnographies) were performed due to a lower number of reported alarm episodes. Sensitivity analyses, particularly for the assumed apnea and bradycardia parameters, were not performed. The robustness of the study results could be questioned as a result. Nevertheless, it should be noted that reported alarm episodes would probably bring about more additional diagnostic tests than accounted for in the authors' calculations (i.e. 1 polysomnographic test). This observation consequently seems to corroborate the paper's findings.

The third study our literature search yielded is an Incremental Cost Analysis by Zupnancic[111]. The incremental cost-effectiveness per life year gained (LYS) for screening strategies of QT-intervals in prevention of SIDS was assessed. The assessed intervention concerns an ECG-analysis of newly-borns on the third day of life. Observed QTc-intervals exceeding 440 milliseconds are used as a threshold value for suspected SIDS disposition. The diagnostic parameters are based on preceding prospective research[88]. Authors find the results of their analysis to be "within the range for other accepted health interventions", provided high risk SIDS subgroups can be successfully identified. This precluding condition, however, is not convincingly met. Other important precluding hypotheses regard the efficacy of preventive SIDS treatment and further research on the aetiology of the SIDS-syndrome.

4.3 DISCUSSION

The preceding analysis demonstrates the scarceness of scientific evidence on the evaluated topic. The very nature of the SIDS-pathology, characterized by an uncertain aetiology and low incidence, does not seem to allow for "hard" clinical evidence. Consequently, no diagnostic test parameters for polysomnographic readings or home monitoring are readily available. This lack of clinical evidence is reflected by the paucity of relevant economic analyses.

Furthermore, our review showed that the few available studies are of poor quality. Inadequate statement of costs and effect(s), unclear definition of protocols and the application of unrealistic hypotheses are the main shortcomings undermining the various study results.

Regarding the use of home monitoring we would conclude there are indications event-recorded monitoring is a cost-minimising approach when compared to parental observation of monitoring. Further research should focus on clinical protocols applying to Belgium and should include an assessment of robustness for changes in technical parameters defining an alarm episode.

As far as the use of polysomnographic tests is concerned, we find the available evidence to be limited. Only one study was found, indirectly linking polysomnographies (through ECG screening) to SIDS prevention. Furthermore, the abiding uncertainty surrounding the diagnostic performance would not seem to allow for any valid cost-effectiveness estimation to be performed.

Key points

- **Due to the ambiguous aetiology and low incidence of SIDS, few economic evaluations are reported in the literature. Moreover, the existing literature is of poor quality.**
- **There are tentative indications event-recorded monitoring is a cost-minimizing strategy compared to parental observation of monitoring.**

5 BELGIAN SITUATION

5.1 INTRODUCTION

5.1.1 Scope

This chapter aims to present readers with an overview of the evolution, trends and budgetary impact of the use of polysomnographic interventions (PSG) and home monitoring for patients during their first life year in Belgium[i]. All data available to researchers were explored in order to present a descriptive analysis highlighting elements that should be of active interest to policy makers.

The chosen perspective is governmental. Consequently, unless indicated otherwise, all financial data relate to RIZIV/INAMI [ii] reimbursed medical interventions and regard direct[iii] monetary[iv] costs. Data were primarily derived from:

- RIZIV/INAMI databases ("DocHosp, DocN")
- Belgian Ministry of Health (Hospital database MKG/RCM)
- Two large national Belgian sickness funds ("mutualities")

Further background on data sources as well as methodological notes on calculations and specific software procedures can be found in the statistical appendix ("Data Sources and Methods") enclosed to this report.

5.1.2 Background

5.1.2.1 Polysomnographic Hospital Stays

Currently there is no binding legislation on hospitals performing (paediatric) PSGs. In order for the patients(' parents) to receive a reimbursement a physician's prescription is required. In all, we estimate about 66 hospitals (cf. infra) dispose of the necessary polysomnographic equipment.

5.1.2.2 Infant home monitoring in Belgium

Currently, in order to qualify for public reimbursement of an infant home monitoring device, parents should go through one of 12 accredited centres (12 out of the 66 hospitals estimated to perform PSGs). Home monitoring usually starts before the age of one, although infants can be monitored beyond their first life year, depending on the moment the monitoring begins. In general monitoring is prescribed for a maximum duration of 7 months and can be prolonged one time for a duration of maximally 5 months. Monitoring can be suspended at any given time when a (supplementary) PSG shows normal readings. Patients qualifying for reimbursement should adhere to one of 6 specifically defined categories.

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[i] In certain cases infant home monitoring can be extended beyond the first life year.
[ii] the Belgian third party public payer
[iii] e.g. no allowances made for income loss due to parents' waiting time, etc.
[iv] e.g. no allowances made for psychological wellbeing of parents, etc.

By and large reimbursement covers 6 categories of infants[114]:

- ALTE babies, with prior polysomnographic testing performed:
  - In case the ALTE could only be interrupted through a considerable stimulation or mouth-to-mouth respiration and an ensuing PSG did not offer a sufficiently clear and remediable cause;
  - In case stopping the ALTE did not require a considerable stimulation or mouth-to-mouth respiration and a full medical check-up did not indicate a sufficiently clear and remediable cause, but an ensuing PSG did prove to be anomalous;
- SIDS siblings;
- Preterm infants born before the 31st week of gestation with a birth weight below or equal to 1.500 grams, who required cardio-respiratory monitoring until the time of their dismissal from the neonatology department;
- Neonates born with drug-dependent mothers who persisted in hard drug use throughout pregnancy;
- Preterm infants born between the 30th and 37th week and neonates with a birth weight below or equal to 2.500 gram, with prior (anomalous) PSG testing performed;
- Infants not meeting any of the above criteria, but with whom a PSG for alternative medical indications showed up anomalous.

Furthermore, preterm infant home monitoring usually starts before a PSG has been administered and minimally lasts up to when they have reached the age a PSG can be performed ("corrected" age of 8 weeks).

As the above regulation has been introduced from 2003 onward, data collected from the accredited centres have only been available for a short time frame.

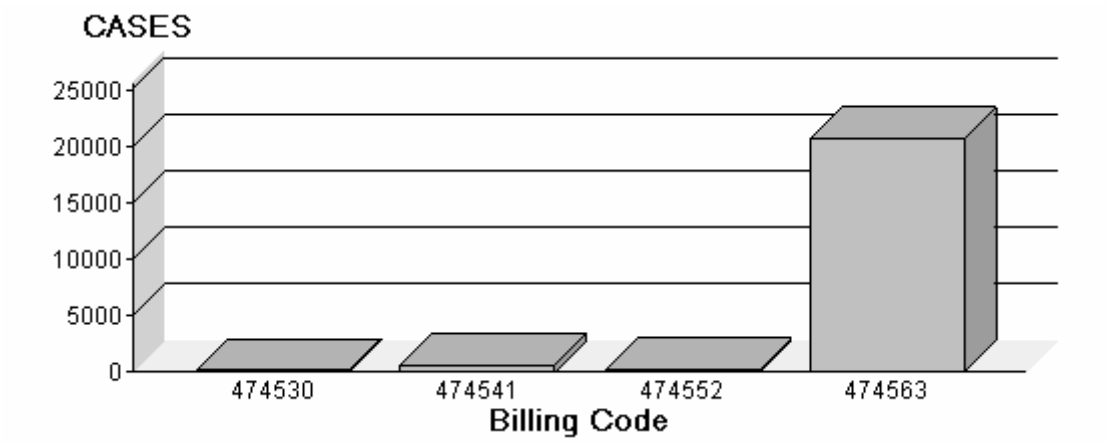
5.2 GLOBAL EVOLUTION OVER THE LAST DECADE

5.2.1 Polysomnographies

Figure 8 depicts the number of paediatric poly(somno)graphic interventions in 2004 reimbursed by RIZIV/INAMI (see also the appendix to chapter 1). As clearly demonstrated, the vast majority of these interventions concern the billing code 474563, "paediatric interventions, polysommetric examination with a minimal duration of 6 hours". The billing code applies to interventions in an inpatient environment. This observation corresponds well with the scope of our report: polysomnographic examinations (of infants below the age of one) in a hospital environment. In all, 20.637 examinations were reported for the year 2004[i].

[i] for the other codes, 474430, 474541 and 474552 respectively 9, 619 and 213 examinations were reported for 2004.

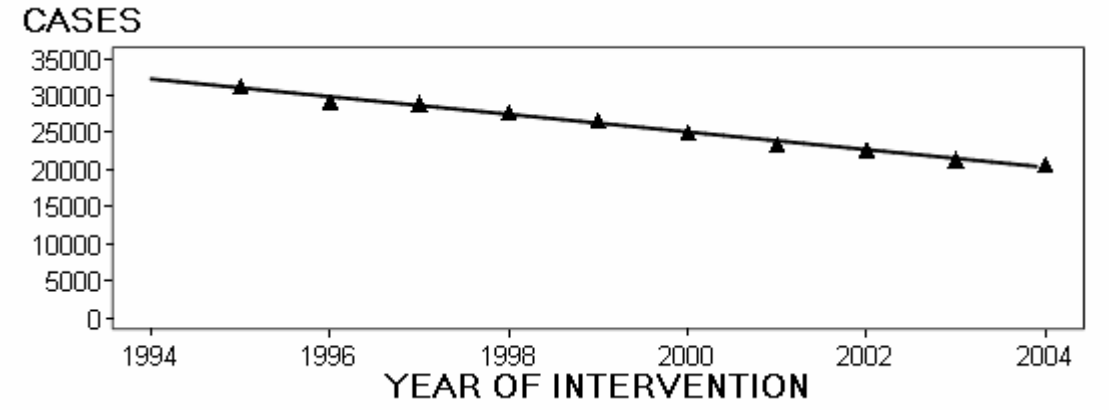
Figure 8. Relevant RIZIV/INAMI billing codes: number of interventions for 2004



Source: RIZIV/INAMI data "N Codes"[114]

The reported number of polysomnographic interventions for paediatric inpatients (these concern patients below the age of 16 at the time[i]) has decreased considerably over the past decade as indicated by figure 9 (scatter plot and fitted (OLS) trend line). Between 1995 and 2004 the overall number of interventions went down by a (regressed) average of about 1.188 interventions per additional year. As the birth rate remained relatively stable for Belgium over the same period (116.000 births in 1995 against 116.000 births in 2004[115, 116]) and the number of preterm births has been reported to increase[117], this appears to be a remarkable observation. Of course, given the rudimentary nature of the plotted data (concerning all paediatric interventions, also covering children beyond their first life year) this conclusion should be made with due methodological caution. Further in this chapter, the age distribution for polysomnographic interventions will be analyzed in depth.

Figure 9. PSG cases by Code 474563: Inpatient PSG, paediatric patient population



Source: RIZIV/INAMI data "N Codes"[114]

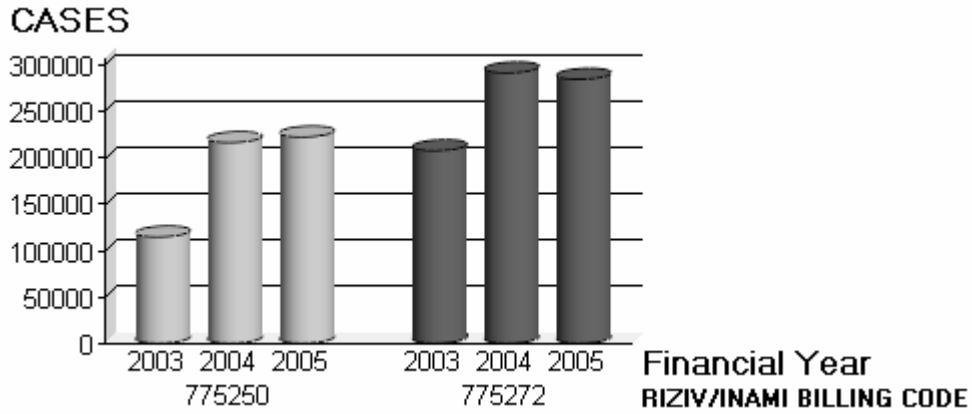
[i] Personal communication by a member of the scientific validation committee dated December 11th 2006.

5.2.2 Home Monitoring

Figure 10 offers an overview of the number of days infants in Belgium received cardio-respiratory home monitoring through RIZIV/INAMI-reimbursement in 2003, 2004 and 2005. As the covered time span is relatively short (recent regulation on reimbursement was introduced from 1st Jan 2003) we opted to display the data by financial year[i].

As can be concluded from figure 10, the use of home monitoring devices without event recorded memory (code 775272) exceeds the use of devices with memory function (code 775250), although the relative use has slightly decreased between 2004 and 2005

Figure 10. Cases of Home Monitoring (number of days)



Source: RIZIV/INAMI data "N Codes"[114]

Key points

- **The number of overnight paediatric PSG hospital stays has declined considerably over the last decade.**
- **A tendency to a relative higher use of memory-enabled devices can be assumed.**

5.3 DATA ON UTILISATION

5.3.1 Polysomnographies

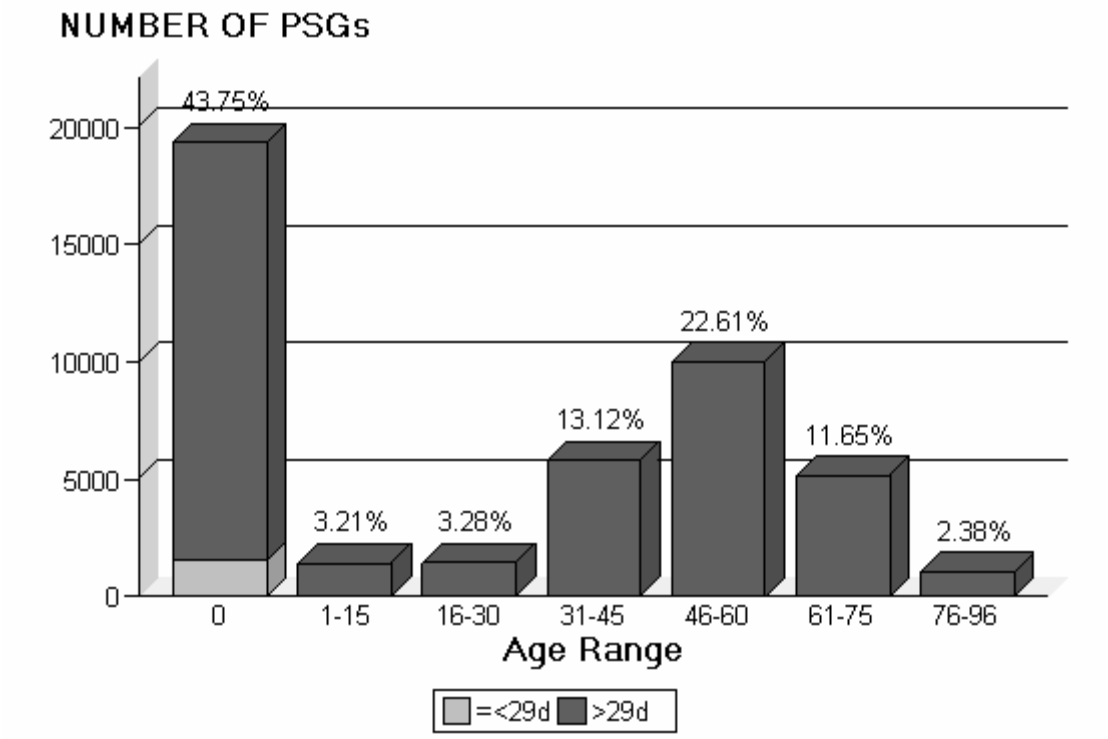
5.3.1.1 Age distribution

In figure 11 we plot the frequency of polysomnographic interventions by patient age groups, including the corresponding percentages. These data concern inpatient polysomnographies as identified by their ICD-9 procedure code[ii] both for a paediatric and adult inpatient population (see also the appendix on data sources and methods). In total 43.634 hospital stays involving a PSG were reported for 2004.

As we can derive from the graph, polysomnographies for patients up to 1 year account for well over 40% of all interventions, or 19.335 hospital stays, in 2004.

[i] i.e. by year of third party invoice settlement, allowing for a wider time frame.
[ii] Code 8917: "POLYSOMNOGRAM"

Figure 11. Hospital Stays with a PSG in 2004 (N=19.335), Frequencies by Age Groups (years + indication for patients below 30 days)



Source: Ministry of Health "MKG/RCM database"[118]

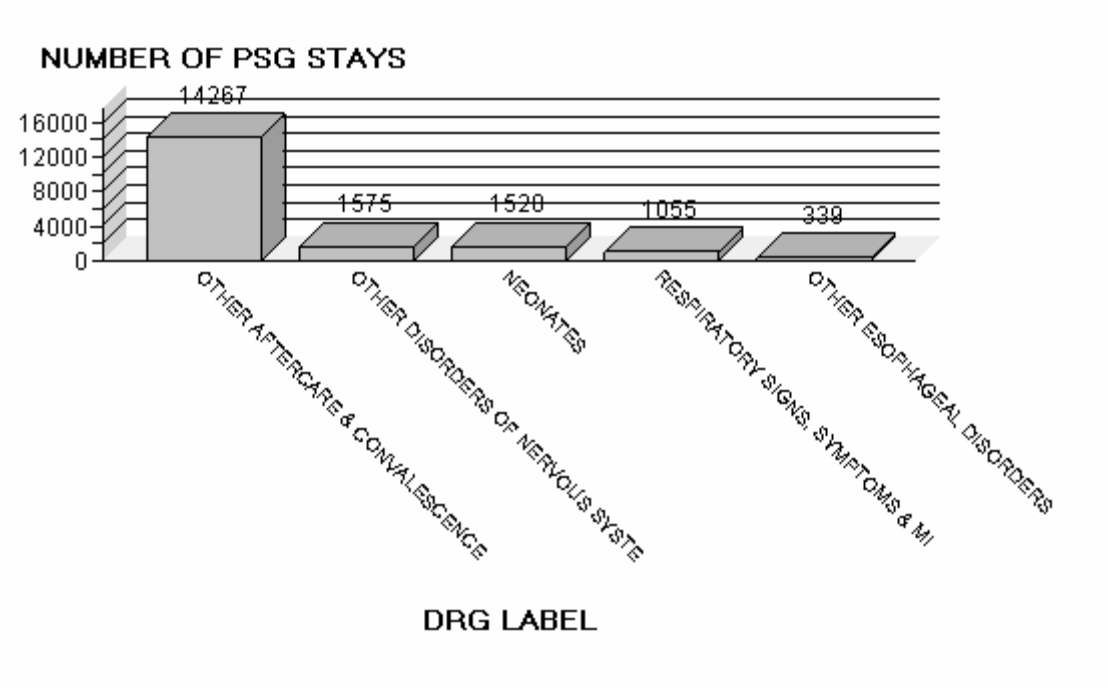
5.3.1.2 Distribution by Diagnostic Group

The data in figure 12 concern all Belgian hospital stays with a polysomnography (as defined by ICD9 procedure code) performed on patients up to 1 year in 2004. Five main categories (with APR-DRG-codes[i] (version 15) relating to neonates taken as one category) account for 96% of all hospital stays in question, with the most important category (APR-DRG 862, "other aftercare and convalescence") representing 73% of stays. As a consequence, questions are raised with regard to patient characteristics for the overwhelming majority of all PSG-stays.

The four remaining categories displayed in the graph, taken together, make up 23% of all PSG stays. These categories correspond to symptomatic groups (respiration, nervous system, digestive system) or a patient subpopulation (neonates) for which the link with the polysomnographic intervention is intuitively clear. It should be duly noted nevertheless that the APR-DRGs 058, 144, 243 all three concern "other" disorders for respectively the nervous system, respiratory system and digestive system.

[i] "All Patients Refined Diagnosis Related Groups", constituting a unique label (with subdivision by degree of severity) per hospital stay.

Figure 12. Hospital Stays with a PSG in 2004, Frequencies by DRG Groups



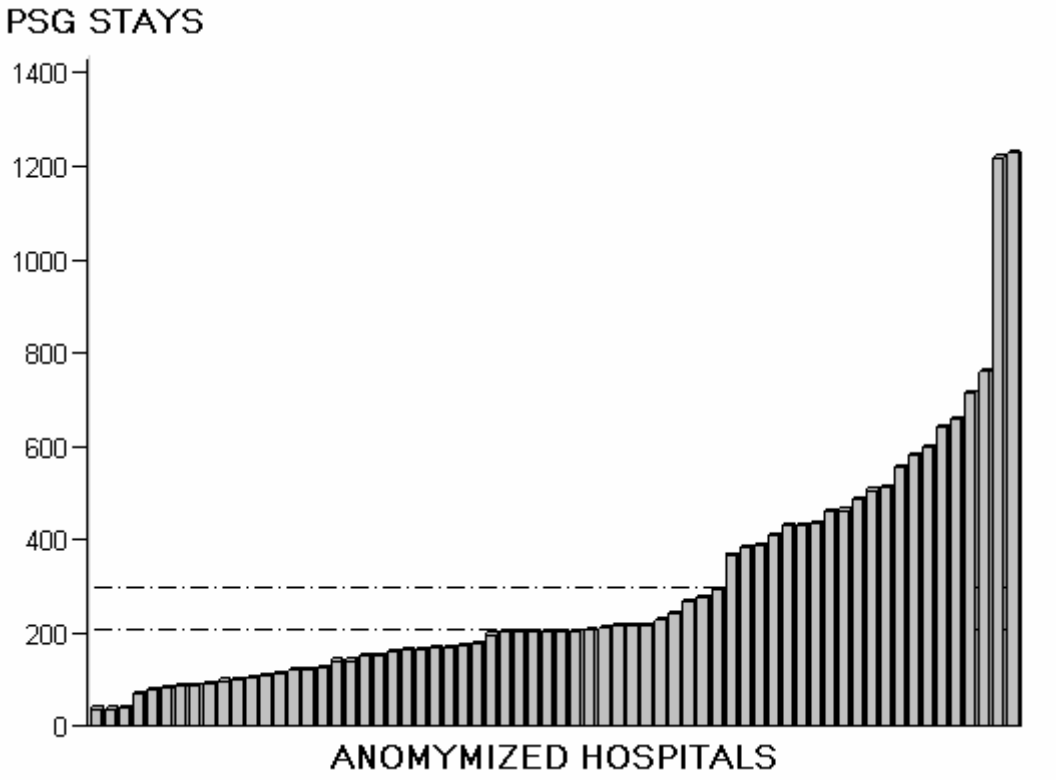
Source: Ministry of Health "MKG/RCM database"[118]

5.3.1.3 Distribution by Hospital

Figure 13 depicts the number of polysomnographies for patients below the age of one for 2004 per hospital (in ascending order). Some interesting observations can be made. Examinations were reported for 76 hospitals in total. Ten hospitals, however, report an unlikely low number for that year (below 10 PSGs). Hence, it seems plausible these data stem from codification errors. Consequently, in figure 13 we plot the number of PSG stays for patients below the age of one for 66 anonymized Belgian hospitals in 2004.

There are two hospitals that stand out, performing over 1.200 interventions in 2004 (accounting for almost 13% of all PSG stays). Our expectation that twelve hospitals (the "accredited centres") would account for a markedly higher number of polysomnographies is not met. In all, the spread of the distribution is considerable (standard deviation, median, mean respectively equalling 246, 205 and 292). Mean and median are plotted on the graph (dashed lines).

Figure 13: Hospital Stays with a PSG in 2004 (N=19.335), Frequencies by Anonymized Hospitals (N=66)



Source: Ministry of Health "MKG/RCM database"[118]

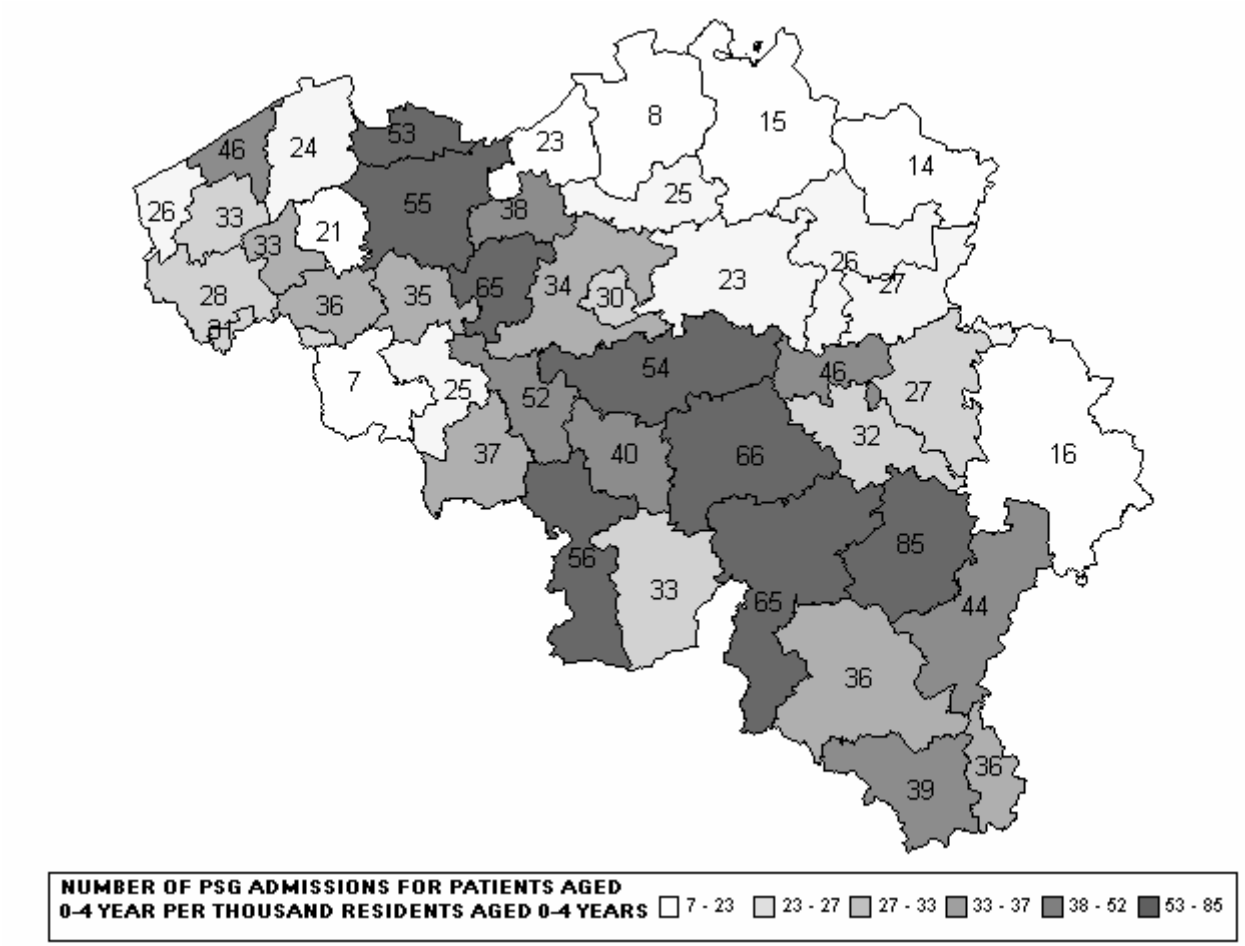
We analyzed the frequency dispersion between hospitals for the period between 1995 and 2004 by different measures of concentration: the Gini coefficient, the coefficient of variation and an exponential measure (for further details on calculations and software see our appendix). We found the overall dispersion of the distribution per hospital to have remained stable, with larger hospitals and smaller hospitals respectively having lost and gained in overall stake.

In conclusion, we find the distribution of overnight polysomnographic interventions to be widely dispersed among hospitals. This overall dispersion has remained stable for the last decade with smaller hospitals having gained a higher stake in the overall number of interventions and larger hospitals having lost some of their share. No clear-cut explanation for this wide dispersion could be found.

5.3.1.4 Geographical Distribution

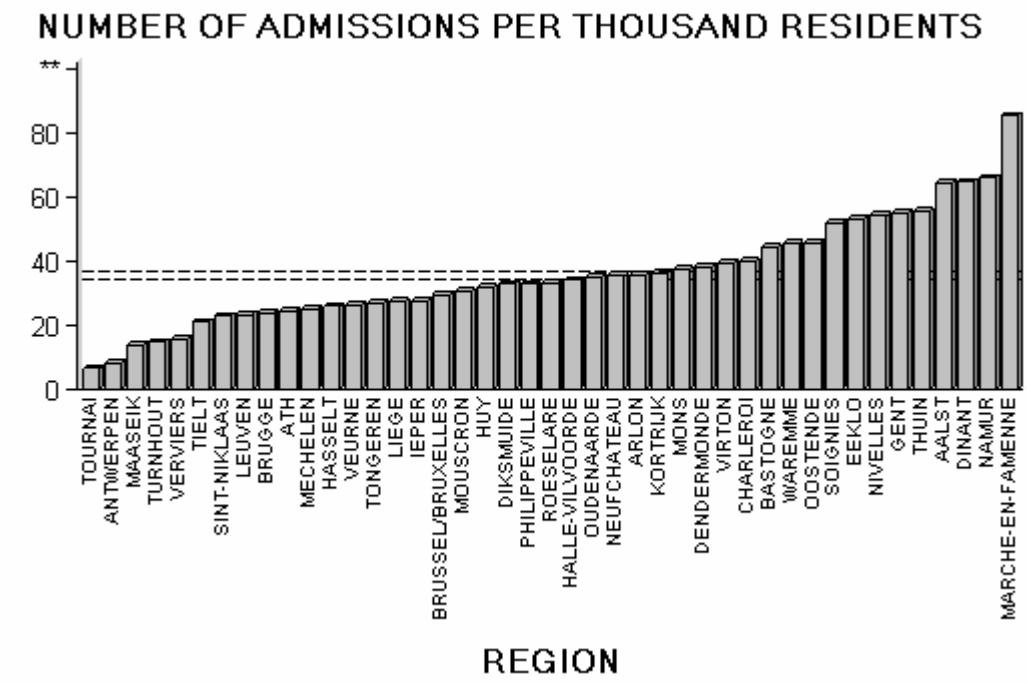
The below chart (figure 14) and following graph (figure 15) indicate that the hospital admission rate for paediatric polysomnographies (for patients up to the age of 4) varies considerably between distinct regions, ranging from as low 7 per 1000 (Tournai/Doornik) up to 85 per 1000 (Marche-en-Famenne), with a nation-wide average of 36, median of 33 (see reference lines) and standard deviation of 16. The data were calculated based on the indicated place of residence for patients. As we only had access to demographic data grouped per categories of 5 years, the denominator in figures 14 and 15 are all children aged 0 to 4 year in a given region.

Figure 14. Belgian density map for PSG admission rates (2004 data, indirectly standardized)



Source: Own calculations based on data by Ministry of Health and National Institute for Statistics[118, 119]

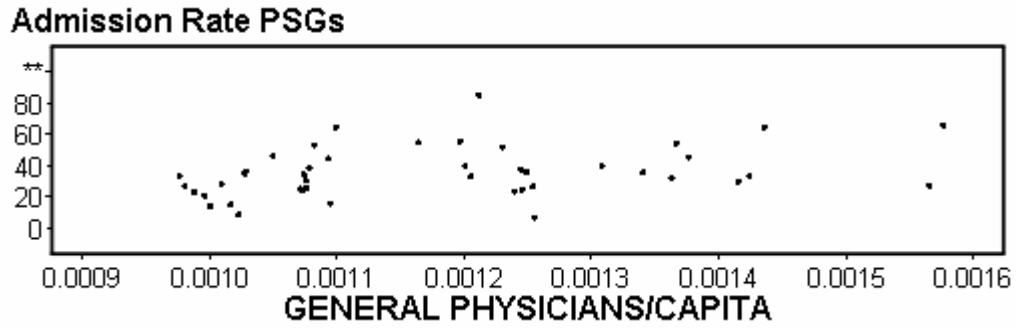
Figure 15. Cumulative Density for PSG Admission Rates per Region, Number of PSGs per 1000 residents aged 0 to 4 years, 2004.



Source: Own calculations based on data by Ministry of Health and National Institute for Statistics[118, 119]

We did not find any significant association (Pearson correlation) at regional level between admission rates and the median revenue declared with tax services (proxy for socio-economic gradient). Nor did any significant association with the number of paediatricians per capita show up, nor with the number of hospital beds (overall or specifically in paediatric wards) per capita (proxies for supply side factors). A moderately positive association (R= 0,33) was found between the number of generalists per capita of overall population and the PSG admission rate. As data on the number of general physicians were available from two sources (RIZIV/INAMI and Ministry of Health) we ran the Pearson correlation on both data sets. In both cases a significant positive association was confirmed.

Figure 16. Scatter Plot for 2004 PSG Admission Rates and General Physicians per Capita (1 observation per region)



Source: Own calculations based on data by Ministry of Health and RIZIV/INAMI[118, 114, 119]

Further exploratory analyses can be found in our methodological appendix.

Therefore we conclude there is a very wide geographical dispersion in admission rates: coefficients of variations equalling 46% and 85% respectively between regions ("arrondissementen", 43 out of a total of 43 in our sample) and lower level municipalities ("gemeenten", 545 out of a total of 589 in our sample). A recent in-house report on regional variation in admission rates per municipality for various surgical interventions[120] found coefficients of variation ranging from 22% (hysterectomy) to 40% (athroscopy of the knee), labelling the latter as "very high disparity"[◦]. Furthermore, we found no straightforward explanation for this dispersion.

Key points

- **Almost half of inpatient overnight PSGs regard patients below the age of one.**
- **The vast majority of PSG hospital stays for our population concerns a related diagnostic that remains unspecified.**
- **We observe a considerable variability between hospitals performing PSGs on our population. No clear explanation for this dispersion could be found.**
- **Similarly, we found a wide dispersion based on patients' place of residence without any straightforward explanation.**

5.3.2 Home Monitoring

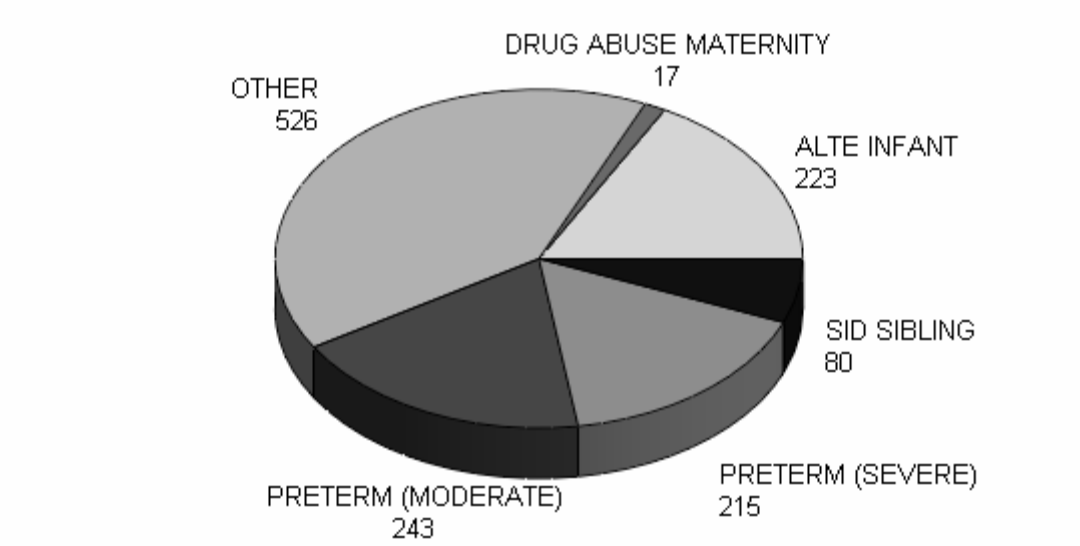
5.3.2.1 Patient distribution by reimbursement category

As can be derived from the below chart (figure 17) the highest frequency of monitored patients (526 patients out of a total of 1304) can be found in category 6 "other patients with an anomalous PSG". This category concerns non-preterm babies with whom a preceding PSG test indicated anomalies in the sleeping pattern. As can be expected preterm and ALTE infants taken together make up the largest part of the studied population (681 patients) whilst the remaining two categories account for a very limited share of all patients.

Furthermore, it is noteworthy that the vast majority of patients receiving reimbursed home monitoring concern patients legally required to have undergone a PSG test (1072 out of 1304 patients).

◦ It should of course duly be noted that surgical interventions in general stem from a more clear-cut patient case aetiology than polysomnographic interventions for infants presumably would, in part explaining the higher dispersion found in our analysis.

Figure 17. Patients receiving monitoring at home 31st Dec 2004 (N=1.305)

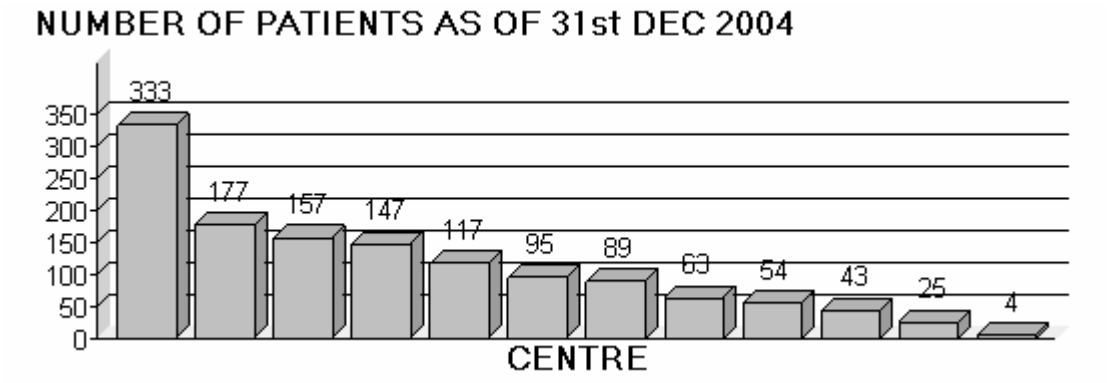


Source: RIZIV/INAMI[114]

5.3.2.2 Patient distribution by accredited centre

The relative range in caseload between the highest and lowest ranking centre is considerable: 333 patients as compared to 4 (see figure 18). Perhaps this dispersion is due to organisational factors as the regulation on monitoring centres is relatively recent.

Figure 18. Home Monitoring Caseload Distributed by Treatment Centre (N=1.305)



Source: RIZIV/INAMI[114]

5.3.2.3 Patient distribution by type of monitoring device

As indicated by table 2 the majority of patients were monitored by means of devices without memory capacity. The use of monitor types varies considerably according to the type of centre patients are related to.

Table 2. Number of patients under monitoring (reference date: 31/12/2004)

CENTRE	PATIENTS/TYPE OF MONITOR			% OF PATIENTS ON MONITORS WITH MEMORY CAPACITY
	WITHOUT MEMORY	WITH AND WITHOUT MEMORY	WITH MEMORY	
1	4	0	0	0%
2	319	14	0	0%
3	77	4	66	45%
4	0	0	54	100%
5	0	0	43	100%
6	37	7	113	72%
7	0	0	117	100%
8	0	0	95	100%
9	121	11	45	25%
10	17	0	8	32%
11	64	7	18	20%
12	53	9	1	2%
Total	692	52	560	43%

Source: RIZIV/INAMI[114]

5.3.2.4 Further Data

The data transmitted by RIZIV/INAMI on home monitoring devices included data on the average duration of monitoring by category (6 indications) and treatment centre. As these data regard the average duration for patients who finished their monitoring in a given year, the underlying patient population does not correspond to the indicated patient numbers that reflect numbers at a precise date for the same 6 categories and 12 centres. Unfortunately, there is no indication in the summary tables (see the appendix to this chapter) we received on the amount of patients the indicated average durations apply to. Consequently, as various treatment centres transmit their data files individually no reliable overall average duration can be estimated. Furthermore, duration of monitoring follow-up is not specified according to the type of device (with/without memory). The need for such a data lay-out would, however, seem self-explanatory as one of the main differences reported in the literature on the topic is the difference in duration of follow-up according to absence/presence of a memory function.

We conclude we cannot make a reliable estimate of average duration based on the RIZIV/INAMI data, nor can we derive a meaningful assessment of the relationship between the type of equipment and average duration. We would therefore urge RIZIV/INAMI to change its data protocol to include the number of patients derived duration that averages apply to in its summary tables and to make the distinction between the two types of devices in their data collection protocol.

In table 3 we present data obtained with the LCM, the largest Belgian mutuality (representing over 43% of all Belgian residents at June 30th 2004). We found that, for the cohort of concerned babies born in 2004 (46.465 in all, out of which 987 babies were monitored for at least one day during their first life year), the duration in follow-up varied according to the type of monitor that was administered. The average duration for memory-enabled monitors apparently exceeds the average follow-up period for simple monitoring devices. This observation may point to heterogeneity between both patients groups (i.e. that Belgian physicians tend to favour memory-enabled devices for more severe patient cases) as centres can put both memory-enabled and simple devices at the patient's disposal.

Table 3. Indicated duration (number of reimbursed monitoring days) by type of monitor administered before the age of one with infants born in 2004.

	Number of Monitoring Days	Number of Patients	Monitoring Days/patient
Memory-Enabled Monitoring	89.592	531	169
Simple Monitoring	61.225	381	161
Both simple and memory-enabled monitoring	15.267	75	204

Source: LCM (Belgian sickness fund)[121]

Key point

- **A large group (around 40%) of monitored patients regard an unspecified subpopulation that presented anomalous PSG findings. The majority of monitored patients are legally required to have undergone a preceding PSG.**
- **The size (measured by patient numbers) of the twelve officially recognized treatment centres varies considerably.**

5.4 INTERNATIONAL COMPARISON

5.4.1 Communications by international experts

In line with the low incidence and vast semantic difficulties underlying the SIDS, we found it hard to obtain comparable international utilisation data. We contacted the various members of the SIDS international organisation[122] with a request for information on (diagnostic) monitoring practices with newly-borns. A list with experts' references is included in the appendix to this chapter.

We received replies for the following countries:

- The US: no protocol has been developed regarding polysomnographic recordings of high risk infants. Medical practices vary according to centres and physicians. Third party reimbursement is dependent on various payor specific criteria.
- The UK: overnight monitoring is only performed (and reimbursed) for infants with previous SIDS cases in the family who have previously been issued an apnea monitor.

- Israel: monitoring devices are only recommended in case of a medical condition (e.g. ALTE), in which case the intervention is reimbursed by health insurance.
- The Netherlands: monitoring is only reimbursed for specific indications (preterm infants, ALTE, etc.) Reimbursement for monitoring of SIDS siblings has also been accepted by insurers.
- France: PSG is reimbursed by public third party when prescribed by a specialist physician. PSGs are regularly prescribed to ALTE infants. No specific allowance is made for SIDS screening/prevention

5.4.2 Available data and related analysis

In looking for comparable data on utilisation we adapted our demand to the use of polysomnographic interventions and home monitoring devices for the population of infants below one year of age, waiving the specific notion of "SIDS prevention".

Through a grey search and expert contacts we identified possible sources for Canadian, French and Australian data. Given the very specific nature of the data demand, however, it proved difficult to extract corresponding data from Canadian and French databases. The Canadian data (pertaining to Québec, the MedEcho database) did not sufficiently specify the precise technical intervention (as polysomnographies are listed under the 0319[P] code which appeared too general for further analysis). The French data we obtained from the CNAMS (employment-based sickness fund) pose considerable problems with regard to the specific age category of our interest.

Below we present data quoted by Marshall e.a. (Australian publication, accepted for publication in Clinical Sleep Medicine, July 2006). These data pertain to 2004 and are repeated in a recent Canadian publication (pre-print 2006).

Table 4. Reported PSG utilisation in general population in 2004

	Australia	United Kingdom	Canada	United States
PSGs per 100. 000 residents (2004)	308	42,5	370,4	427
Total Population (per 1.000,2006)	20.264	60.609	33.098	298.444
Derived Number of PSGs	62.413	25.759	122.595	1.274.356

Source: Own calculations based on Marshall e.a. [123] , CIA World Factbook [124]

As an illustration of the profound methodological difficulties in collecting reliable international data on this specific topic we mention the figure Marshall quotes for Belgium (117,2 PSGs per 100.000 residents), a considerable underestimation by any measure. By ICD-9 procedure code 8917 we estimate the overall number of PSGs for inpatients at around 44.000 in 2004 for Belgian residents. Following RIZIV/INAMI billing codes we count about 52.000 PSGs in total. Restricting our scope to inpatients (as is the case in our report) we find there were some 42.139 interventions in 2004 for all Belgian residents. This latter figure implies a PSG rate per 100.000 residents of 406 in 2004, putting Belgium well above the UK, Australia and Canada and at a par with the United States.

[P] « 0319 AUT.MESURES/EXAMENS NON CHIR.DU SYST.NERV./ORG.DES SENS »

We contacted both the Canadian and Australian authors with regard to our findings and requested for possible data on PSG utilisation by age categories. The Canadian authors stated they had no direct access to local data, whereas the Australian authors replied in a personal communication dated September 25th 2006 that in Australia"the proportion of PSGs given to children (0-16) is usually about 5% per year. About 56% of those were children 0-4 in 2004. Generally, it looks like about half of the paediatric load was 0-4 year olds".

Applying the percentages quoted by the Australian authors (56% of 5% of a total number of 62.413PSGs for 2004) we can deduct around 1.700 PSGs were administered to patients aged 0-4 years old in Australia. We obtained the number of 19.957 PSGs performed in patients aged 0-4 years in 2004 for Belgium from our hospital data (19.335 PSGs for patients aged below one year). Correcting for overall population size, this would imply children aged 0 to 4 years in Belgium stand a chance of being administered a PSG that is more than 22 higher than that of their Australian counterparts.

If we were to assume the number of PSGs administered to Belgian residents below the age of 1 was only 288 and thus proportional to the number of PSGs administered to patients aged 1 to 4 years old in Belgium, patients in Belgium aged 0-4 years would stand a chance of being administered a PSG that is only 0,94 times as high as the chance their Australian antipodes do.

We stress once more considerable methodological caution should be paid in interpreting international comparative data for assessing very specific technical interventions across varying health care systems for very specific subpopulations, as is the case in our report. Nevertheless, we conclude there is tentative evidence the PSG utilisation rate for patients below 1 year in Belgium is particularly high.

Key points

- **Contacts with international experts indicate PSG testing of patients below one year is not a widely established practice.**
- **The (scarce) international data we found tentatively show that the number of overnight PSGs performed in Belgium for infants below one year is particularly high.**

5.5 BUDGET ANALYSIS

Our calculations apply to public third party reimbursed costs for 2003 (2003 €) as this was the most recent year for which we disposed of financial data for Belgian hospital stays (aggregate data by the TCT, Technische Cel/Cellule Technique, see appendix).

5.5.1 Polysomnographies

5.5.1.1 Data

MKG/RCM Database

We requested frequency data on the number of hospital stays for patients up to 1 year including a polysomnographic intervention through the MKG/RCM hospital stays database. These frequency data were sorted by DRG and corresponding degree of severity (DRG-DS).

TCT Database

Through the TCT-website (see appendix) we have access to aggregate cost data on Belgian hospital stays (all patients) for the years 1997, 2000, 2001, 2002 and 2003. We opted to apply data for the most recent year available in our calculations. The lay-out of these aggregate data includes per APR-DRG per related degree of severity:

- Average length of stays (days, integer value),
- Total average cost per stay, subdivided into a fixed part (fixed lump sum per day spent in hospital) and remaining cost items such as pharmaceuticals, physicians' fees (variable part).

We calculated average daily hospital costs, split into a fixed part and a variable part, the latter part covering the potential cost for the PSG.

Combined Data

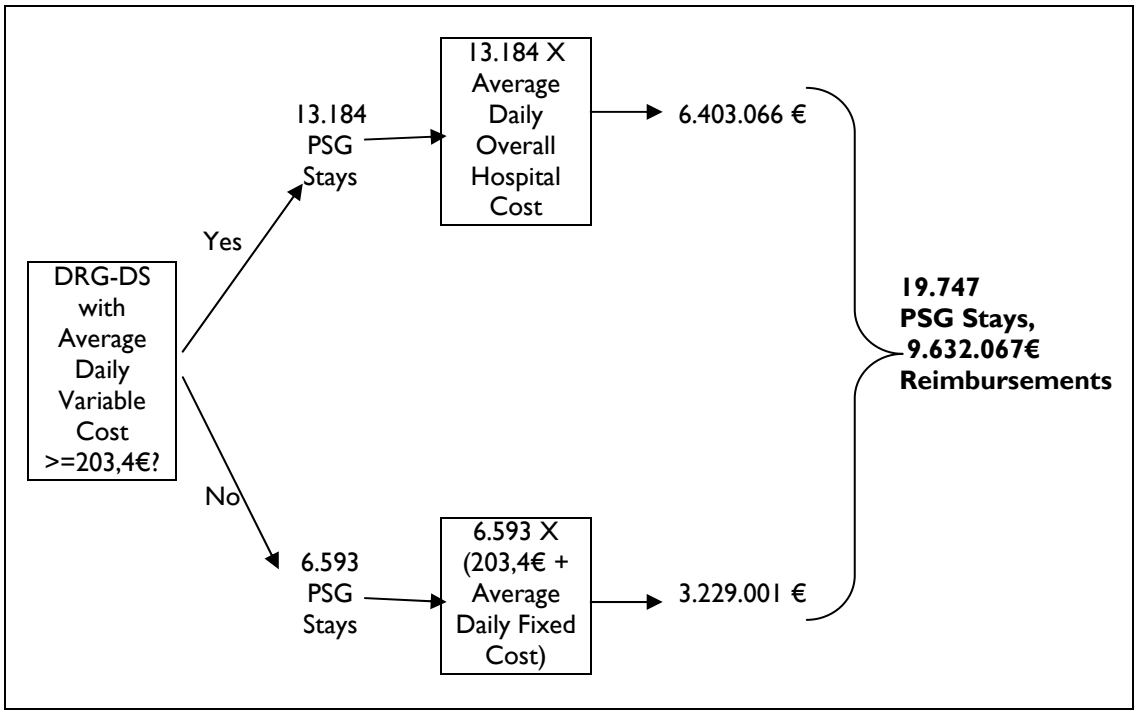
Per combination of DRG and Degree of Severity (DRG-DS) we disposed of the number of PSG stays for patients below one year and the average cost per day (both average fixed and average variable cost) for this type of hospital stay (averages applying to hospital stays for all patients in Belgium).

5.5.1.2 Calculations

Following our cost algorithm (see below) we estimated overall hospital costs for PSG testing of patients up to 1 year in Belgium:

- If the average daily variable cost exceeded or was equal to 203,4€ (the average PSG intervention reimbursement we calculated for 2003), we multiplied the average overall daily cost with the number of concerned hospital stays (13.184 in total)
- If the average daily variable cost was lower than 203,4€, we assumed the latter amount to be the correct (minimal) estimate for all variable costs and multiplied the sum of this amount and average daily amounts for remaining cost items by the number of related stays (6.593 PSG stays in all).

Figure 19. Cost Algorithm for PSG Hospital Stays (2003 data)



5.5.1.3 Results

In total we estimate the cost at 9.632.067€ for 2003 (or about 488€ per PSG), which corresponds to around 0,06% of all RIZIV/INAMI reimbursed medical interventions for that year.

5.5.1.4 Limitations and Assumptions

As we had no access to the MKG-MFG database that would allow us to assess reimbursed hospital costs in detail, we resorted to an approximate estimate. All calculations that aim to identify and attribute costs to complex cost items entail various hypotheses.

Similarly, the above estimate is subject to various assumptions:

- We use overall cost averages (across all patient age groups) to estimate costs for a specific population below one year of age;
- We assume that PSG testing adds one day to the overall LOS[q];
- Applying an average cost per day in hospital implies that overall hospital stay costs would be directly proportional to LOS. It would, however, seem more plausible that days involving technical interventions (such as a PSG) are more costly. This bias will be partly offset by applying minimal variable costs in our calculations for cases where they seem unrealistically low (i.e. below the reimbursement for one single PSG test).

We conclude that the estimate we derived is rather conservative and dominated by our assumption that overall hospital costs within a given diagnostic group are mainly driven by LOS.

[q] This assumption was confirmed by the external experts group linked to the report.

5.5.2 Home Monitoring

In determining the overall (reimbursed) costs for cardio-respiratory monitoring for patients below 1 year, we used the RIZIV/INAMI overall data (per billing code) to derive aggregate costs per year of intervention. For 2003 the combined cost for both memory-enabled and simple monitoring devices was 4.747.110€.

Table 5. Reported costs for Home Monitoring Reimbursement

YEAR	TYPE OF MONITOR	EXPENDITURES	NUMBER OF REIMBURSED DAYS	AVERAGE REIMBURSEMENT / DAY
2003	Memory-enabled Monitoring	1.812.494 €	157.213	11,5 €
	Simple Monitoring	2.934.616 €	290.541	10,1 €

Source: RIZIV/INAMI ()

5.5.3 Polysomnographies and Home Monitoring

5.5.3.1 Overall Result

Combining our cost estimates for overnight PSGs and Home Monitoring in 2003 we can derive an overall cost of 14.379.177€ (0,09% of all RIZIV/INAMI reimbursed medical interventions that year).

5.5.3.2 Cost Allocation Model

Data

In table 6 we show data obtained from two large Belgian sickness funds representing almost 73% of all RIZIV/INAMI insured Belgian residents at June 30th 2003. These data regard the cohort of babies born during the year 2004 who received at least one Polysomnographic / Polygraphic test before the age of one, either as an outpatient or inpatient as identified by the presence of billing codes 474563 (inpatient PSG), 474552 (outpatient PSG), 474541 (inpatient PG) and/or 474530 (outpatient PG).

Data were grouped according to the presence of at least one day of monitoring, sorted by type, before the age of one. With a total of 76.973 babies born for affiliates of the concerned sickness funds, we find that about 16% of all babies born in 2004 were tested. For the population of our scope in particular, patients before the age of one receiving a polysomnography in hospital (billing code 4747563) we estimate[r] around 15% of all babies belong to this group.

Furthermore, for the cohort of babies born in 2004 with the largest Belgian sickness fund we obtained additional data indicating that around 6% received cardio-respiratory monitoring[s] without a preceding P(S)G (either as inpatient our outpatient).

Contrary to our findings on the reported differences in duration of monitoring (see table 6) these data show that memory-enabled monitors are associated with a lower per capita P(S)G rate.

[r] Billing code "474563" overnight paediatric polysomnographies for outpatients accounts for 96% of all cases of paediatric P(S)G, outpatients and inpatients, in 2004 (RIZIV/INAMI data for all Belgian residents).
[s] These patients account for around 5% of all reimbursed monitoring days in the cohort.

Table 6. P(S)G-utilisation for Sickness Fund Affiliates: P(S)Gs before the age of one for patients born in 2004

	Number of P(S)Gs	Number of Patients	P(S)G/Patient (Inpatient or outpatient)
Memory-Enabled Monitoring	1.309	788	1,7
Simple Monitoring	1.989	938	2,1
Both simple and memory-enabled monitoring	369	165	2,2
No monitoring	10.366	10.058	1,0
SUM	14.033	11.949	1,2

Source: LCM and NVSM (Belgian Sickness Funds)[121, 125]

Calculations

We apply the relative population proportions from table 6 and the additional data from the largest Belgian sickness fund:

- number of P(S)G/Patient from table 6 (derived from a population of around 77.000 infants)
- number (and percentage of total) of monitoring days for infants (not) having undergone a preceding P(S)G test (derived from a population of around 46.500 infants)
- (proportion of) average durations for monitoring period of infants with preceding P(S)G by type of monitor (derived from a population of around 46.500 infants)

and split up overall cost in 2003 for PSG testing (see table 7) and home monitoring according to the type of monitoring patients received.

Results

The cost aggregate for patients following the PSG-monitoring protocol is an estimated 14.038.010€ (2003€) or over 98% of total costs for overnight inpatient PSG testing and home monitoring. This merely reflects the fact that the overwhelming majority of infants under home monitoring also have passed or will pass at least one PSG test.

Our data (see table 7 on the next page) indicate monitored babies account for higher per capita overall costs seeing not only the cost for monitoring days should be allowed for, but also a higher proportion of PSGs per infant can be observed in this group.

Within the group of monitored infants, the memory-enabled group presents the highest per capita cost. The higher average number of monitoring days and higher daily cost for this type of devices are only partially offset by a lower PSG rate. This may indicate substitution effects are at work, similar to the ones reported in the literature (see chapter IV)

Table 7. Cost Allocation Model for PSG-Monitoring Protocol: cost estimate for 2003

	#PATIENTS (Estimate)	# PSGs	AVERAGE DURATION OF MONITORING (Days) (Estimate)	#MONITORED DAYS IN ALL (Days) (Estimate)	PSG-STAYS (€)	MONITORING (€)	TOTAL BUDGET 2003 (€,%)		COST/PATIENT (€)
Memory-Enabled Monitoring	1.109	1.842	156	172.761	898.479 €	1.986.750 €	2.885.229 €	20,55%	2.602 €
Simple Monitoring	1.320	2.799	147	193.914	1.365.279 €	1.958.531 €	3.323.810 €	23,68%	2.518 €
Both simple and memory-enabled monitoring	232	519	183	42.573	253.155 €	460.662 €	713.817 €	5,08%	3.077 €
No monitoring	14.153	14.587		0	7.115.155 €	0 €	7.115.155 €	50,68%	503 €
SUM/OVERALL	16.814	19.747		409.248	9.632.067 €	4.405.943 €	14.038.010 €	100,00%	835 €

Source: calculations (see appendix) based on MKG/RCM data by Ministry of Health and utilisation data by LCM and NVSM[118, 121, 125]

Assumptions and Limitations

In presenting the overall budget for 2003 in the way we did in the above table we made various assumptions:

- We extrapolated findings for two subgroups of infants born in 2004 (n1= 30.508 and n2=46.456) to all infants in Belgium below the age of one in 2003: both possible time trend and cohort effects have not been accounted for.
- We assumed that the overall proportions of PSG/Patient for the sickness fund data that were derived for poly(somno)graphic interventions with both inpatients and outpatients are similar for the subgroup of polysomnographic inpatients. This subgroup (identified by billing code 474563) made up 96% of all interventions with Belgian residents for the four related billing codes (474563-474552-474541-474530) taken together in 2004.

Although it should be stressed the split-up of overall costs for 2003 according to monitored patient warrants caution due to the applied hypotheses, we find it a very convenient manner of displaying the overall budgetary impact for inpatient polysomnographic testing and cardiorespiratory monitoring among patients up to one year. It should come as no surprise we find higher overall costs associated with monitored infants as this reflects medical practices for this particular population, which in their turn aim to address the specific medical needs of this patient group.

Key points

- **We estimated the overall cost for overnight inpatient PSG testing for the scoped population at 9.632.067€ (2003€).**
- **Adding the reimbursed cost of home monitoring, we estimated an overall cost of 14.379.177€, or 0,1% of all public third party reimbursements in 2003.**
- **Not surprisingly, our cost allocation model indicates home monitored patients create higher overall costs, mainly due to a higher PSG/Capita ratio in this subgroup.**

5.6 MAIN CONCLUSIONS AND DISCUSSION

5.6.1 Home Monitoring

An overriding conclusion should be that data on home monitoring reimbursement are lacking. On the one hand, current legislation came into practice from 2003 onward, leaving little hindsight to rely upon. On the other hand, the current methodology of data collection leaves to be desired on some points.

From the data we analyzed we notice a tentative tendency to a relatively higher use of memory-enabled devices. Most patient cases involve a prerequisite PSG test, leading us to derive overall cost estimates for a combined "PSG-Monitoring"-protocol. Finally, policy makers should consider whether the current geographic coverage through the 12 acknowledged treatment centres is optimal.

5.6.2 Polysomnographies

We observed a wide variation between hospitals and regions regarding the frequency of PSG hospital stays. No straightforward explanation could be found for this dispersion. Moreover, diagnostic indications for the targeted patient group were found to be too general to derive any meaningful conclusion.

Furthermore, personal communications by international experts and tentative explorations of available data seem to suggest polysomnographic interventions in Belgium are performed in disproportionately high number for patients below the age of one. Remarkably, our data show the use of paediatric PSGs in Belgium was even more frequent in the recent past.

The above observations raise questions with regard to the reason(s) why patients in Belgium below the age of one are so frequently tested (our estimates suggest around 15% of babies in Belgium receive at least one overnight PSG in their first life year).

A further elaboration of the (policy) implications of this chapter's conclusion is offered in chapters VIII (General Conclusions and Disussion) and IX (Recommendations).

6 CONTENT, EVOLUTION AND IMPACT OF PREVENTION MESSAGES TO REDUCE RISK OF SIDS

6.1 GENERAL OVERVIEW

As shown in chapter I, the incidence of SIDS has dramatically decreased. This trend in several countries could be explained for a considerable part by the various prevention campaigns and therefore by changes in parental behaviour[126]. Examples of this evolution can be found in Japan[127], the United Kingdom, The Netherlands, the United States and Sweden[128] and many other countries.

Part of the trend can also be attributed to changes in the definition of SIDS or to a better identification of SIDS (as autopsies were performed more frequently).

However, after a clear decrease in the number of deaths due to SIDS, rates are now levelling off. In spite of prevention measures, a certain number of deaths still remain unexplained. By looking in detail at the information gathered by the government on the remaining cases it could be possible to determine the patients and conditions at higher risk, such as those in lower socioeconomic groups, stress brought on by infants' first days in nursery, situations in which unsafe bedding material is used, etc. Nevertheless, the prevention measures have proven their efficacy in reducing infant mortality. We will expound how these prevention messages are transmitted to the wider population and -if found to be the case- the place of the PSG in it.

6.1.1 Content

The prevention campaigns aim to change the modifiable components in parental behaviour that are related to an increased risk of sudden death by encouraging prevention measures.

They are all related to the environment of the (sleeping) infant: sleeping position, bedding, overheating, smoking (mother/father; active/passive), co-sleeping, absence of breast-feeding[126] and more recently, pacifier use during sleep[6]. None mentioned the use of PSG.

Others risk factors are to be considered but are not modifiable: socio-economic status, cultural membership, age of infant, sex, seasonality[126]. These factors are therefore not targeted by prevention messages.

While the final objective of prevention campaigns is to curb infant mortality, the intermediary outcome targeted is to change risky behaviour into safe behaviour. As the sleeping position is the most obvious risk factor, the recommendations mainly concern(ed) this factor. After all, the sleeping position is the behavioural component that can most easily be modified as related changes are simple in application and not expensive[126].

Gilbert et al. have systematically reviewed the literature about the infant sleeping position and the SIDS between 1940 and 2002[129]. The historical review for the UK shows that in 1944 the usual sleeping position for babies was on their back. As two thirds of the babies who died during sleep were found face down in their bed, a health promotion campaign recommended to avoid the front position. This recommendation was maintained until the mid-50s when many publications took preference for the front sleeping position, a standpoint

advocated until the early nineties. This change in stance was based on a body of physiological research suggesting advantages related to the prevention of gastro-oesophageal reflux, prevention of scoliosis, faster psychomotric development, etc (cited by Mckee[130]). At this point the key message changed, again favouring the side position, against the front position. Finally, from 1995 on, the back position was consistently championed.

Interestingly, an association can be made with markedly lower incidence rates in East Germany, where the prone position had already been avoided for 20 years before the 'back to sleep' campaign in the reunified Germany,[131].

The 'back to sleep' message is now unanimously advocated on the international forum.

Other detrimental factors apart from the supine position were identified: the presence of grandparents[132] in the home or the fact that the baby was a firstborn[132] or rather the opposite: a sibling[133], the age of the mother (20-29 year-old) and the infant (younger than 8 weeks)[133].

Nevertheless, it appears that the risk associated with maternal age, increased parity and male gender have not increased after the back-to-sleep campaign, while the risk of low birth weight infants, the prenatal exposure to smoking, the fact that the mother was unmarried has increased[134].

6.1.2 General Effectiveness

The media found to be the most efficient in passing SIDS prevention messages in the US are television (72%), followed by magazines, midwives, health visitors and last –and it this case least- doctors (11%)[135].

The question of why only a small proportion of parents were informed by their doctor could be answered through the study of Moon et al[132]: among 3717 physicians (GPs, paediatricians, and gynaecologist) it was observed that, while, by and large they are aware of the established risk factors and recognized the importance to discuss the SIDS with parents, but very few discuss the matter routinely or transmit information in writing[132].

It has been shown that informing parents through pamphlets on risk factors of SIDS and prevention measures is an effective approach[136], but the question on the influence of socio-economic status (SES) remains.

As SIDS incidence is associated with SES (mainly race, marital status, employment of mother/father or educational level of mother/father)[137, 138], the fact that prevention messages seem unfit for reaching low SES parents raises a more fundamental question on how to get through to these parents.

The advice best recalled by parents concerns sleeping position, overheating and –quoted by one third of surveyed parents-smoking[135].

In conclusion, prevention campaigns have contributed to a dramatic decrease in the incidence of SIDS in western countries. Nevertheless, rates have been stagnant for the last years, in part probably due to the existence of a high risk population, less reachable or sensitive to health promotion campaign due to its SES, cultural background[138] and/or due to the role of a less malleable behavioural component, i.e. smoking habits.

Regarding the use of the PSG, we did not find indications in the literature that it has any place in prevention campaigns.

6.2 IN BELGIUM

6.2.1 History

Prevention is a competence of the Communities[t] in Belgium. Two public health care organisms are charged with the follow-up of babies, infants and children, i.e. the ONE (Office de la Naissance et de l'Enfance) in the French-speaking part of the country and K&G (Kind & Gezin) for the Flemish Dutch-speaking part. Decisions are then taken autonomously, while further collaborations would be welcome in the mind of their representatives.

In 1994, the ONE decided to dedicate a budget of French-speaking Community reserved for the training of child nursery staff in raising awareness for the quality and safety of the children's sleep[139]. It consisted of training program, first by instilling an awareness of existing liaison persons (one paramedic and one daytime baby minder per family) in an effort to induce a reflection on sleep safety. The campaign was led by means of video material, posters, pamphlets and instructive booklets.

In October 1994, K&G launched its campaign with a preliminary survey on the sleeping environment among 3800 parents to infants aged up to 10 months and repeated the questioning one year after, i.e. March 1995[140].

In 1995, taking the lead from other industrialized countries, a collaboration of the ASTRA foundation and the Ministry of public Health led to a national consensus on infant death prevention, concretized in a national prevention campaign launched in 1995-1996, realized simultaneously by the ONE and K&G.

In 1998-99 the "Centre for Patient Education", a not-for-profit organisation funded by the French-speaking Community, launched its own campaign in the French speaking community[141]. From that point on, campaigns have been mutually co-ordinated as to optimize their effectiveness.

The most recent general campaign in the French-speaking part of Belgium was launched in 2000-2001 "Reducing SIDS risks together"[142].

Since the last national campaign (1995), no specific large-scale campaign was launched in the Flemish part of Belgium, but prevention messages continue to be publicized (available folders or parent dossiers). A Flemish Group of Paediatricians (Werkgroep Studie en Preventie van Infantiele Mortaliteit in Vlaanderen) ensuring the follow-up of recent scientific findings. They prepare an update of the messages, scheduled for 2007, and a campaign should be launched in 2008-2009.

Recently (2006) a national program to help future parents give up smoking has been implemented backed by various organizations involved in the field of health care, smoking behaviour or children's welfare. For the moment, we do not dispose of reliable figures on the prevalence of smoking in pregnant women in Belgium: they are partial, outdated or based on non-scientific sources in view of the difficulty to collect such data (e.g. due to social disapproval s matter)[143, 144, 117]

We observe that, following a phase of actively canvassing prevention messages, currently most of the prevention messages have become part of routine advices given to parents, in general publications targeting young parents, in the

[t]Public organisms based on Belgium's three official languages: Flemish (Dutch speaking) Community, French-speaking Community and German speaking Community.

maternity hospital or by the physicians. Smoking remains a crucial point requesting active awareness.

6.2.2 The campaigns:

The campaigns targeted parents, the general public, but also health professionals or –in one case- children's health professionals.

Hospitals, university centres, professional associations (scientific societies, or groups), well baby clinics (ONE/Kind & Gezin), social insurance funds, parents associations and the national infant mortality observatory were involved in national campaigns, applying different media, depending on the targeted audience:

For the professionals:

- Campaigns by ONE and K&G: Conferences-debate, medical press, flyers, video, e-mail, campaign by phone.
- Campaign Centre d' Education du Patient (CEP): conference, hospital information stands, patient education assessment, video, flyers, posters.

For the parents: press articles, television spots, radio spots, video, posters in settings related to the health or the nursery, flyers given by the professionals to the parents and also, for the ONE campaign, 'télétexte'[u], adding to the infant health record.

6.2.2.1 Content:

National campaign focused on the safety during the sleep. No mention of PSG testing was made

The campaign of the Centre for Patient Education focused on the four main key points to reduce the risk of SIDS: Sleeping position, smoking during pregnancy and after birth, temperature in the sleeping room and safety in the bed.

The message, i.e. 'It is so easy to begin life the right way' emphasized the simplicity of behavioural factors that reduce the risk of SIDS.

Stop Smoking national campaign

To help future parents give up smoking during pregnancy, a program was set up, including financial support for consultations and substitution products. Both pregnant mothers as well as future fathers are targeted.

The campaign used radio advertisements, written press and internet resources (website and e-cards) to raise future parents' awareness of the existence of the program.

6.2.2.2 Impact of the campaigns

The evaluation of the first K&G campaign, 1 year after its launching revealed that, even if the percentage of people smoking in the presence of a baby remained unchanged, the supine sleeping position has become more general and the use of a duvet has decreased[140].

[u] Belgian TV cable based message board ("teletekst" in Dutch)

In 1995, the Astra foundation has conducted a survey to evaluate parental behaviour before the national campaign began. Four hundred mothers of children under 1 year were surveyed.

A similar study was conducted in 1996-1997 by the ONE to verify the impact of its last campaign, interviewing 740 mothers in the French-speaking community.

The evaluation concerned changes in behaviour in line with publicized prevention messages[145].

Questions about the sleeping environment of infants were included in the first (1997), but not the second (2001) nor third (2004) Health Interview Survey for Belgium. There were responses from 150 mothers of a baby born in 1996 or 1997; 49% reported a 'quality' sleeping environment for their child ('quality' being defined as meeting all listed recommendations, taken from Kind and Gezin messages)[146]

Results[145] show that the message concerning the sleeping position has had a noticeable effect in the ASTRA study , though be it in favour of the lateral position and not of supine one, as recommended. The key advice on infants' sleeping temperature was not successfully transmitted. Moreover, one third of the parents did not measure temperature. The use of a bedding cover has diminished as recommended and the surveillance of the sleeping baby has increased; just as the knowledge on the bed (type and organisation) has improved. Nevertheless, the parental smoking habits after birth pose a persistent problem in the implementation of the prevention practices[145, 147].

The poor impact on behaviour could partly be explained by the quality of the involved media. Indeed, in 1999, the Observatory of the infant death ordered an audit of 28 French-speaking media used in prevention of SIDS for the general public (articles and folders distributed to the mothers in maternity homes, daytime nurseries, the prevention organisation, etc)[148]. The assessment criteria focused on content (i.e. are main prevention measures all mentioned) and readability (is the message understandable, easy to remember, etc.) This last approach uses two objective measures: easiness to read (by a count of the syllables by 100 words) and human interest aroused in the reader (by the count of words appealing directly to people and direct sentences).

Results show that 71% of the tools are of sufficient quality for content, 54% are easy to read and 43% are of personal appeal (to readers). In total, only 25% meet all tree criteria.

Moreover, these results indicate that parents from a low educational are less likely to pick up on prevention messages. More, in the Astra foundation study, it was highlighted that parents mainly prefer oral communication as the main source of information about their infant's health[149].

In October 1999, after the last CEP campaign, 1130 young parents of babies aged between several weeks to 8 months have been surveyed[150]. A survey was also carried out with health professionals to evaluate poster stands (n=142) and conferences (n=356) that were part of the general campaign. Results showed that 80% of responding parents complied with the 4 key points of the campaign[v], this percentage surely overestimates genuine parental compliance seeing the likely selection bias (written questionnaires as survey tools). Nevertheless, parents with a low level of general comprehension, below 24 years of age or first-time parents should still be particularly targeted because they remain vulnerable.

[v] Supine position, no smoking during pregnancy and after birth, adequate temperature and safety in the bed

For the professionals, 33% reported having changed their mind on the favoured sleeping position after the campaign, 9% on the effect of tobacco (but 88% were already convinced of this in advance)[150].

The application of a PSG as a diagnostic tool appeared in the 'professional' part of their awareness campaign. Nevertheless, it appears in the evaluation of the campaign that this piece of information was the least consulted by the physicians[151].

The evaluation by the patient education centre revealed also that some parents frowned at the prevention campaigns. Indeed, if it was 'so simple to…' the message is seen to instil guilt with parents who lost a baby. Likewise, the messages were felt to unsettle people who already experienced enough difficulties in their new role of young parents as it was. Consequently, the overall tone of prevention messages was altered.

In conclusion, there is still work to make parents adopting prevention behaviours. Specific groups have to be particularly targeted. Nevertheless, smoking behaviour[147] and SES are difficult to modify. Whether this could easily be achieved remains an open question.

6.3 PLACE OF THE PSG IN THE PREVENTION OF SIDS

We did not find any scientific publication on the use of PSG as a SIDS prevention tool or screening tool to detect infants at risk of SIDS.

PSG has been acknowledged as a diagnostic tool for many purposes but not as a specific nor even an adequate means to 'prevent' or 'screen for' SIDS. This question is dealt with in detail by the clinical literature review of our report.

Key points

- **Since 1994, active SIDS prevention campaigns, targeting multiple stakeholders, have been launched in Belgium.**
- **Campaigns focus on modifiable behaviours, i.e. smoking and the sleeping environment.**
- **Results have been measured several times in terms of evaluating behaviours modifications and tend to show increase in adopting safe behaviours. Smoking remains difficult to modify.**
- **PSG testing does not appear as a prevention measures advised in the campaigns.**

7 PARENTAL ISSUES

As clearly demonstrated in chapter V, the number of PSGs performed on paediatric patients has dropped considerably over the last decade. PSGs can be performed for a variety of medical reasons. Nevertheless, as the overall rate of preterm infants has risen over the same period in Belgium, it appears paradoxical that the admission rate for PSG hospital stays should decline in such a pronounced way. At the same time, we found tentative indications that the number of PSGs performed in Belgium on patients up to 1 year is disproportionately high, even after a decade of steady decline in its utilisation.

Another element highlighted by our organisational analysis is the vast dispersion in PSG use for infants between various regions and hospitals without any easily identifiable explanation that could be pointed out. As no clear medical indication was offered through the hospital databases for the vast majority of these PSGs, it appeared essential to actively explore the field as to identify possible explanations for the observed PSG utilisation rates.

Finally, as established in chapter VI, the use of a PSG is not broached, let alone stressed, in various SIDS prevention campaigns targeting parents. Hence, questions on the reasons underlying the use of PSGs in Belgium for neonates remain unanswered.

Consequently, we decided to interview representatives from the health profession as well as a selected population of parents following a detailed research protocol (related documents can be found in the appendix to this chapter). As our intent was to find a variety of possible reasons underlying infant PSG utilisation, we excluded parents with whom the infant presented a clear medical indication from our scope. Specifically, our research is aimed at parents of infants up to the age of one with whom an overnight PSG test is performed in the absence any clear medical indication. This population is considered the most interesting as our approach is an exploratory one, aspiring to formulate possible hypotheses (and thus preferably as wide a variety as feasible) on the decision process leading up to the infant's PSG.

More in detail, we will discuss various information channels for SIDS and PSG, why do some parents have their baby tested, why do some paediatricians prescribe it in the absence of an evident medical indication and what does a PSG test cost to parents.

In the context of SIDS prevention, we also address the use of monitors. Indeed, in case of a 'positive' PSG or for siblings of SIDS infant, a monitor will be prescribed. It is well known that the use of monitors could reassure parents but that it could also be a source of anxiety or a psychological or social disturbance[152]. We reviewed the literature for the last decade to illustrate the state of knowledge on this matter. Also, we briefly touch upon the financial repercussions monitoring entails for parents.

7.1 MAIN SOURCE OF INFORMATION ABOUT THE PSG FOR THE PARENTS IN BELGIUM

7.1.1 Methodology

In order to identify and describe sources of information as well as the decision process leading up to the actual PSG test, we explored several approaches:

- A wide internet search for grey literature on the topic, also covering websites (internet forums, etc.) that discuss the topic of PSG testing in the context of SIDS.
- We interviewed experts in the SIDS prevention or in paediatric healthcare we have met representatives of the two public organizations in charge of infant and child health, i.e. ONE and K&G, a representative of the French-speaking paediatricians, i.e. the Association of French-speaking Paediatricians, The Groupement Belge des Pédiatres Francophones (GBPF), and one of the Dutch-speaking ones, Vlaamse Vereniging voor Kindergeneeskunde (VVK).
- We have examined the prevention and information tools available or distributed to the parents and the professionals (K&G, ONE and CEP).
- We have interviewed the parents of 15 infants who underwent a (first-time) PSG test, in 4 different hospitals – 1 in Brussels, 1 in Wallonia and 2 in Flanders. Our full research protocol is featured in the appendix to this chapter. The protocol was accepted by various ethical committees (ULB (Free Brussels University) faculty of medicine- local committees of participating hospitals included) Four interviews (2 French-speaking and 2 Flemish-speaking) had to be excluded from further analysis because infants were prescribed a PSG for medical purposes (preterm birth, low birth-weight or heavy illness problems), while criteria for parent selection where clearly explicated to the paediatrician in charge of the selection at the local hospital. Results of 11 retained interviews (6 French-speaking 5 Dutch-speaking) are summarized in a table presented in this chapter's appendix.

7.1.2 Results

The corpus of our analysis contains interviews with representatives of paediatricians' organizations, of public institutions in charge of the well-being of children, interviews with parents, and prevention tools (flyers, books, websites) as well as forums/message boards on the Internet.

We did not carry out an exhaustive qualitative research. Results presented here are only descriptive and restricted to specific infants. We will present our analysis in an effort to identify a range of hypotheses. As such, we do not pass any judgement on the generality of our findings with regard to common everyday practice. Conclusive (in)validation of our findings is conditional on further and widespread prospective research to which our analysis will help lead the way.

We make a distinction between 'formal' channels of prevention messages and informal ones.

Formal channels can be found through the media, health professionals and day time nursery professionals. Formal transmission of information occurs in the context of the individual relationship between the parent and the professional, in the context of an organized prevention campaign. Indirectly, these formal prevention messages will also be conveyed by broader popular media.

Informal channels are restricted to (mouth-to-mouth) personal relationship: friends, family members, acquaintances.

7.1.2.1 Formal channels:

Prevention instances:

Information about SIDS

There is no reference (entry) to SIDS on the ONE website[153]. Prevention messages are spread via the health record of the individual child, published by the ONE and given to the mother at the maternity hospital, or via different flyers regarding safety during sleep and/or via the infant consultations.

Kind and Gezin put forth prevention recommendations in the context of safe sleeping, which are -among others- related to the SIDS[154]. In the Flemish community, messages are diffused by means of 'het ABC van baby tot kleuter[w]' book[155] distributed at the maternity hospital. Preventive behaviour to reduce risk of SIDS is clearly described, even recommended in the health record of the child to be read, while in the ONE documents SIDS is not specifically mentioned rather than safety measures in general.

While prevention material does exist and is available, some of the parents we met reported not to have seen this material in the context of the ONE. In the Flemish community, every parent mentioned the K&G book, even if they hadn't read it yet[x].

On the website of the centre of patient education, one page is devoted to the PSG, indicating that it should be reserved for specific cases and not used as a diagnostic tool[151].

Normally, health professionals and nursery professionals get up-to-date information about prevention measures and should be able to pass this information on to parents. Nevertheless, when interviewing parents, it seems that prevention messages are not systematically conveyed at the maternity home or even by the paediatrician. That is why some of the parents suggest that information sessions at the maternity hospital would be held to explain first-time parents how they have "to deal" with their infant (sleeping position, bed organization, prevention measures, etc) as already done for breast-feeding. In one of the hospital where we interviewed parents, it seems that a booklet is given to parents featuring all of those tips.

Some parents reported having just received precise information on SIDS prevention, when they come to pass the PSG.

[w] "the ABC from baby to toddler"

[x] one of the reason mentioned was that the book was considered to sizable.

Information about PSG

At present, no recommendation is directly given to the parents regarding the PSG examination by the ONE. The PSG does not appear as such in the prevention tool-kit available for parents through the ONE material (flyers, health record, and website). Indeed, the ONE recommends an examination (but not specifically a PSG) in their "Guide of preventive medicine for infant and child"[156] in case of, during his/her sleep, the complexion of the infant changes (to blue or white), if the baby seems to suffocate, is sweating so much or his/her clothes are wet, his/her perspiration smells unpleasantly, is snoring or making conspicuous throat noises when not linked to a cold. This information targets health care professionals (paediatricians and GPs mainly).

K&G are more explicit on the PSG or even the SIDS: there are several pages on their website specifically on SIDS[157]. They explicitly recommend a PSG in several cases (in a direct appeal to parents): in case of previous SIDS cases in the family or in case the infant presents a chronic respiratory problem, the infant has a yellow complexion, he/she is a preterm infant, he/she has a low weight at birth, he/she turns blue when crying and if parents would be worried, even in the absence of the afore-mentioned conditions. In other words, the message to parents and professional is: "if you have a doubt or a worry about the sleep of an infant, have the test done". In the brochures of K&G the place of polysomnography and monitoring has been mentioned: not as a preventive issue but in a sense that they could be prescribed in some cases by the paediatrician. When listening to the representative of K&G, however, they have no mission or message to deliver on this matter: K&G will refer parents to the doctor in charge of the child. And, indeed, parents reported that nor K&G (neither the ONE) spontaneously bring up the PSG topic during their consultations or home visits. Ominously, however, the PSG test is named 'Sudden death-test' in a literal translation from Dutch ("wiegendoodtest") in the messages provided by K&G.

Professional organisations

In Belgium, professionals do not advocate the PSG as a diagnostic tool for the risks of SIDS but stress that a PSG could be part of a more comprehensive patient examination.

Nevertheless, some parents have heard of the test in the context of SIDS, through a health professional. In particular among the parents we met, because a prescription by the paediatrician was delivered or after a visit to the emergency department. This happened therefore when a specific problem of health occurs or when parents talk of the SIDS. In our sample, composed of parents who had the test done out of parental anxiety, no paediatrician has directly or spontaneously suggested the test if the parents did not talk about it.

One parent reported also to have heard of the test on television, in a program on premature babies, set at a paediatric hospital service.

7.1.2.2 Informal channel:

The informal channel of information of PSG is the first one mentioned by the parents: they have heard of the test by acquaintances, friends, family[y], other

[y] They also mentioned internet, but did not point to particular informational websites or parental forums.

young mothers met in the context of infant consultations, day care nursery[z], post-natal physiotherapy, often they carry this information to their paediatrician.

Formal and informal channels are therefore confronted. They are also mixed when parents talk about SIDS or PSG in a medical or nursery context.

In resonance with ongoing practices in Belgium (or perhaps even in amplifying them), insurance companies explicitly mention coverage of (complementary cost for prescribed) PSG tests for infants. Remarkably, one parent mentioned she first heard of PSG through her working environment as her employer (an insurance company) covers the test through its standard policies.

7.1.2.3 Why do some parents have their child tested in the absence of any obvious medical reason?

In default of any obvious reason for test an infant, we identify several reasons why some parents decide to take the test anyway:

- The PSG is considered a screening tool for SIDS:

Parents worry about their infant's health and survival. They dread the idea to live the sudden death of their child and consider this option plausible because, as we have already mentioned it, they heard of cases in their broad social environment or from their insurance company. Once they are aware a test exists, they consider the test as an elementary measure of precaution, regardless of its price. This way, parents seek to be reassured their child is not at risk for the SIDS.

Many of them think that the test will show that their infant is not at risk, that he/she has a healthy sleeping pattern. The most anxious parents would expect to learn their child will be handicapped or might even die soon. The way they interpret test results seems clearly linked to the way they were informed about the test. Many of the parents have no idea of what may follow after a positive PSG. Few have received information on the test and what it measures by the prescribing doctor. The information, if transmitted, is transmitted by nurses. In one particular hospital, all three of the parents we met were unaware of what was happening with their baby. Indeed, it appears that parents at this particular hospital received no explanations on the test (what it measures) and are ignorant with regard to the possible aftermath of an anomalous test result.

More so, if the test is not valid because results are unclear or the baby has not slept enough, parents return home without a monitor. This situation lasting up to the following PSG test is very stressful to them.

On the one hand, for parents who had their baby tested, the test is identified as a preventive measure, and if necessary, getting a monitor in their mindset would be a further way to prevent SIDS. This clearly indicates the parental interest for SIDS prevention in general. On the other hand, however, though able to identify many of the prevention measures diffused by the formal channel, not every parent can name them all. Smoking is not cited by parents. In particular it would seem with parents who smoke (or have only recently quit smoking). This may indicate parents do not spontaneously connect the recommended measures and the risk of SIDS in their minds.

Some parents claim never to have heard about sleep safety campaigns, but it would seem there is a recurrent feeling that the advice on infant sleeping

[z] Members of this research project's experts' group and the two Belgian members of the scientific validation committee also reported that some day care nurseries in Belgium require a PSG test as a precluding condition to take an infant under their care.

position changes regularly in time: at one time children should sleep on the stomach, the other time on their side and then again on their back. As a result, this particular point of advice seems subject to changing 'fashion trends", rather than based on firm scientific evidence.

- Previous children having received a PSG

If parents had a previous child tested, they will repeat the test with younger siblings. This observation emerges from our interviews. This parental "strategy" was also mentioned by the representative of Dutch-speaking paediatricians, referring to a study that indicated that parents want to do everything of what they have done for their previous child, regardless of the intermittent evolution in related recommendations.

- Physicians' advice

Finally, once the parents asked practitioner for a test, the paediatrician was not opposed and prescribed it, at times without any explanation of the specific aim of the test: precise measurements and potential follow-up. Obviously, as we only met parents who received the prescription, it may well be the case that several paediatricians or GPs do not comply with the parent's demand for a PSG. Nevertheless, representatives of K&G, as well as representatives of paediatricians, admitted that some medical doctors refer patients too quickly for a PSG or even a home monitoring device. Moreover, even if the test would indicate there is no reason to get a home monitor, some parents still express the intention of buying an apnea mattress.

7.1.2.4 Why do professionals prescribe a PSG beyond medical necessity?

We can only emit hypotheses on this point because we did not meet doctors who admitted having prescribed a PSG test if it was not medically justified.

Nevertheless, some considerations could be made from the interviews with the health professional representatives and the parents.

Parents declared that their paediatrician accepted or suggested a PSG as a means to put them at ease.

Health professionals report that some paediatricians prescribe it to protect themselves in case of the death of one of his/her little patients, thus from litigious claims and/or to safeguard their reputation, particularly if they work in a small community.

To prescribe a PSG out of financial incentives, i.e. to unrightfully derive income from social security regulations was reputed to be a common practice in the past, but appears to have gone out of date, as stated by the interviewed health professionals.

7.1.2.5 What does a PSG cost to parents?

From the interviews, it appears that parents have no idea on the actual price of a PSG. Some of them have private (complementary) insurance covering patient co-payments, others have not. In both cases, they state not to worry about the price because they believe that the test could save their baby's life, and that no price seems too high in this case.

In reality, whereas the PSG itself is fully reimbursed[aa], patients are due all expenses concerning regular hospital stays. These co-payments depend on the

[aa] as it is considered an intervention that would only be performed in case of "major" health risks.

number of days in hospital, patient medication, the physician's fee (fixed RIZIV/INAMI agreed part). Further additional costs concern higher fees for physicians not adhering to RIZIV/INAMI tariffs and patients' special preferences (e.g. requesting a single bed room). One parent in our sample quoted an overall patient cost for a –standard- infant overnight PSG ranging between 70€ and 80€, which corresponds well with the 75,25€ one major Belgian hospital mentions on its website[158].

7.2 IMPACT OF MONITORING ON PARENTS

7.2.1 Methodology

We have searched Medline, Psychinfo and Embase and (unsuccessfully) Sociological Abstract Database from 1996-2006.

We selected articles written in French, English or Dutch. Search strategies appear in the related appendix with the table of evidence.

We also consulted French-speaking and Flemish-speaking forums on websites identified by Google and further completed available information with results from our interviews.

7.2.2 Results

7.2.2.1 Utilisation of a home monitor or other electronic surveillance devices

If a baby has experienced an ALTE, is a SIDS sibling, is prematurely born (<30 weeks pregnancy), is born weighing less than 1.500 grams, is born between the 31st and 37th week of pregnancy and tested positive on a PSG, is born weighing less than 2.500 grams and tested positive on a PSG, or if results of the PSG for an other indication are abnormal, a home cardio-respiratory monitor will be prescribed. As we have already mentioned, these devices can only be prescribed by one of twelve accredited reference centres.

Besides 'official' monitors, 'unofficial' products exist in the market to detect apnea, mainly apnea mattresses. Following an advice from the Commission for the safety of consumers[bb] from 1999, cardio-respiratory monitors can not be prescribed without the agreement of one of the accredited centres, neither can second-hand monitors be sold[159]. Nevertheless, so-called "apnea mattresses" are still on sale. This product detects every movement of the baby including respiratory movements and will sound an alarm in case of cessation of these movements during a certain time. No specific message about this type of product is clearly put forth by the ONE whereas K&G clearly states devices restricted to respiratory monitoring do not suffice for proper monitoring[154]. However, some parents used these devices or intended to use them if they would not receive a monitor through the official (prescribed and reimbursed) way.

One parent reported having a friend who asked for a PSG because her baby's apnea mattress alarm went off very often, causing a lot of worries. Another stated that the baby's professional daytime minder prefers that children under her responsibility are monitored.

Professionals and prevention organizations advice against such products for safety concerns: they could falsely reassure parents when there are signs that

[bb] Commission de la sécurité des consommateurs: This governmental organization aims to give advices to the Minister of the Economy

must lead them to consult a medical doctor. More so, they could become a source of stress because of untimely alarms. Health professionals also condemn the commercial exploitation of parental concerns[152].

Nevertheless, in general, there is no official message on the use of a monitor by prevention instances. Decision and prescriptions belong to the medical domain and thus to physicians. Nevertheless, the ONE in its role of 'control of daytime nurseries' has stated that if an infant is prescribed a monitor and attends a nursery certified by the ONE, the nursery would not accept him/her without the home monitor, even if parents decided not to use it.

7.2.2.2 Compliance with home monitoring

Even if the effectiveness of a home monitor is still debated, parents have to be compliant with its use, for medical purposes as well as economic purposes in case the monitoring is covered with third party funding[160]. Some articles have reported issues on this matter or suggest interventions to increase the compliance.

Carbone et al have compared the compliance in parents of SIDS siblings, preterm infants with apnea and infants who experienced an ALTE during the first months of use and concluded that parents for the two latter groups used the monitor more frequently and on average for longer periods than the parents of SIDS siblings[161]. The authors explained this partly by the fact that these parents experienced different emotions and displayed different behaviour taking comfort in the low probability of a recurring SIDS diagnosis. They incriminate also the debate on the efficacy of SIDS sibling monitoring. Following the authors, this group has to be carefully educated and supported.

More recently, in the context of the CHIME study, Silvestri et al have tried to measure frequency of monitor use and factors that influence it in order to validate a predictive model for monitor use[162]. They reported that monitor use in the first week was the most important variable for predicting subsequent use. They suggest therefore to assess adherence to recommendations early on.

In improving compliance, simple education, behavioural and case management of non-compliant families seem to be promising strategies[160]. Kurz et al have also reported that an intensive counselling program consisting of clear explanations of PSG readings, indications for monitoring and infant resuscitation, recall of SIDS preventions measures and the intervention of a trained and experienced nurse to called family 2 days after discharge and respond to question on duty leads to better utilization of the monitor.

7.2.2.3 Impact on emotional health

From our interviews, it appears that several parents are aware a monitor could be used to watch over their baby during sleep. Some of them had used it for a previous child. The monitor is perceived as both a source of stress and a source of reassurance. This concurs with ongoing discussions on SIDS forums. As monitors are perceived as a security measure, more anxiety could be created when the monitoring is discontinued.

In general, the literature reports that home monitoring of infants at risk of apnea/bradycardy can affect family members' physical and emotional health.

Several studies focused on consequences for the parents, and specifically mothers, when they have an infant in need of electronic home surveillance. The mental wellbeing of the parents could be endangered as well as the stability of the couple. These studies carried out in the 80's were often retrospective and

could not distinguish the psychological impact (stress, anxiety, depression, etc.) caused by the health status of the child or by the monitoring itself. It is therefore difficult to identify the effects attributable to the illness or to the presence of a monitor. However, on the other side, this presence could be a source of comfort and support.

The study of Abendorth et al showed that during a period of home monitoring, parental anxiety decreased through time, an evolution, however, similar to that of parents whose infant was not monitored[163]. Nevertheless, the overall levels of depression and hostility evolve differently between groups: depression increased in the group of parents who have a home monitor and it decreased in the other group. The hostility level increased in the group with monitor during time but remains unchanged in the one without monitor. During the period of the study, no change occurred in the family functioning.

Williams et al have investigated the effect of monitor on maternal fatigue, comparing mothers of preterm infant discharged with or without monitor[164]. They found that the fatigue increased in the weeks following discharge for mothers whose infant was monitored while it decreased in the comparator group.

The study of Kurz et al indicated that 60% of the families are reassured by the use of a home monitoring, emphasized by the fact that 75% of the families would use the monitor again in the same circumstances, despite the side effects[165]. A positive impact from an intensive counselling program on parental stress was also shown[165] Nevertheless, frequent alarms could be very stressful for parents[166, 165]. That is why an Austrian study has analysed alarms of 26 symptom-free infant under monitor. It was shown that only 15 alarms on 218 classified as true by the parents were genuine true alarms[167]. Authors assumed that, by analysing short-term monitoring records, it is possible to reassure parents and shorten monitoring duration. Another possible approach is offered through an intensive counselling program as for instance evaluated in an Austrian study[165]. The program consisted of standard instructions and advice on infant cardiopulmonary resuscitation. It was observed the program helped to decrease stress and aggressive reactions to alarms (shake the baby or lift him/her up) but do not reduce anxiety.

An interesting conclusive observation is made by Kurz et al[165]: more than 30% of parents to monitored babies would not leave the infant alone with a caretaker, which the authors interpret as an indication of social isolation.

7.2.2.4 Financial impact

If a monitor is prescribed following INAMI/RIZIV regulations, no co-payment is due. From January 1st 2003 on, only cardio-respiratory devices are reimbursed by RIZIV/INAMI[168].

One couple of parents in our sample mentioned paying a rental fee of 12€ per month to their sickness insurance fund for the MR10 respiratory monitor. Rental prices of 75€ per month and purchasing prices of 832€ for the MR10 monitor are reported in the commercial circuit[169].

Apnea mattresses cost more or less 140€. As for the PSG, costs related to the surveillance of the child are not perceived as barrier because it could save his/her live.

An active second-hand market exists on popular websites (e.g. www.ebay.be) for various monitoring devices.

7.3 DISCUSSION

7.3.1 PSG

We specifically set out to meet parents who had their infant pass a PSG for non medical reasons. What follows only concerns this very specific population.

It seems that a (non-quantified) number of babies undergo a PSG to reassure parents for the risk of a SIDS baby. Parents believe that the PSG is a screening tool, although they acknowledge the technology is not totally accurate.

First of all, regarding SIDS prevention in general, information is conveyed by both an official and informal channel. In the latter one, Internet appears as a source for information, which is likely to pass information of varying quality.

The PSG seems not to be suggested to parents by formal channels, except by K&G on their website, where it is advised to talk about it to the paediatrician or the GP in a number of cases.

Nevertheless, the PSG is still perceived as a screening tool by several parents. We could hypothesize that this belief is reinforced by the parents themselves: once knowledge, i.e. information that PSG is a screening test preventing SIDS, is considered a truth for someone, it becomes a personal belief. These beliefs are also transmitted by the different channels. In the case of infant health or well-being, parent's health beliefs could be reinforced by or confronted with medical staff or paramedics (doctor, nurses in the maternity, physiotherapists), family, friends, acquaintances (of acquaintances), other young mothers met in the context of infant consultations, day care nursery, post-natal physiotherapy.

Interviewed parents' beliefs are that SIDS could be screened by a PSG and that it gives them more or less assurance that their child is not at risk. If health professionals do not invalidate this assumption, they implicitly reinforce the belief. Moreover if a PSG is called a 'SIDS-test' (as the literal translation from Dutch reads) it confirms indirectly its utility to detect risk of SIDS. Even if this word is used to popularize messages, it should be banished from the prevention material because it reinforces the inherently wrong message that the test is specifically SIDS-related.

In our contemporary society, the idea of 'not doing anything' to save an infant is intolerable. This probably explains why parents in turn convey the message that the PSG should be used by every parent, or at least proposed to every parent.

Indeed, in western –medicalized- societies, the death of an infant has become rare and therefore less tolerable than before. Therefore, adults will do everything in their power to avoid it. The PSG is perceived in general as a means to reassure adults, i.e. parents and physicians that nothing will happen to the baby they are responsible for. More so, for the professional, it is seen as a safeguard against potential legal action, what appears also in he literature regarding prescription of home monitoring[170]. The responsibility of all of the adults in charge of the infant is preserved. That could be illustrated by the story reported by one parent who told us that the nurse of her child would prefer that every child she cares had first had a PSG and is watched by monitor.

The criteria identifying infant as candidates for a PSG should be explained by the general practitioner or the paediatrician to the parents during the consultation. To achieve this, messages have to be reinforced among health professionals and physicians should be trained to reduce anxiety and fears of parents other than through the prescription of a PSG test when they feel that it is unjustified.

A way to reinsure parents could be, as suggested by the secretary of the Belgian group of French-speaking paediatricians (GBPF). He suggests training parents to administer first aid to their infant if they find him/her unconscious in bed. But this practice has to be first evaluated in terms of organisation (who, when, where?), acceptability, resources, efficacy, effectiveness, etc.

We can also imagine that every infant presenting to be PSG tested without a clear medical reason should first be referred by his/her physician to a sleep laboratory consultation before the prescribing a PSG. This second suggestion has also to be evaluated but it will present the following advantages: Several parents declared having received or read major prevention advices only at the time of the test, in the sleep laboratory. This indicates that by-passed classical prevention messages. Therefore, having a specific consultation on the sleep of their infant could be an occasion to catch up with the 'recalcitrants'. Moreover, as the information is mainly transmitted by written media, remaining more adequate for educated people and less for other people, it will be the occasion to convey orally the messages. However, the fact that information was only noticed at hospital was not reported by every parent, in every hospital. By systematically referring parents to sleep laboratories the parent information protocol can be standardized.

Further, next to the fact that parents do not know the effective utility of the test, we have also noticed that some parents did not know what the PSG measures. Regarding this lack of basic information, we can ask then the question if, for anxious parents, are they going to be reassured after the test, and if so on what?

All these elements pointed to the importance of giving good quality information that allows parents to renounce to the test because it is not justified by the health status of their infant and gives health professionals the occasion to satisfy the parental need for reassurance.

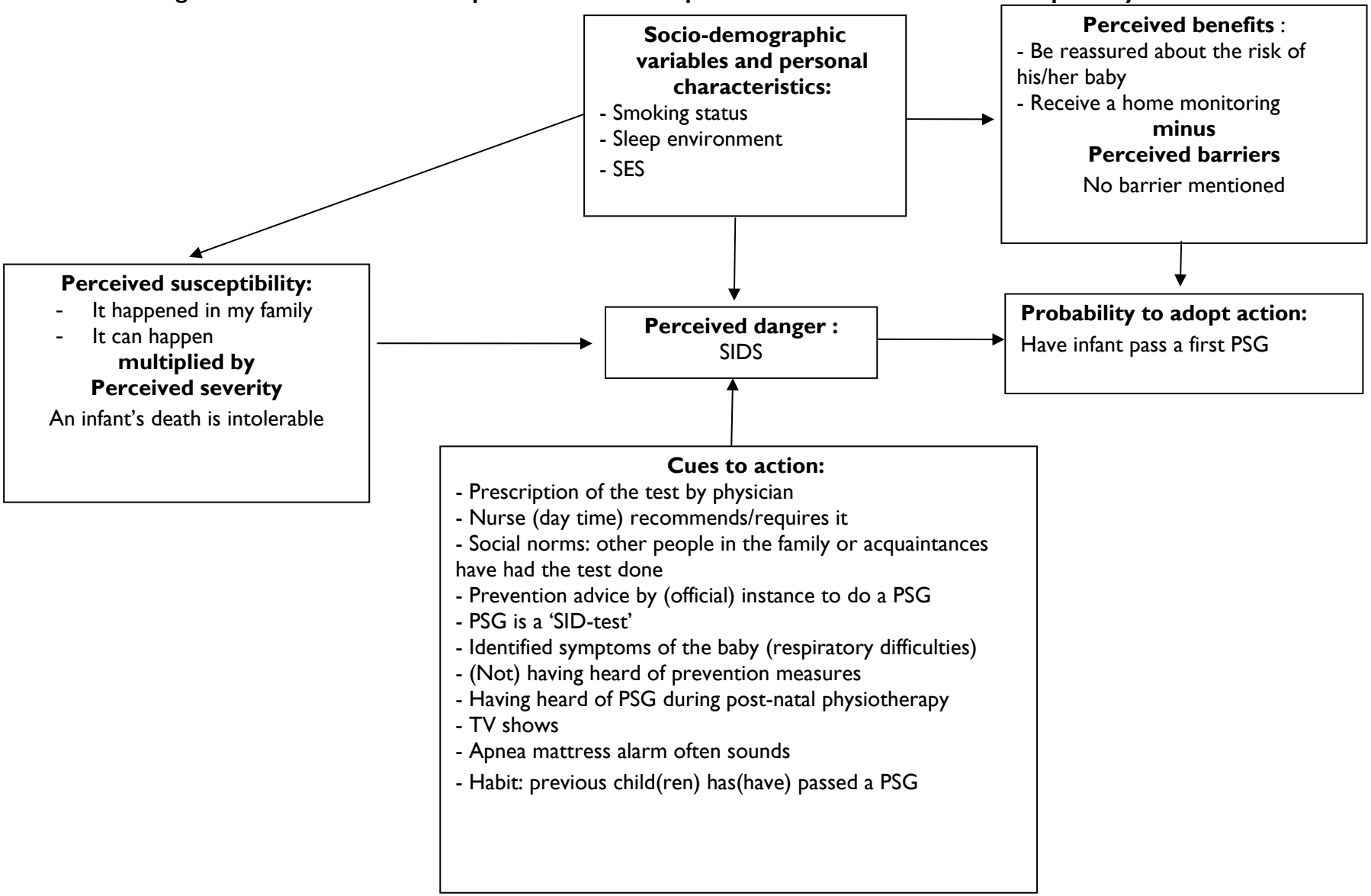
Tentative model on parental motivations

To further the reflection on how to improve the correct use of PSG, we have resumed our findings in a tentative model inspired by the Health belief Model developed by Becker and Rosenstock[171] (see figure 20 below). This model has been applied to health behavior in various instances, i.e. preventive health behavior and clinic use, i.e. physician visit. It has never been validated as such for SIDS prevention. It is based on our findings but could be used also in case of a medically justified decision to pass a PSG. We will only present the model including examples of our findings with the purpose of pointing out possible suggestions to reduce potential inappropriate use of PSG.

Our findings suggest that parents perceive, rightfully or not, the PSG as a screening test for SIDS, SIDS being the danger they perceive and having a PSG being the right strategy to respond to this danger.

The fact that parents see their infant as vulnerable to SIDS and the severity of such an event, i.e. death, will also determine the perceived danger.

Figure 20. Tentative model of parental decision to pass a first PSG to their infant – inspired by the Health Belief Model[171]



Suggestions based on the model:

- Socio demographic and personal characteristics could influence the way danger is perceived, i.e. SIDS. Targeting some specific groups of parents more at risk because of their habits, for example (smoking behavior), still remains recommendable in our tentative model.
- It is impossible to influence the perceived severity of an infant death. However we could probably influence the perceived susceptibility to SIDS by pointing out the most recent incidence rates of SIDS to parents and by clearly explaining the trends in this incidence: SIDS is a scarce event by any measure. To improve and support this knowledge, autopsies have to be more encouraged and performed more systematically.
- The decision to pass a test will also be influenced by the difference between perceived benefit, i.e. 'detect risk of SIDS' and in consequence saving their baby life, and perceived barriers. Notice that no respondent has mentioned barriers to the test. However, we can imagine that, as we only have met parents who managed to do the test, it is quite logical. Nevertheless, we can assume the existence of disadvantages or barriers to parents: for example, to sleep one night at hospital next to one's baby covered with electrodes. Such a statement would probably not be sufficient to counterbalance the benefit of the test. Just as we can guess that the financial aspect would carry little weight regarding perceived benefit, but is a possible target.
- One other perceived benefit is the advantage of having his/her child under electronic surveillance (home monitoring). To raise barriers here, we could point to inconveniencies such as stress, disturbed sleeping patterns, social isolation. Nevertheless, it would not be very ethical to insist on it because if a child really needs a monitor, it will highten parental anxiety. The goal can also be achieved by referring to apnea mattresses that are dangerous and very uncomfortable.
- Environmental influences include perceived prevention measures. If no message has come across, this would increase the perceived danger.
- Social norms, i.e. family, medical and paramedical, acquaintances, etc will downplay or reinforce this perceived danger. As they are well 'placed' both to get correct information and to be in the know of circulating beliefs, nurses and physiotherapists need to be better informed on the use of PSG.
- The term 'SID-test' should be banished from all official channels, as we have already pointed out.
- Habits could also contribute to the decision of passing the test. In our case, habits would for instance be 'if my previous children had one, every next child will too'. By reserving the PSG test to babies who really need it, the prevalence of previous children who were tested will decrease. In consequence fewer parents could develop the habit to test their children.

- Finally, we could interact directly with the response to the perceived danger: other things be done in response to SIDS (anxiety): by adopting prevention measures (insisting on their proven efficacy and explaining why the message on the sleeping position has been changing in the course of time), or by teaching parents how to resuscitate a child.

We do not have enough information on the different elements in the model as the principal research question was not on the adoption of prevention measures. Nevertheless, we can suggest that, it will also be important if we change the model from 'pass a PSG' into 'adopt prevention measure', and particularly 'put baby on its back to sleep',

7.3.2 About the home monitor

Many articles pointed to the difficulties caused in the family by the use of a Home monitor. Therefore it is important to restrict home monitoring to infants for which convincing medical indications apply.

To help parents, who received a home monitor, to manage their anxiety, it is essential to train them to distinguish false from true alarms and how to intervene in case of an emergency. These instructions are normally passed on by nurses at discharge. Furthermore, it parents' psychological troubles, to give them support and a system of technical duty is in place[152]. Concerning the psychological management at the moment of the discontinuation of the use of the home monitor, the use of recording monitor is suggested as useful[152].

The existence of unofficial electronic surveillance devices on the market raises questions. Indeed, they could do more harm than good. Admittedly, the evaluation of this hypothesis is difficult. However, as some parents intend to buy an apnea mattress in case of normal test results, it is very important to assess their mindset after the results of the PSG, eventually to take their anxiety in charge, and, in any case, to remind them of general prevention measures.

Key points - hypotheses

- **Official prevention and professional organisation do not recommend PSG as a screening tool**
- **Parents test their child if it is unnecessary because:**

PSG is perceived by certain parents as a risk of SIDS screening tool for all children. This is further reinforced by a series of factors: use of 'SIDS-test' denomination, the fact that it is not refuted by some of the physicians or that it is suggested on the website of K&G;

Previous child(ren) has(have) been PSG tested;

Physician prescribed it.

- **Some physicians seem to prescribe PSG too rapidly as to reassure parents or to protect themselves from litigious claims.**
- **Home monitors have to be properly used and have a profound impact on the family life. Therefore, their use should remain restricted to children who really need it under strict medical follow-up. The utilisation of unofficial material (apnea mattresses) raises safety concerns.**

8 GENERAL CONCLUSIONS AND DISCUSSION

Despite considerable declines in incidence, SIDS still continues to be the leading cause of post-neonatal infant deaths (deaths between 28 days and one year of age. Due to its specific definition (by "default" of a clear cause of death) SIDS data need to be analyzed with due caution.

The risk of SIDS in individual infants is determined by complex interactions, not yet fully understood, between genetic and environmental risk factors, some of them modifiable, some not. Clinical strategies in prevention of SIDS can target either the reduction of SIDS risk through modifiable (causal) risk factors or the identification of high risk infants.

8.1 CLINICAL EFFECTIVENESS OF THE PSG AND HOME MONITORING FOR INFANTS UNDER ONE

8.1.1 Technical Efficacy

Our literature review shows that both for polysomnographies and home monitoring in infants validated reference values are lacking. Moreover, a technical validation of these devices is absent. There are insufficient guarantees for the quality in the interpretation of results. Specifically for home monitors sold or rented through commercial circuits ("over-the-counter" devices) questions can be raised.

8.1.2 PSG and Home Monitoring in detection of cardio-respiratory (CR) events

We did not find sufficient evidence-based findings to answer the following questions:

(a) Are CR events in infants at risk for those events harmful –in particular as regards the infants' further psychomotor development?

(b) Is there a correlation between events that were detected by a PSG in preterms/dysmatures and recurrent CR events in the months following the PSG?

(c) Can home monitoring of CR events improve the psychomotor development of patients for whom a PSG indicated a high risk for CR events?

With regard to the usefulness of the PSG in clarifying the underlying diagnosis with children who experienced an ALTE, we did not find data to confirm in which cases a PSG could help to establish such a diagnosis. Neither did we find evidence-based recommendations or guidelines on the necessary technical examinations in an ALTE infant. The diagnosis of an ALTE is (by definition) based on subjective parental interpretations.

The usefulness of a PSG in infants suffering from other medical conditions (e.g. neuromuscular conditions, etc.) for which clinical symptoms of CR failure present themselves is generally accepted in the scientific literature, despite the fact that little evidence can be found. The medical indication for a close follow-up of these infants in their home environment is clear. The question whether home monitoring can offer sufficient guarantees remains to be answered.

Oximetry (an optional measure for some monitoring devices) can, because of its proneness to false alerts constitute an alternative in only a limited number of cases. A similar rationale for home monitoring applies to children of drug-addicted mothers.

8.1.3 PSG as a screening tool for SIDS risk, Home Monitoring as a means of prevention for SIDS

Because SIDS is a rare phenomenon and because of obvious ethical reasons no randomized controlled trials (RCT) exist to determine the predictive value of a PSG in the RSIK assessment for SIDS among healthy infants or among infants belonging to an a priori at risk group (e.g. preterm infants). The existing literature clearly shows, based on large-scale observational cohort studies, that the PSG cannot predict SIDS occurrence.

The impact of home monitoring on SIDS incidence can not be determined by means of RCTs. Large scale observational cohort studies did not show a decline in SIDS incidence after the introduction of Home Monitoring.

8.2 COST-EFFECTIVENESS OF THE PSG AND HOME MONITORING

There are few economic evaluations of the cost-effectiveness of a PSG and home monitoring for the prevention of SIDS and more in general a sudden unexplained infant death in the scientific literature. The existing literature is of very low quality. There are indications that the use of memory-enabled home monitoring devices (devices that have the capacity to register CR parameters during an alarm event and to record them for subsequent analysis) are cost-minimizing when compared to the use of simple monitoring devices (without a memory function); in the latter case only parental observations are reported to the responsible physician for further analysis.

8.3 PSG AND HOME MONITORING FOR INFANTS IN BELGIUM: A QUANTIATIVE DESCRIPTION

In 2004, 19.335 PSGs were performed in Belgian hospitals in patients below one year. The total number of "paediatric PSGs" (PSGs in children below the age of 16) has decreased from 31.236 in 1995 to 20.637 in 2004, a decline of 34%. Given the considerable stake patients under one account for in paediatric PSGs, the observed decline can mainly be traced to this group.

The number of paediatric PSGs varies widely between hospitals, with an average (standard deviation between parentheses) of 292 (246) hospital stays in 66 different hospitals for 2004.

Moreover, the admission rate for PSGs in the age group between 0 and 4 years varies strongly between various regions, from as low as 7 stays per 1.000 children to 85 per 1.000 with a national average of 36, median of 33 and standard deviation of 16.

As indicated by APR-DRG (All Patient Refined Diagnosis Related Groups) data 73% of PSG-stays in under-ones for 2004 concern an indication that is particularly vague ("other aftercare and convalescence"), raising the suspicion these hospital stays mainly regard healthy infants.

Based on health insurer data for nearly 77.000 born in 2004 we estimate about 15% of infants in Belgium undergo at least one PSG as an inpatient in their first

life year. This number seems very high compared to foreign countries, as specifically confirmed by our analysis of Australian data.

At a cost of around 500€ (publicly reimbursed cost) and a further 75€ patient co-payment (possibly covered by a private complementary insurance) per PSG-stay the wide-scale practice of performing PSGs in infants is expensive to society (even when only taking the direct monetary cost into account). We estimate the total cost for Belgian social security at 9.632.067€ in 2003.

The available data on home monitoring indicate a trend to a progressively higher use of memory-enabled devices at the expense of more simple devices. Based on health insurer data for 46.465 infants born in 2004 we derived an average duration of monitoring of respectively 169 and 161 days for monitors with and without memory capacity. This finding contradicts expectations raised by the scientific literature on the topic. This may imply an underlying protocol is at work (e.g. whereby more severe patient cases are preferentially equipped with memory-enabled devices).

The publicly reimbursed cost of infant home monitoring (starting during the first life year) was 4.747.110€ in 2003, adding up to an estimated total of 14.379.177€ for PSG-examinations and home monitoring in 2003 in patients below the age of one (almost 0,1% of all publicly reimbursed medical expenses that same year).

8.4 THE PARENTAL PERSPECTIVE: A QUALITATIVE EXPLORATION

Taking the parental perspective as a frame of reference, we sought to find possible answers to the question why so many infants in Belgium are tested. This analysis encompasses interviews with representatives of paediatricians' associations and public institutions responsible for child welfare and healthcare, interviews with parents as well as the perusal of prevention-tools (folders, book(let)s, websites) and internet discussion boards.

The various SIDS prevention campaigns that targeted parents since the nineties did not mention the use of the PSG.

The representatives of Belgian healthcare professionals we interviewed state that the PSG is not advocated as a general diagnostic tool, but is seen as part of a more comprehensive patient examination. This standpoint did not appear to concur with the use of the phrase "wiegendoodtest" (literally meaning "SIDS-test" in Dutch) in one of the organization's documents to designate a PSG in an infant. Furthermore, on the website of Kind en Gezin, a prominent child welfare and healthcare organization "very worried" parents are advised to have their child undergo a PSG without any further medical indication at play.

We interviewed a number of parents (a sample of 11 infants) who had their child tested in default of a clear medical indication as we assumed this way we could explore as wide a variety as possible of parental reasons to undergo a PSG test. Our findings are hypotheses that require further research for conclusive (in)validation:

- PSG is unrightfully seen as a screening tool by parents for all children and hence as a safety measure all parents should take.
- Parents who had an older child tested, will have all younger sibling tested later on.
- Physicians prescribe the PSG too easily, sometimes without further explanation on the specific purpose of the test, precise measurements or the possible aftermath of the test.

The scientific literature shows that home monitoring may have an impact on the emotional and social wellbeing of parents.

8.5 DISCUSSION

Despite of the decrease in the number of PSGs over the last years, polysomnographic examinations in infants are still widely performed in Belgium with over 19.000 hospitals stays of infants under one involving a PSG. The use of a PSG may be envisaged for certain groups, such as preterms/dysmatures, ALTE-infants and infants suffering from very specific conditions. Six large scale epidemiologic studies show that the PSG does not contribute to the detection and eventual prevention of SIDS.

There is a considerable variety in medical practices, either going by the patient's place of residency or the 66 involved hospitals. The majority of these stays relate to an indication that is particularly vague, raising the suspicion that mainly healthy infants are being tested. A comparison to international data confirms the high utilisation of the PSG among infants in Belgium. Given the large population of infants who are tested annually it should not come as a surprise that about 40% of home monitored infants are term infants who did not experience an ALTE. CR events will frequently be observed on a PSG for many infants. The appearance of CR events on a PSG, however, is no precursor of the SIDS. Nevertheless, it will take considerable effort to reassure parents in this case and a fundamental question is raised about the eventual usefulness of this examination.

A disproportionate use of PSG testing will probably result into a higher utilisation of monitoring devices. As hard diagnostic parameters are lacking, we cannot determine the precise number of false-positives.

Our qualitative exploration suggests that Belgian health care professionals do not propagate the PSG as a screening instrument. However, inconsistencies emerge in the information transmitted to parents who are not always contradicted in their belief that a PSG test is a sound safety measure every parent should take.

Our research points to an inappropriate use of the PSG in Belgium at a considerable cost for examinations and hospital stays to the social security and the patients' parents in particular.

Key points

- **Both for polysomnographies and home monitoring in infants validated reference values are lacking.**
- **We did not find sufficient evidence-based findings to answer questions on the interaction between cardiorespiratory events, psychomotor development and the role of PSGs and home monitors.**
- **We did not find data to confirm (nor dismiss) the usefulness of the PSG in clarifying the underlying diagnosis with children who experienced an ALTE, recommendations.**
- **The usefulness of a PSG in infants suffering from other medical conditions (e.g. neuromuscular conditions, etc.) is generally accepted despite the fact that little evidence can be found.**
- **There are few economic evaluations of the cost-effectiveness of a PSG and home monitoring in prevention of a sudden unexplained infant death. The existing literature is of very low quality.**
- **Despite the decrease in the number of PSGs over the last years, polysomnographic examinations in infants are still widely performed in Belgium.**
- **There is a considerable variety in medical practices. The majority of PSG stays relate to an indication that is particularly vague, raising the suspicion that mainly healthy infants are being tested.**
- **A disproportionate use of PSG testing will probably result into a higher utilisation of monitoring devices.**
- **Belgian health care professionals do not seem to propagate the PSG as a screening instrument**
- **Parents are not always contradicted in their belief that a PSG test is a sound safety measure every parent should take.**
- **Our research points to an inappropriate use of the PSG in Belgium at a considerable cost to social security.**

9 RECOMMENDATIONS

9.1 POLYSOMNOGRAPHIES

In order to improve the appropriateness of polysomnographies performed in infants in Belgium, three complementary and optional strategies could be followed:

- Offering better information to the broad public, in particular young parents through authorized bodies, such as Kind/ONE and insurance funds;
    - The limitations of PSGs in prevention of SIDS could be mentioned explicitly in related documents and on websites.
    - Conflicting information could be avoided, e.g. the use of "wiegendoodtest" to designate a PSG in an infant could be banished and it could be envisaged that parental anxiety is no longer quoted as a sound reason for PSG testing.
- Reinforcing the role of health care workers and day time baby minders:
    - Physicians should provide correct information on the usefulness of PSG testing if the topic occurs. They could stress the general SIDS prevention measures to reassure anxious parents as these have been proven to be effective.
    - Instructions for relevant health care workers (maternity hospital nurses, daytime baby nurses, physiotherapists delivering ante and post-natal physiotherapy) could be stressed, conveying correct information on the usefulness of the PSG.
- Reforming the current regulation:
    - We estimated the number of hospitals performing PSGs in infants at 66 in 2004. Currently nineteen Belgian hospitals dispose of a neonatal intensive care unit (NICU). Ten out of these nineteen hospitals are also accredited centres for infant home monitoring of which there are twelve in total. Consequently, the combined numbers of hospitals with a NICU and accredited centres make up twenty-one hospitals in total. These twenty-one hospitals appear most qualified to perform polysomnographies in infants. As a consequence, it could be recommend to restrict the use of PSGs in infants to these hospitals. There are possible repercussions on the functioning and financing of the paediatric departments at the 45 hospitals that will no longer be allowed to perform PSGs in infants.
    - The quality of the paediatrician's analysis of paediatric PSG readings could be improved, e.g. by

making specific training mandatory for the entire sleep research team.

- An alternative or complementary recommendation is that parents whose infant is a candidate for a PSG are systematically referred to a paediatrician specialized in PSG assessment, who will decide on the actual prescription.

9.2 HOME MONITORING

The current regulation under which only monitors measuring both heart rhythm and respiration qualify for reimbursement is in line with the findings in the literature on the topic.

The question whether reimbursement should be limited to event-recording monitors is less easily answered. It is not clear to what extent all alarm reports from memory-enabled devices have to be analyzed by responsible physicians. Nor is it clear what would constitute the added value of repeated PSGs as compared to reports from memory-enabled devices.

We note that high PSG utilisation rates probably result into higher utilisation rates for home monitoring. If our recommendations on proper PSG utilisation prove to be successful, the number of infants under home monitoring is expected to decline accordingly.

9.3 DATA COLLECTION AND FURTHER RESEARCH

Questions about the sleeping environment of infants were included in the first (1997), but not the second (2001) nor third (2004) Health Interview Survey for Belgium. We recommend that future Health Interview Surveys systematically address this topic, so long term data on the prevalence of behavioural factors could be collected in order to assess the impact of prevention campaigning.

At present, the twelve accredited centres responsible for infant home monitoring are not required to report the average duration of monitoring split up per type of device (i.e. memory-enabled or not). They are, however, asked to report patient number both for January 1st of the present year and December 31st of the previous year, which seems to add little value for further analysis. We included a proposal to improve the current data collection in the appendix to this chapter. A further option is for the data collection with the twelve accredited centres is to keep a separate registry of home monitoring in infants with specific medical conditions (e.g. neuromuscular conditions). This way data on patients that generally are monitored for a considerably longer period van be analyzed separately.

Studies of good quality are lacking in the scientific literature, more specifically on the effect of CR events in infants on further psychomotor development. The set-up of such studies is recommendable.

Key points

- **The number of hospitals allowed to perform PSGs in an infant population could be reduced.**
- **The quality of the paediatrician's analysis of paediatric PSG readings could be improved.**
- **The number of physicians allowed to prescribe a PSG for infants could be limited.**
- **The topic of PSG testing could be mentioned explicitly in prevention messages and conflicting information could be avoided.**
- **Physicians could provide concrete information on the usefulness of PSG testing if the topic occurs.**
- **Instructions for relevant health care professionals could be stressed.**
- **A general debate could clarify to what extent alarm reports from memory-enabled devices have to be analyzed by responsible physicians.**
- **Future Health Interview Surveys could systematically address the topic of infant sleeping safety.**
- **The current data collection with the twelve accredited centres could be improved.**
- **Further prospective research is needed on the effect of CR events in infants on further psychomotor development.**

10 SCIENTIFIC VALIDATION

The members of the scientific validation committee unanimously confirmed that the report was scientifically valid provided certain improvements were made with respect to:

- General readability, requiring:
  - a list of abbreviations,
  - an introductory chapter,
  - a more understandable manner in presenting clinical evidence tables,
  - an English translation of the qualitative research protocol,
  - a list of contacted international experts.
- General content, requiring:
  - a more comprehensive discussion of the role bradycardia's play.
  - a more balanced approach of conflicting evidence in clinical literature in some cases.
  - a discussion on the prevalence of maternal smoking in Belgium.
  - a discussion on the ethical and societal implications of the report's findings.

Remarks on general readability were all taken into account for the final lay-out of the report. The suggested improvements for the report's content were also made with the understanding that ethical and societal implications were primarily discussed in the report's recommendations, which are the sole responsibility of the Belgian Health Care Knowledge Centre.

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I APPENDIX TO CHAPTER I

I.1 RIZIV-INAMI BILLING CODES FOR POLY(SOMNO)GRAPHIC INTERVENTIONS

RIZIV/INAMI Code[a]	Label_NL	Libellé_FR	Label_ENG (Translation suggested by KCE)[b]
474530	Verstrekkingen die tot het specialisme kindergeneeskunde (FJ) behoren : Neurodiagnostische polygrafie, verricht tijdens het eerste levensjaar, met gelijktijdig en continu registreren, gedurende ten minste twee uur, van minimum 6 derivaties (zoals hartfrequentie, ademhalingsbewegingen en -frequentie, E.M.G., E.O.G., E.E.G., druk in en/of pH van oesofagus of andere parameters) met protocol en uittreksels uit de tracés	Prestations relevant de la spécialité en pédiatrie (FJ) : Polygraphie de neuro-diagnostic faite pendant la première année de vie, avec enregistrement simultané et continu, pendant au moins deux heures, de minimum 6 dérivations (telles que fréquence cardiaque, mouvements et fréquence respiratoires, E.M.G., E.O.G., E.E.G., pression et/ou pH oesophagien ou autres paramètres) avec protocole et extraits de tracés	Paediatric Interventions: Neurodiagnostic polygraphy, during first life year, simultaneous and continuous recording for at least 2 hours of minimally 6 parameters (such as heart frequency, respiratory movements and respiratory frequency, E.M.G, E.O.G., E.E.G., oesophageal pressure and/or pH or other parameters) including protocol and print-outs of polysomnographic readings
474541	Verstrekkingen die tot het specialisme kindergeneeskunde (FJ) behoren : Neurodiagnostische polygrafie, verricht tijdens het eerste levensjaar, met gelijktijdig en continu registreren, gedurende ten minste twee uur, van minimum 6 derivaties (zoals hartfrequentie, ademhalingsbewegingen en -frequentie, E.M.G., E.O.G., E.E.G., druk in en/of pH van oesofagus of andere parameters) met protocol en uittreksels uit de tracés	Prestations relevant de la spécialité en pédiatrie (FJ) : Polygraphie de neuro-diagnostic faite pendant la première année de vie, avec enregistrement simultané et continu, pendant au moins deux heures, de minimum 6 dérivations (telles que fréquence cardiaque, mouvements et fréquence respiratoires, E.M.G., E.O.G., E.E.G., pression et/ou pH oesophagien ou autres paramètres) avec protocole et extraits de tracés	Paediatric Interventions: Neurodiagnostic polygraphy, during first life year, simultaneous and continuous recording for at least 2 hours of minimally 6 parameters (such as heart frequency, respiratory movements and respiratory frequency, E.M.G, E.O.G., E.E.G., oesophageal pressure and/or pH or other parameters) including protocol and print-outs of polysomnographic readings

[a] Billing codes of which the penultimate number is even or uneven refer respectively to inpatient or outpatient care,

[b] E.M.G: electromyography; E.O.G.: electro-oculogram; E.E.G. : electronecophalography

RIZIV/INAMI Code[a]	Label_NL	Libellé_FR	Label_ENG (Translation suggested by KCE)[b]
474552	Verstrekkingen die tot het specialisme kindergeneeskunde (FJ) behoren - Polysommetrisch onderzoek met eem minimumduur van zes uur met protocol en uittreksels uit de tracés : Continu en gelijktijdig registreren dat ten minste het E.E.G., het E.O.G., het E.C.G., de continue oxymetrie en twee ademhalingsparameters omvat	Prestations relevant de la spécialité en pédiatrie (FJ) - Examen polysomnographique d'une durée minimum de six heures avec protocole et extraits des tracés : Enregistrement continu et simultané comprenant au moins l'E.E.G., l'E.O.G., l'E.C.G., l'oxymétrie continue.et 2 paramètres respiratoires	Paediatric Interventions: polysomnographic examination with a duration of at least 6 hours, including protocol and print-outs of –at least- following polysomnographic readings: continuous and simultaneous recording of at least E.E.G, E.O.G. E.C.G., continuous oximetry and 2 respiratory parameters.
474563	Verstrekkingen die tot het specialisme kindergeneeskunde (FJ) behoren - Polysommetrisch onderzoek met een minimumduur van zes uur met protocol en uittreksels uit de tracés : Continu en gelijktijdig registreren dat ten minste het E.E.G., het E.O.G., het E.C.G., de continue oxymetrie en twee ademhalingsparameters omvat	Prestations relevant de la spécialité en pédiatrie (FJ) - Examen polysomnographique d'une durée minimum de six heures avec protocole et extraits des tracés : Enregistrement continu et simultané comprenant au moins l'E.E.G., l'E.O.G., l'E.C.G., l'oxymétrie continue.et 2 paramètres respiratoires	Paediatric Interventions: polysomnographic examination with a duration of at least 6 hours, including protocol and print-outs of–at least- following polysomnographic readings: continuous and simultaneous recording of at least E.E.G, E.O.G. E.C.G., continuous oximetry and 2 respiratory parameters.
477234	* Verstrekkingen die tot het specialisme neuropsychiatrie (FM) behoren : Polygrafie (elektro-encefalogram, elektrocardiogram, psychogalvanische weerstand) met verslag en uittreksel uit het tracé ten minste 6 gelijktijdige elektro-encefalografische derivaties	* Prestations relevant de la spécialité en neuropsychiatrie (FM) : Polygraphie (électroencéphalogramme, électrocardiogramme, résistance psycho-galvanique) avec rapport et extrait du tracé, 6 dérivations électroencéphalographiques simultanées au minimum	Neuropsychiatric Interventions: polygraphy (E.E.G., E.C.G., psycho-galvanic resistance) including a report and print-outs of the readings, simultaneous recording of minimally 6 electroencephalographic parameters
477245	* Verstrekkingen die tot het specialisme neuropsychiatrie (FM) behoren : Polygrafie (elektro-encefalogram, elektrocardiogram, psychogalvanische weerstand) met verslag en uittreksel uit het tracé ten minste 6 gelijktijdige elektro-encefalografische derivaties	* Prestations relevant de la spécialité en neuropsychiatrie (FM) : Polygraphie (électroencéphalogramme, électrocardiogramme, résistance psycho-galvanique) avec rapport et extrait du tracé, 6 dérivations électroencéphalographiques simultanées au minimum	Neuropsychiatric Interventions: polygraphy (E.E.G., E.C.G., psycho-galvanic resistance) including a report and print-outs of the readings, simultaneous recording of minimally 6 electroencephalographic parameters

RIZIV/INAMI Code[a]	Label_NL	Libellé_FR	Label_ENG (Translation suggested by KCE)[b]
477374	Verstrekkingen die tot het specialisme neuropsychiatrie (FM) behoren : Polysomnografisch onderzoek met een minimumduur van zes uur met protocol en uittreksels uit de tracés : Continu en gelijktijdig registreren dat ten minste het E.E.G., het E.O.G., het E.C.G., de continue oxymetrie en twee ademhalingsparameters omvat	Prestations relevant de la spécialité en neuropsychiatrie (FM) : Examen polysomnographique d'une durée minimum de six heures avec protocole et extraits des tracés : Enregistrement continu et simultané comprenant au moins l'E.E.G., l'E.O.G., l'E.C.G., l'oxymétrie continue et 2 paramètres respiratoires	Neuropsychiatric Interventions: polysomnographic examination with a duration of at least 6 hours, including protocol and print-outs of polysomnographic readings: continuous and simultaneous recording of at least E.E.G, E.O.G. E.C.G., continuous oximetry and 2 respiratory parameters.
477385	Verstrekkingen die tot het specialisme neuropsychiatrie (FM) behoren : Polysomnografisch onderzoek met een minimumduur van zes uur met protocol en uittreksels uit de tracés : Continu en gelijktijdig registreren dat ten minste het E.E.G., het E.O.G., het E.C.G., de continue oxymetrie en twee ademhalingsparameters omvat	Prestations relevant de la spécialité en neuropsychiatrie (FM) : Examen polysomnographique d'une durée minimum de six heures avec protocole et extraits des tracés : Enregistrement continu et simultané comprenant au moins l'E.E.G., l'E.O.G., l'E.C.G., l'oxymétrie continue et 2 paramètres respiratoires	Neuropsychiatric Interventions: polysomnographic examination with a duration of at least 6 hours, including protocol and print-outs of polysomnographic readings: continuous and simultaneous recording of at least E.E.G, E.O.G. E.C.G., continuous oximetry and 2 respiratory parameters.

1.2 RIZIV/INAMI BILLING CODES FOR HOME MONITORING DEVICES

RIZIV/INAMI Code	Label_NL	Libellé_FR	Label_ENG (translation suggested by KCE)
775250	Revalidatieovereenkomst inzake cardiorespiratoire monitoring thuis bij pasgeborenen en zuigelingen met verhoogd risico op plotse dood : cardiorespiratoir toezicht met een monitor met geheugen	Convention type de rééducation fonctionnelle relative au monitoring cardiorespiratoire à domicile de nouveau-nés et de nourrissons présentant un risque de mort subite : surveillance cardio-respiratoire avec un moniteur avec mémoire	[…] cardiorespiratory monitoring at home for neonates presenting a heightened risk for sudden death: cardiorespiratory surveillance by means of an event recording monitor
775272	Revalidatieovereenkomst inzake cardiorespiratoire monitoring thuis bij pasgeborenen en zuigelingen met verhoogd risico op plotse dood : cardiorespiratoir toezicht met een monitor zonder geheugen	Convention type de rééducation fonctionnelle relative au monitoring cardiorespiratoire à domicile de nouveau-nés et de nourrissons présentant un risque de mort subite : surveillance cardio-respiratoire avec un moniteur sans mémoire	[…] cardiorespiratory monitoring at home for neonates presenting a heightened risk for sudden death: cardiorespiratory surveillance by means of an monitor without event recording

2 APPENDIX TO CHAPTER III

HTA Polysomnography and Home Monitoring in Prevention of SIDS: Search April-May 2006

Note: How to read "Appendix to Chapter III"

2.1: First the databases searched and the search strategy are described, followed by the retained results.

2.2-2.3-2.4: Search Strategy and Number of Results: the first column gives the ID of the search, followed by the date and database searched. Then, the first "Keywords" are mentioned, and the number of resulting articles. If the number of results is considered to be too high to read all titles and abstracts, in the last column "not valid" is indicated. The second "Keywords" are then added to the first Keywords, and if necessary still a third "Keywords" is added. Finally, titles and abstracts of the search results are selected according to the Methodology explained in Chapter 3.2, and the number of relevant articles retained is mentioned in the last column.

2.5-2.6: Analysis of Results: first the search ID (see 2.1- 2.2- 2.3- 2.4) from which the result was retained is mentioned, then (if one search strategy yielded several articles) the ID of the result (article) under consideration is presented, followed by the reference. The next columns concentrate on the analysis of the references.

2.1 DATABASES FOR GUIDELINES: SEARCH STRATEGY DESCRIPTION AND NUMBER OF RESULTS

The following search strategy was developed:

A. Databases for Guidelines:

NeLH Guidelines Finder (UK)

National Guideline Clearinghouse (USA)

ANAES (France)

CMA Infobase (Canada)

CDC Recommends (Centre for Disease Control, USA)

Health Services /Technology Assessment Texts (HSTAT, USA)

ICES (Institute for Clinical Evaluative Sciences, Canada)

New Zealand Guidelines Group

Scottish Intercollegiate Guidelines Network (SIGN - UK)

National Institute for Clinical Excellence (NICE - UK)

Agency for Healthcare Research and Quality(AHRQ - USA)

Were searched for:

Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS

OR:

polysomnography or sleep monitoring or physiologic monitoring of sleep

All papers concerning polysomnography and/or sleep monitoring in infants or young children were retained.

6 results were retained after selection on title and abstract.

2.2 GUIDELINES RESULTS

ID Search	Source	ID Results	Results	Author	Reference	Publication Date	Topic
PSG-G-1a	NELH Guidelines Finder		Reduce the risk of cot death: an easy guide	Publisher: Foundation for Study of Infant Deaths (UK)	NLH	12/05/2004	SIDS-prevention (no references)
PSG-G-2a	NGC	PSG-G-2a-1	Apnea, sudden infant death syndrome, and home monitoring.	American Academy of Pediatrics	Pediatrics 2003 Apr;111(4 Pt 1):914-7.	2003	Home monitoring (SIDS- ALTE-other infants), short note on polysomnography (SIDS- ALTE)
		PSG-G-2a-2	The changing concept of sudden infant death syndrome: diagnostic coding shifts, controversies regarding the sleeping environment, and new variables to consider in reducing risk.	American Academy of Pediatrics Task Force on Sudden Infant Death Syndrome	Pediatrics 2005 Nov;116(5):1245-55.	2005	SIDS-prevention; short note on homemonitoring (SIDS-ALTE-other infants)
PSG-G-4a	CMA Infobase	-	Guidelines for perinatal care : sudden infant death syndrome : newborn guideline 13, Mar 2001, British Columbia Reproductive Care Program	British Columbia Reproductive Care Program		13/03/2001	SIDS-prevention
		-	Reducing the Risk of Sudden Infant Death Syndrome in Canada	Canadian Foundation for the Study of Infant Deaths, the Canadian Institute of Child Health, the Canadian Paediatric Society and Health Canada	Paediatrics & Child Health 1999;4(3)223-4	revised 2001	SIDS-prevention
PSG-G-6a	Health Services Technology Assessment Texts (HSTAT)	PSG-G-6a-1	Infantile Apnea and Home Monitoring. NIH Consens Statement Online 1986 Sep 29-Oct 1;6(6):1-10. Pediatrics 1987;79(2): 292-299	panel of experts (no references)	HSTAT; Pediatrics 1987;79(2): 292-299	1986	Home monitoring (SIDS- ALTE- other infants); pneumogram (12 or 24h heart rate and thoracic impedance)

2.3 DATABASES FOR SYSTEMATIC REVIEWS: SEARCH STRATEGY DESCRIPTION AND NUMBER OF RESULTS.

ID Search	Date	Source	Keywords 1	Keywords 2	Number of results	Title/abstract
PSG-SR-1	30/04/2006	Cochrane DSR Full Range	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS		22	(no new results)
PSG-SR-2		-	ALTE or apparent life threatening event		0	0
PSG-SR-3		-	polysomnography		24	0
PSG-SR-4		-	sleep monitoring or physiologic monitoring of sleep		0	0
PSG-SR-5		-	apnea or sleep apnea, central or sleep apnea, obstructive sleep apnea syndrome		96	not valid
PSG-SR-6		-		Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	4	no new results
PSG-SR-7		-	respiratory rate or anoxia or hypoxia		246	not valid
PSG-SR-8		-		Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	1	no new results
PSG-SR-9		-	bradyarrhythmia or bradycardia or arrhythmia		237	not valid

ID Search	Date	Source	Keywords 1	Keywords 2	Number of results	Title/abstract
PSG-SR-10		-		Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	2	no new results
PSG-SR-11		-	electroencephalography		10	0
PSG-SR-12		-	electrocardiography or ECG		160	not valid
PSG-SR-13		-		newborn or infant	14	0
PSG-SR-14		-	electromyography or EMG		80	not valid
PSG-SR-15		-		newborn or infant	5	0
PSG-SR-16		-	electrooculography or eye movements		17	0
PSG-SR-17		-	oximetry or blood gas monitoring, transcutaneous or oximetry, pulse or transcutaneous oximetry or cutaneous oximetry		59	not valid
PSG-SR-18		-		newborn or infant	22	0
PSG-SR-19		DARE Full Range	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS		4	0
PSG-SR-20		-	ALTE or apparent life threatening event		0	0
PSG-SR-21		-	polysomnography		9	1 (Adults)
PSG-SR-22		-	sleep monitoring or physiologic monitoring of sleep		1	0
PSG-SR-23		-	apnea or sleep apnea, central or sleep apnea, obstructive sleep apnea syndrome		13	no new results
PSG-SR-24		-	respiratory rate or anoxia or hypoxia		28	0

ID Search	Date	Source	Keywords 1	Keywords 2	Number of results	Title/abstract
PSG-SR-25		-	bradyarrhythmia or bradycardia or arrhythmia		67	not valid
PSG-SR-26		-		newborn or infant	7	0
PSG-SR-27		-	electroencephalography or EEG		16	0
PSG-SR-28		-	electrocardiography or ECG		37	0
PSG-SR-29		-	electromyography or EMG		28	0
PSG-SR-30		-	electrooculography or eye movements		2	0
PSG-SR-31		-	oximetry or blood gas monitoring, transcutaneous or oximetry, pulse or transcutaneous oximetry or cutaneous oximetry		6	0
PSG-SR-32		ACP Journal Club Full Range	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS		3	1 new result
PSG-SR-33		-	ALTE or apparent life threatening event		0	0
PSG-SR-34		-	polysomnography		10	0
PSG-SR-35		-	sleep monitoring or physiologic monitoring of sleep		10	0
PSG-SR-36		-	apnea or sleep apnea, central or sleep apnea, obstructive sleep apnea syndrome		19	0
PSG-SR-37		-	respiratory rate or anoxia or hypoxia		44	not valid
PSG-SR-38		-		newborn or infant	3	0
PSG-SR-39		-	bradyarrhythmia or bradycardia or arrhythmia		111	not valid
PSG-SR-40		-		newborn or infant	2	0

ID Search	Date	Source	Keywords 1	Keywords 2	Number of results	Title/abstract
PSG-SR-41		-	electroencephalography or EEG		12	0
PSG-SR-42		-	electrocardiography or ECG		98	not valid
PSG-SR-43		-		newborn or infant	0	0
PSG-SR-44		-	electromyography or EMG		2	0
PSG-SR-45		-	electrooculography or eye movements		1	0
PSG-SR-46		-	oximetry or blood gas monitoring, transcutaneous or oximetry, pulse or transcutaneous oximetry or cutaneous oximetry		14	0
PSG-SR-47		CRD Databases (HTA) Full Range	Sudden Infant Death Syndrome or sudden infant death or SIDS		3	0
PSG-SR-48		-	cot death or crib death or SID		1	0
PSG-SR-49		-	ALTE or apparent life threatening event		525	not valid
PSG-SR-50		-		infant	10	0
PSG-SR-51		-	polysomnography		8	0
PSG-SR-52		-	sleep monitoring or physiologic monitoring of sleep		1	no new results
PSG-SR-53		-	apnea or sleep apnea, central or sleep apnea, obstructive sleep apnea syndrome		23	0
PSG-SR-54		-	electroencephalography or EEG	newborn or infant	0	0
PSG-SR-55		-	electrocardiography or ECG	newborn or infant	0	0
PSG-SR-56		-	electromyography or EMG	newborn or infant	0	0
PSG-SR-57		-	electrooculography or eye movements		0	0

ID Search	Date	Source	Keywords 1	Keywords 2	Number of results	Title/abstract
PSG-SR-58			oximetry or blood gas monitoring, transcutaneous or oximetry, pulse or transcutaneous oximetry or cutaneous oximetry		1	0

2.4 MEDLINE SEARCH STRATEGY AND NUMBER OF RESULTS

ID Search	Date	Source, Year Range	Keywords 1	Keywords 2	Keywords 3	Keywords 4	Results-Number	After Title & Abstract
PSG-PubMed-1	30/04/2006	Medline via Pubmed 1966-2006	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS				7236	Not Valid
PSG-PubMed-2		-	polysomnography or sleep monitoring or physiologic monitoring of sleep or sleep monitoring				9925	Not Valid
PSG-PubMed-3		-	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	polysomnography or sleep monitoring or physiologic monitoring of sleep or sleep monitoring			281	15
PSG-PubMed-4		-	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	apnea or sleep apnea, central or sleep apnea, obstructive sleep apnea syndrome or "respiratory rate" or anoxia or hypoxia			1271	Not Valid
PSG-PubMed-5		-			diagnosis		823	Not Valid
PSG-PubMed-6		-			predictive value of test		2	0
PSG-PubMed-7		-			diagnosis or diagnosis, differential or diagnostic test		823	Not Valid
PSG-PubMed-8		-				review	149	5
PSG-PubMed-9		-			diagnostic techniques		97	3
PSG-PubMed-		-			diagnosis, differential		23	0

ID Search	Date	Source, Year Range	Keywords 1	Keywords 2	Keywords 3	Keywords 4	Results-Number	After Title & Abstract
10								
PSG-PubMed-11		-	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	bradyarrhythmia or bradycardia or arrhythmia			350	Not Valid
PSG-PubMed-12		-			review		84	4
PSG-PubMed-13		-	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	electroencephalography			95	2
PSG-PubMed-14		-	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	electrocardiography			220	Not Valid
PSG-PubMed-15		-			review		27	2
PSG-PubMed-16		-	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	electromyography			43	0
PSG-PubMed-17		-	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	electrooculography or eye movements			27	0
PSG-PubMed-18		-	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	oximetry or blood gas monitoring, transcutaneous or oximetry, pulse or transcutaneous oximetry or cutaneous oximetry			37	1

ID Search	Date	Source, Year Range	Keywords 1	Keywords 2	Keywords 3	Keywords 4	Results-Number	After Title & Abstract
PSG-PubMed-19		-	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	follow-up study or longitudinal study or cohort study			442	Not Valid
PSG-PubMed-20					polysomnography		37	2
PSG-PubMed-21					sleep monitoring or physiologic monitoring of sleep or sleep monitoring		50	4
PSG-PubMed-22		-	apparent life threatening event or ALTE				374	Not Valid
PSG-PubMed-23		-		polysomnography or sleep monitoring or physiologic monitoring of sleep or sleep monitoring			46	1
PSG-PubMed-24		-		Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS			140	Not Valid
PSG-PubMed-25		-			polysomnography		21	1
PSG-PubMed-26		-			sleep monitoring or physiologic monitoring of sleep or sleep monitoring		33	0
PSG-PubMed-27		-		follow-up study or longitudinal study or cohort study			40	4

ID Search	Date	Source, Year Range	Keywords 1	Keywords 2	Keywords 3	Keywords 4	Results-Number	After Title & Abstract
PSG-PubMed-28		-	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	polysomnography or sleep monitoring or physiologic monitoring of sleep or sleep monitoring	practice guideline		1 (prevention)	0
PSG-PubMed-29		-			meta-analysis		0	0
PSG-PubMed-30		-			RCT		3	0

2.5 EMBASE SEARCH STRATEGY AND NUMBER OF RESULTS

ID Search	Date	Source-Limits	Keywords 1	Additional Keywords 2	Additional Keywords 3	Results-Number	After Title & Abstract *(other articles than search A-B-C)*
PSG-Embase-1	30/05/2006	Embase (2004-2006)- map to preferred term- explode- include as keyword	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS			1350	Not Valid
PSG-Embase-2		-	polysomnography or sleep monitoring or physiologic monitoring of sleep			1444	Not Valid
PSG-Embase-3		-	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	polysomnography or sleep monitoring or physiologic monitoring of sleep		32	0 (no new findings)
PSG-Embase-4		-		apnea or sleep apnea, central or sleep apnea, obstructive sleep apnea syndrome or "respiratory rate" or anoxia or hypoxia		90	1
PSG-Embase-5		-	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	bradyarrhythmia or bradycardia or arrhythmia		145	Not Valid
PSG-Embase-6		-			review	48	0
PSG-Embase-7		-	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	electrocardiography		25	0
PSG-Embase-8		-	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	electroencephalography		6	0
PSG-Embase-9		-	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	electromyography		5	0
PSG-Embase-10		-	Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or	electrooculography or eye movements		2	0

ID Search	Date	Source-Limits	Keywords 1	Additional Keywords 2	Additional Keywords 3	Results-Number	After Title & Abstract *(other articles than search A-B-C)*
			SIDS				
PSG-Embase-11			Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	oximetry or blood gas monitoring, transcutaneous or oximetry, pulse or transcutaneous oximetry or cutaneous oximetry		0	0
PSG-Embase-12			Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	follow-up study or longitudinal study or cohort study	polysomnography	1	0
PSG-Embase-13			Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	follow-up study or longitudinal study or cohort study	sleep monitoring or physiologic monitoring of sleep or sleep monitoring	1	0
PSG-Embase-14			apparent life threatening event or ALTE	polysomnography or sleep monitoring or physiologic monitoring of sleep or sleep monitoring		2	0
PSG-Embase-15				Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS		17	0
PSG-Embase-16				follow-up study or longitudinal study or cohort study		10	0
PSG-Embase-17			Sudden Infant Death Syndrome or cot death or crib death or sudden infant death or SID or SIDS	polysomnography or sleep monitoring or physiologic monitoring of sleep or sleep monitoring	practice guideline	3	0
PSG-Embase-18					meta-analysis	0	0
PSG-Embase-19			polysomnography or sleep monitoring or physiologic monitoring of sleep or sleep monitoring	RCT		1	0

2.6 GUIDELINES AND SYSTEMATIC REVIEWS: ANALYSIS OF RESULTS

Primary article ID		Title, Reference	Topic	References Primary article	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak	Message	Primary article: Recommendations	Level of Evidence (Fryback&Thornbury Levels) +:good test +-:moderate test -:weak test ?: no information
	PSG-G-2a-1:	Pediatrics 2003	Home monitoring (SIDS-ALTE-other infants), short note on polysomnography (SIDS- ALTE)				Home monitoring should not be prescribed to prevent SIDS. It may be warranted for premature infants with recurrent (extreme) events, but should be limited by age of 3 weeks postterm or cessation of the events. It may be warranted for infants with ALTE, tracheostomy, CPAP, chronic lung disease, or rare medical disorders affecting regulation of breathing. Monitors with event-recording are	Laboratory polysomnography: L1 technical efficacy:?; L2 diagnostic accuracy :weak test (low to moderate quality of evidence); L3-6:not valid
	GuidA	Apr;111(4 Pt 1):914-7.						Home monitoring:L1 technical efficacy:? ; L2 diagnostic accuracy:weak test (high quality of evidence) ; L3-6:not valid

Primary article ID		Title, Reference	Topic	References Primary article	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak	Message	Primary article: Recommendations	Level of Evidence (Fryback&Thornbury Levels) +:good test +-:moderate test -:weak test ?: no information
							advised; no current monitor detects obstructive apnea reliably; parents should be advised that monitoring has not been proven to prevent SIDS; parents should be encouraged to follow preventive measures (supine sleep position, safe sleep environment, no smoking)	
GuidA-ref1				Ramanathan R, Corwin MJ, Hunt CE, Lister G, Tinsley LR, Baird T, Silvestri JM et al. Cardiorespiratory events recorded on home monitors: Comparison of healthy infants with those at increased	Good: large prospective cohort study (N=1079).	home monitoring: extreme apnea/bradycardia exists in ALTE, SIDS-siblings, symptomatic and asymptomatic preterm children and healthy controls ALTE's and preterms show trend for increased rate, but		

Primary article ID		Title, Reference	Topic	References Primary article	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak	Message	Primary article: Recommendations	Level of Evidence (Fryback&Thornbury Levels) +:good test +-:moderate test -:weak test ?: no information
				risk for SIDS.JAMA. 2001 May 2;285(17):2199-207.		only in preterms this is statistically significant. This increase in preterms ceases at about 3 weeks postterm age whereas SIDS incidence is highest at 2-3 months.		
GuidA-ref2				Hoffman HJ, Damus K, Hillman L, Krongrad E.Risk factors for SIDS. Results of the National Institute of Child Health and Human Development SIDS Cooperative Epidemiological Study. Ann N Y Acad Sci. 1988;533:13-30.	Moderate: very large population-based retrospective case-control study (N=757 SIDS-cases, N controls=1514). Study included about 10% of total live-birth population of th USA during the inclusion period. (+-: partly based on interviews)	maternal interview, medical records: no association apnea of prematurity and SIDS		

Primary article ID		Title, Reference	Topic	References Primary article	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak	Message	Primary article: Recommendations	Level of Evidence (Fryback&Thornbury Levels) +:good test +-:moderate test -:weak test ?: no information
GuidA-ref3				Southall DP, Richards JM, Rhoden KJ, Alexander JR, Shinebourne EA, Arrowsmith WA, Cree JE, Fleming PJ, Goncalves A, Orme RL. Prolonged apnea and cardiac arrhythmias in infants discharged from neonatal intensive care units: failure to predict an increased risk for sudden infant death syndrome. Pediatrics. 1982 Dec;70(6):844-51.	Moderate: large prospective cohort study (N=1157): (+-: ancient registration methodology)	Laboratory pneumosomnography (breathing/ECG) has never been shown to predict SIDS (preterms and/or small for gestational age)- prolonged apnea more frequent in preterms		
GuidA-ref4				Southall DP, Richards JM, Stebbens V, Wilson AJ, Taylor V, Alexander JR. Cardiorespiratory function in 16 full-term infants with sudden infant death	Moderate: prospective cohort study (N=16, N controls=16): (+-: ancient registration methodology)	Laboratory pneumosomnography (breathing/ECG) has never been shown to predict SIDS (16SIDS- matched controls)		

Primary article ID		Title, Reference	Topic	References Primary article	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak	Message	Primary article: Recommendations	Level of Evidence (Fryback&Thornbury Levels) +:good test +-:moderate test -:weak test ?: no information
				syndrome. Pediatrics. 1986 Nov;78(5):787-96.				
GuidA-ref5				Monod N, Plouin P, Sternberg B, Peirano P, Pajot N, Flores R, Linnett S, Kastler B, Scavone C, Guidasci S. Are polygraphic and cardiopneumographic respiratory patterns useful tools for predicting the risk for sudden infant death syndrome? A 10-year study. Biol Neonate. 1986;50(3):147-53.	Weak: large retrospective case-control study (N=650; N=146; N controls=204) (-: only 1/5 SIDS had autopsy)	Laboratory polysomnography (PSG) has never been shown to predict SIDS (in SIDS-siblings and ALTE).		

Primary article ID		Title, Reference	Topic	References Primary article	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak	Message	Primary article: Recommendations	Level of Evidence (Fryback&Thornbury Levels) +:good test +-:moderate test -:weak test ?: no information
GuidA-ref6				Rosen CL, Frost JD Jr, Harrison GM. Infant apnea: polygraphic studies and follow-up monitoring. Pediatrics. 1983 May;71(5):731-6.	Weak: retrospective cohort study (N=26) (-: inconsistent methodology)	idem: Laboratory polysomnography (PSG) has never been shown to predict SIDS. (ALTE, 35% atypical cases)		
GuidA-ref7				MacKay M, Abreu e Silva FA, MacFadyen UM, Williams A, Simpson H. Home monitoring for central apnoea. Arch Dis Child. 1984 Feb;59(2):136-42.	Weak: retrospective cohort study (N=64)	Epidemiologic studies failed to document impact of home monitoring on incidence of SIDS.		
GuidA-ref8				Ward SL, Keens TG, Chan LS, Chipps BE, Carson SH, Deming DD, Krishna V, MacDonald HM, Martin GI, Meredith KS, et al. Sudden infant death syndrome in infants evaluated by apnea	Weak: retrospective cohort study (-: poor data collection)	idem: Epidemiologic studies failed to document impact of home monitoring on incidence of SIDS.		

Primary article ID		Title, Reference	Topic	References Primary article	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak	Message	Primary article: Recommendations	Level of Evidence (Fryback&Thornbury Levels) +:good test +-:moderate test -:weak test ?: no information
				programs in California. Pediatrics. 1986 Apr;77(4):451-8.				
GuidA-ref9				Hunt CE. Sudden infant death syndrome and other causes of infant mortality: diagnosis, mechanisms, and risk for recurrence in siblings. Am J Respir Crit Care Med. 2001 Aug 1;164(3):346-57.	Thorough review, including 2 large population based mortality studies (N=352.475 (Oyen N 1996); N=251.142 (Guntheroth WG 1990) of good quality; however no data on environmental risk factors included (+-: no search strategy included)	Situation in SIDS-siblings unclear: inconclusive evidence but genetic susceptibility to SIDS may exist, although the risk is most likely extremely low. Combined genetic-environmental influence most likely		
GuidA-ref10				statement, no evidence given	no references: no evidence	none of the current monitors detects obstructive apnea reliably		

Primary article ID		Title, Reference	Topic	References Primary article	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak	Message	Primary article: Recommendations	Level of Evidence (Fryback&Thornbury Levels) +:good test +-:moderate test -:weak test ?: no information
	PSG-G-6a-1	Infantile Apnea and Home Monitoring. NIH Consens Statement Online 1986 Sep 29-Oct 1;6(6):1-10. (HSTAT); Pediatrics 1987;79(2): 292-299	Home monitoring (SIDS-ALTE-other infants)-pneumogram (12 or 24h heart rate and thoracic impedance)	Expert Panel: no references	no references: no evidence	Apnea of prematurity:- risk factor for unexplained ALTE: no evidence- no risk factor for SIDS: evidence- subsequent morbidity: no evidenceALTE is a risk factor for sudden death or SIDS and may be associated with increased morbiditycurrently available home monitors (impedance): safe but many false alarms; a set of minimal standards for monitors is needed; "over-the-counter" monitors should be strongly discouragedHome	Pneumograms (12 or 24h heart rate and thoracic impedance) are not predictive for SIDS in normal children, ALTE, asymptomatic preterms, SIDS siblings. It might be helpful in clinical management. Cardiorespiratory monitoring not indicated for normal infants or asymptomatic preterms. Cardiorespiratory monitoring indicated for severe ALTE, symptomatic preterm infants, siblings of two or more SIDS victims, infants with certain conditions like	no evidence

Primary article				References Primary article	Evaluation (Dutch Cochrane Library Checklists)	Message	Primary article: Recommendations	Level of Evidence (Fryback&Thornbury Levels)
ID		Title, Reference	Topic		+: good +-: moderate -: weak			+:good test +-:moderate test -:weak test ?: no information
						monitoring:-might be effective for some cases of ALTE (case studies) -effect in SIDS-siblings: no studies available - effect in premature infants no studies available but might be alternative to prolonged hospitalization-other conditions e.g. bronchopulmonary dysplasia or tracheostomy may warrant home monitoring but studies are lacking-SIDS rates are not declined since introduce of home monitors and proportion of SIDS victims with history of apnea is small-no	central hypoventilation. Monitor discontinuation should be based on infant's condition, one or more normal pneumograms not necessary. Some literature to discontinue monitoring for ALTE after 2 or 3 months without alarms. Counseling necessary before starting monitoring at home since it might be stressful. Multidisciplinary approach advised. Prospective, well-designed studies warranted.	

Primary article				References Primary article	Evaluation (Dutch Cochrane Library Checklists)	Message	Primary article: Recommendations	Level of Evidence (Fryback&Thornbury Levels)
ID		Title, Reference	Topic					+:good test
					+: good			+-:moderate test
					+-: moderate			-:weak test
					-: weak			?: no information
						randomized studies concerning prevention of death by homemonitoring. (cont)		

Primary article ID		Title, Reference	Topic	References Primary article	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak	Message	Primary article: Recommendations	Level of Evidence (Fryback&Thornbury Levels) +:good test +-:moderate test -:weak test ?: no information
	PSG-SR-47-1	A review of guidelines for referral of patients to sleep laboratories. Hailey D, Tran K, Dales R, Mensinkai S, McGahan L. (CCOHTA) 2005 (Technology Report Issue 55):144	Polysomnography (SIDS-ALTE-other infants): review of evidence found in available guidelines		Review of Guidelines:1. Evidence table of references of other guidelines (Kerbl R 2000, Ipsiroglu OS 2000) and including Gordon D 1986: see CCOHTA 2005 report (appendix 14): 7 large prospective cohort studies (3 already mentioned: GuidA-ref3; GuidA-ref4; GuidA-ref5; others: Br Med J (Clin Res Ed) 1983;286(6371):1092-6; Waggener TB 1990; Kahn A 1988; Schechtman VL 1991)moderate quality evidence (mostly no blinded studies)2.Other guidelines: Kerbl R	1.PSG no routine screening tool in SIDS:moderate quality evidence2.PSG use supported in exploration of ALTE-symptoms with objective abnormality (abnormal blood gas...) or suspected disorders of respiratory control (e.g. OSA, obstructive sleep apnea like in certain syndromatic malformations):no references (no evidence)3. PSG no routine screening in uncomplicated ALTE:no references (no evidence)4. PSG indicated in neuromuscular	PSG no routine screening tool in SIDS or uncomplicated ALTE-PSG use supported in exploration of ALTE-symptoms with objective abnormality (abnormal blood gas...) or suspected disorders of respiratory control (e.g. OSA, obstructive sleep apnea)PSG use indicated in neuromuscular disease, some patients with bronchopulmonary dysplasia, cystic fibrosisRepeat PSG: infants with severe OSA,	Laboratory polysomnography: L1 technical efficacy: weak test (childhood); L2 diagnostic accuracy :weak test (moderate quality evidence in SIDS; no evidence in ALTE; low quality evidence in other infants); L3-6:not valid

Primary article				References Primary article	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak	Message	Primary article: Recommendations	Level of Evidence (Fryback&Thornbury Levels) +:good test +-:moderate test -:weak test ?: no information
ID		Title, Reference	Topic					
					2000, Ipsiroglu OS 2000, American Thoracic Society 1996: no references, but consistent with recommendations for older children and adults in CCOHTA 2005 report: no references (noevev.)evidence)3. Other guidelines: Kerbl R, Ipsiroglu OS, American Thoracic Society: 1 reference: Guilleminault C 1979 (see below): no references (no evidence)4. Other guideline: American Thoracic Society 1996:low quality references (case series or small case-control studies):low quality evidence5.	disease, some patients with bronchopulmonary dysplasia, cystic fibrosis...:low quality evidence5. repeat PSG: infants with severe OSA, neuromuscular disease, alveolar hypoventilation syndromeno references (no evidence)6. Lack of standardization of performance or interpretation of certain parameters of PSG and of some normative standards:low quality evidenceNote: Kerbl R: urgent question from parents can be indication for PSG 6. Lack of	neuromuscular disease, alveolar hypoventilation syndromeLack of standardization of performance or interpretation of certain parameters of PSG and of some normative standards	

Primary article ID		Title, Reference	Topic	References Primary article	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak	Message	Primary article: Recommendations	Level of Evidence (Fryback&Thornbury Levels) +:good test +-:moderate test -:weak test ?: no information
					Other guideline: American Thoracic Society 1996:no references (no evidence)6. 2 references (Bentele KH 1988, Hunt CE 1988) (see below); American Thoracic Society 1996: low quality references (case series or small case-control studies):low quality evidence low quality evidence	standardization of performance or interpretation of certain parameters of PSG and of some normative standards:low quality evidenceNote: Kerbl R: urgent question from parents can be indication for PSG		
GuidB-ref1				Kerbl R. [SIDS and polygraphy] Wien Klin Wochenschr. 2000 Mar 10;112(5):204-8.	Guideline: Evidence table of references: see CCOHTA 2005 report (appendix 14)	Conclusions: see CCOHTA 2005 report (appendix 14)		

Primary article ID		Title, Reference	Topic	References Primary article	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak	Message	Primary article: Recommendations	Level of Evidence (Fryback&Thornbury Levels) +:good test +-:moderate test -:weak test ?: no information
GuidB-ref2				Ipsiroglu OS, Kerbl R, Urschitz M, Kurz R. [4th Austrian SIDS Consensus-Consultation and the Viennese SIDS prevention campaign "Secure Sleep"] Wien Klin Wochenschr. 2000 Mar 10;112(5):187-92.	Guideline: Evidence table of references: see CCOHTA 2005 report (appendix 14)	Conclusions: see CCOHTA 2005 report (appendix 14)		
GuidB-ref3				Standards and indications for cardiopulmonary sleep studies in children. American Thoracic Society. Am J Respir Crit Care Med. 1996 Feb;153(2):866-78. [No authors listed]	Guideline: references cite background clinical issues and some low quality evidence (case-series or small case-control studies): see CCOHTA 2005 report (appendix 12)	Conclusions: see CCOHTA 2005 report (appendix 12)		

Primary article ID		Title, Reference	Topic	References Primary article	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak	Message	Primary article: Recommendations	Level of Evidence (Fryback&Thornbury Levels) +:good test +-:moderate test -:weak test ?: no information
GuidB-ref4				Gordon D, Southall DP, Kelly DH, Wilson A, Akselrod S, Richards J, Kenet B, Kenet R, Cohen RJ, Shannon DC. Analysis of heart rate and respiratory patterns in sudden infant death syndrome victims and control infants. Pediatr Res. 1986 Jul;20(7):680-4.	Good: blinded Prospective cohort study (N SIDs =10, N controls=100) [2]	In-depth spectral analysis of cardiorespiratory data can't distinguish future SIDS from normal control infants		
GuidB-ref5				Guilleminault C, Ariagno R, Korobkin R, Nagel L, Baldwin R, Coons S, Owen M.Mixed and obstructive sleep apnea and near miss for sudden infant death syndrome: 2. Comparison of near miss and normal control infants by	Moderate: Prospective cohort study (N Alte =29, N controls=30) (+-: no information on blinding of investigator)	PSG in ALTE and normal control infants shows more mixed and obstructive apnea's (more than 3 sec) during sleep in ALTE		

Primary article ID		Title, Reference	Topic	References Primary article	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak	Message	Primary article: Recommendations	Level of Evidence (Fryback&Thornbury Levels) +:good test +-:moderate test -:weak test ?: no information
				age.Pediatrics. 1979 Dec;64(6):882-91.				
GuidB-ref6				Bentele KH, Albani M.Are there tests predictive for prolonged apnoea and SIDS? A review ofepidemiological and functional studies.Acta Paediatr Scand Suppl. 1988;342:1-21.	Low: thorough Review with 91 references but no search strategy included and quality of included studies mostly low.	Epidemiologic studies show an increased SIDS risk in SIDSsiblings, ALTE, preterms, infants of drug dependent mothers. Pneumocardiography or other polysomnographic methods are not able to predict the risk of ALTE or SIDS, probably due to lack of standardisation of methods.		

Primary article ID		Title, Reference	Topic	References Primary article	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak	Message	Primary article: Recommendations	Level of Evidence (Fryback&Thornbury Levels) +:good test +-:moderate test -:weak test ?: no information
GuidB-ref7				Hunt CE, Brouillette RT, Hanson D.Apnea-onset definition significantly affects pneumogram results.Sleep. 1988 Jun;11(3):286-90.	Low: controlled trial (-:weak description of methodology)	Scoring of amount of apnea's on 40 pneumograms yields significant different result according to the definition use to define apnea (timing of onset…)		
	PSG-SR-32-1	Sleep apnea syndrome: polysomnography in preterm infants.Comite d'Evaluation et de Diffusion des Innovations Technologiques (CEDIT) 1999.	research publication: comparison of average apnea frequency on polysomnography, polygraphy and monitoring of 77 preterms (born before 34 weeks postmenstrual age); the 3 techniques were done at the same time on the same infant	Comite d'Evaluation et de Diffusion des Innovations Technologiques (CEDIT) 1999.	Moderate: observational study (+-: blinded study but each of the 3 modalities was evaluated by another resaercher)	In preterms (born before 34 weeks postmenstrual age), average apnea frequency (more than 10 sec) is more than twice as high on polysomnography than on polygraphy (only heart rate and breathing) and monitoring; for all three modalities two thirds of alarms was false. PSG preferred technique to evaluate certain preterms (severe ill babies,		

Primary article				References Primary article	Evaluation (Dutch Cochrane Library Checklists)	Message	Primary article: Recommendations	Level of Evidence (Fryback&Thornbury Levels)
ID		Title, Reference	Topic					+:good test
					+: good			+-:moderate test
					+-: moderate			-:weak test
					-: weak			?: no information
						evaluation of certain treatments, in order to discharge early from the hospital, SIDSsiblings)		

2.7 MEDLINE AND EMBASE: ANALYSIS OF RESULTS

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
PSG-PubMed-3	PSG-PM3-1	Schluter B, Buschatz D, Trowitzsch E. [Apnea characteristics of children who later died: comparison of sudden infant death with other causes of death.]Wien Med Wochenschr. 1996;146(13-14):321-3.	apnea on PSG in future SIDS-victims	Apnea's of more than 2 sec: obstructive and central apnea's on PSG significantly more frequent in 7 future SIDS victims compared to 13 infants who died from other causes and 40 controls; but large and overlapping ranges between groups	Weak: prospective cohort study in future SIDS-victims (N=7, N controls=40, N infants who died from other causes=13) (-:low number of SIDS, use of different standards compared to most other studies, no information on blinding of evaluator)
	PSG-PM3-2	de Nardi S, Paditz E, Erler T, Gruntzke A. [Reliability of home monitoring with event-recording compared with polysomnography in infants] Wien Klin Wochenschr. 2003 Jul 15;115(12):421-8.	Homemonitoring: reliability	Described type of home monitor (VitaGuard3000) has insufficient sensitivity as compared to PSG: central apnea's (more than 8 sec) and tachycardia's (more than 200/min) on VitaGuard3000 far less common than on PSG, and many false alarms.	Moderate: simultaneous cross-sectional evaluation of monitor and PSG in 20 infants (+-:no information on blinding of evaluator)
	PSG-PM3-3	Daniels H, Naulaers G, Deroost F, Devlieger H. Polysomnography and home documented monitoring of cardiorespiratory pattern. Arch Dis Child. 1999 Nov;81(5):434-6	PSG and Home monitoring (prematures, ALTE)	study of ALTE with abnormal PSG (obstructive apnea more than 15 sec, bradycardia below 50/min) (N=11), and ALTE with normal PSG (N controls=27): PSG in ALTE predictive of later life-threatening events as documented by event-recording home monitor	Moderate: prospective cohort study in ALTE (N=11 with abnormal PSG and N=27 with normal PSG) (+-:small group, no information on blinding of evaluator)

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
	PSG-PM3-4	Franco P, Szliwowski H, Dramaix M, Kahn A.Decreased autonomic responses to obstructive sleep events in future victims of sudden infant death syndrome.Pediatr Res. 1999 Jul;46(1):33-9.	PSG: apnea in future SIDS-victims	Apnea's of more than 3 sec: obstructive and mixed apnea's on PSG significantly more frequent but central apnea's not in 18 future SIDS victims compared to controls	Good: prospective cohort study in future SIDS-victims (N=18, N controls=30)
	PSG-PM3-5	Kato I, Franco P, Groswasser J, Scaillet S, Kelmanson I, Togari H, Kahn A. Incomplete arousal processes in infants who were victims of sudden death. Am J Respir Crit Care Med. 2003 Dec 1;168(11):1298-303.	PSG: arousal in future SIDS-victims	Cortical arousal (EEG-alteration) less frequent but subcortical arousal (movement, heart-rate change, breathing-pattern change) more frequent in 16 future SIDS victims compared to controls	Good: prospective cohort study in future SIDS-victims (N=16, N controls=16)
	PSG-PM3-6	Crowell DH, Brooks LJ, Colton T, Corwin MJ, Hoppenbrouwers TT, Hunt CE, Kapuniai LE, Lister G, Neuman MR, Peucker M, Ward SL, Weese-Mayer DE, Willinger M. Infant polysomnography: reliability. Collaborative Home Infant Monitoring Evaluation (CHIME) Steering Committee. Sleep. 1997 Jul;20(7):553-60.	PSG: intra- and interraterreliability	Reliability (intra- and interrater) for evaluation of PSG in infants: kappa 0.45-0.58 (moderate)	PSG of 15 infants, 7 evaluators: good

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
	PSG-PM3-7	Freed GE, Meny R, Glomb WB, Hageman JR. Effect of home monitoring on a high-risk population. J Perinatol. 2002 Mar;22(2):165-7.	Monitoring: effectiveness	monitoring high-risk infants (preterms, SIDS-siblings, ALTE) reduces the rate of death to a rate comparable to the general population; in a group of 8998 infants 13 out of 14 deaths occurred while not on the monitor (average monitor compliance rate 82%); monitors can be a diagnostic tool; if preterm infants can be sent home from the hospital earlier, monitoring is largely cost-effective.	Weak: large retrospective cohort study (N=8998) in high-risk infants (-:inclusion or exclusion criteria not described, methodology of compliance measurement not described)
	PSG-PM3-8	Di Marco JN, Rimet Y, Poujol A, Cornus P, Mecheri S, Brusquet Y.[Value of the exploration by polysomnography, pHmetry and 24-hour ECG in the siblings of sudden death infants: 212 cases]Arch Pediatr. 1997 Oct;4(10):1019-20.	PSG: apnea in siblings of SIDS-victims	frequency of obstructive or mixed apnea (more than 3 seconds at least 3 times occurring) the same in 201 SIDS-siblings as reported in literature for general population	Weak: Case series (N=201)

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
	PSG-PM3-9	Weese-Mayer DE, Morrow AS, Conway LP, Brouillette RT, Silvestri JM. Assessing clinical significance of apnea exceeding fifteen seconds with event recording.J Pediatr. 1990 Oct;117(4):568-74.	Monitoring with event-recording: significance of prolonged apnea	Event-recording in 182 older infants (interquartile 2-18 months) showed 6% of true events (apnea longer than 15 sec, bradycardia age-related), 32% of false events and 62% movement-related or loose-lead events. Of the true events, 75% were isolated apnea's; only 3.6% were apnea's longer than 15 sec associated with bradycardia. Among 54 patients with apnea longer than 15 sec, monitoring could be discontinued in 50 based on the results of the event-recording. 30/54 were younger than 1 age, 29 of them were discontinued after 8 months (median) of monitoring. Isolated apnea's of 15-20 sec are no reason for prolonged monitoring in the described age-group, which included no prematures or infants with chronic lung disease.	Weak: Case series (N=182)(-: bias in inclusion criteria, i.e. patients who were already monitored and were referred for event-recording)

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
	PSG-PM3-10	Rowland TW, Donnelly JH, Landis JN, Lemoine ME, Sigelman DR, Tanella CJ.Infant home apnea monitoring. A five-year assessment.Clin Pediatr (Phila). 1987 Aug;26(8):383-7.	Laboratory evaluation (pneumogram) and home monitoring: correlation	Laboratory evaluation (pneumogram) and home monitoring: correlation is poor. In 110 ALTE, home monitoring was started on clinical basis (vigourous resuscitation). 15% suffered recurrence of the initial event, detected by home monitoring; correlation with initial laboratory evaluation was poor. Also for 45 SIDS-siblings correlation between initial laboratory evaluation and further events was poor. Of 34 infants with apnea of prematurity (persisting on laboratory evaluation despite theophylline) and of 22 infants with bronchopulmonary or other chronic disease, one respectively none experienced abnormal events while on monitoring.	Weak: case-series (N=211) (-: serious inclusion bias in home monitoring for "ALTE" and "bronchopulmonary or other chronic disease". Laboratory evaluation used old methodology)

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
	PSG-PM3-11	Oren J, Kelly D, Shannon DC.Identification of a high-risk group for sudden infant death syndrome among infants who were resuscitated for sleep apnea.Pediatrics. 1986 Apr;77(4):495-9.	ALTE: subgroups more likely to die	1153 infants monitored at home for unexplained ALTE were studied retrospectively. Of 76 infants with ALTE during sleep only responsive to mouth to mouth resuscitation, and without diagnosis after thorough investigation, 10 infants subsequently died. 8 infants were monitored at the time of death, 6 infants had autopsy. Of the 76 infants, 25 infants with recurrent idiopathic sleep onset apnea requiring vigourous resuscitation as well as 8 SIDS-siblings and 11 infants developing epilepsia had a higher risk of dying than the others (28% respectively 25% and 57% compared to 13% for the whole group)	Weak: retrospective cohort-study (N=76), inclusion criteria vague (-:no autopsy in 4 children, resuscitation based on parent report)
	PSG-PM3-12	Guilleminault C, Pelayo R, Leger D, Philip P. Apparent life-threatening events, facial dysmorphia and sleep-disordered breathing.Eur J Pediatr. 2000 Jun;159(6):444-9.	ALTE: subgroup with mild facial dysmorphia	A subgroup of ALTE presents sleep disordered-breathing associated with mild facial dysmorphia. This sleep disordered-breathing can be detected by PSG.	Good: prospective cohort study of ALTE (N=346) with 46 controls (definition of apnea clear but unconventional)

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
	PSG-PM3-13	Hoppenbrouwers T, Hodgman J, Arakawa K, Sterman MB.Polysomnographic sleep and waking states are similar in subsequent siblings of SIDS and control infants during the first six months of life. Sleep. 1989 Jun;12(3):265-76.	PSG in SIDS siblings	Polysomnographic sleep and waking states are similar in 25 SIDS-siblings compared to 25 controls	Weak: prospective cohort study in SIDS-siblings (N=25) compared with controls (N=25) (+-: no information on blinding of evaluator; no information on diagnosis of SIDS (autopsy), movement detected by sensor on mattress)

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
	PSG-PM3-14	Kahn A, Blum D, Montauk L. Polysomnographic studies and home monitoring of siblings of SIDS victims and of infants with no family history of sudden infant death.Eur J Pediatr. 1986 Oct;145(5):351-6.	PSG and monitoring in asymptomatic infants and SIDS-siblings	Systematic PSG was performed at 8 weeks of age in 2735 infants without and 923 infants with a sibling who died from SIDS. PSG was considered abnormal if values differed from the 95 centiles for 110 normal term infants of the same age were found. PSG was abnormal in 937 infants (509 infants without and 428 infants with a sibling who died from SIDS), a second PSG was proposed at 12 weeks. One SIDS-sibling for which the second PSG was refused died of SIDS. Of the 891 second PSGs performed, 153 were still abnormal, and 150 infants (55 SIDS-siblings or 6% of the initial group; and 95 other infants or 4%) were home monitored; none of them died. Of the three monitors refused, two infants without family history of SIDS died. Of the infants not monitored, 3 SIDS-siblings and 1other infant died of SIDS. Normal PSG does not exclude SIDS, whereas monitoring might prevent SIDS in infants considered to be at risk. SIDS-siblings present a higher frequency of PSG abnormalities at 8 weeks but not at 12 weeks of age.	Moderate: prospective cohort study (N total=3658) in infants with and without a sibling who died from SIDS (+-: no mentioning of autopsy was performed in SIDS-cases)

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
	PSG-PM3-15 (see also GuidA-ref9)	Hunt CE. Sudden infant death syndrome and other causes of infant mortality: diagnosis, mechanisms, and risk for recurrence in siblings. Am J Respir Crit Care Med. 2001 Aug 1;164(3):346-57.	Literature review: Risk of SIDS in siblings of SIDS-victims	Relative risk for recurrent SIDS in SIDS-siblings is the same as relative risk for recurrence of non-SIDS infant mortality. However, not all studies include data of autopsy; and even in autopsy it is difficult to recognize intended suffocation. Also, in most studies no data on epidemiologic factors associated with SIDS are included. A combined genetic-environmental influence is most likely. For neuropathologic studies in SIDS victims and physiological studies in SIDS-siblings, the causal relationship of those anomalies to the pathophysiology of SIDS is unknown. Due to the low incidence rate of SIDS, an estimated increased relative risk of 5 for a subsequent sibling leads to approximately 1% of SIDS-siblings eventually dying due to SIDS. This percentage may even be less, depending on environmental factors	Moderate: thorough literature review (143 references) but search strategy not mentioned
PSG-PubMed-8	PSG-PM8-1	Lequien P, Carpentier C. [Prematurity and sudden infant death syndrome. Polysomnography in question] Arch Pediatr. 1999 Jun;6(6):683-5.	Literature review: Risk factors SIDS; PSG	Increased risk of SIDS in prematures below 28 weeks PMA is evident; but is not proven in bronchopulmonary dysplasia (BPD). Evaluation of oxygen saturation and eventually oxygen supplements are indicated in BPD. PSG no tool to predict SIDS or ALTE	Weak: review of 16 references, search strategy not mentioned

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
	PSG-PM8-2	Kahn A, Sawaguchi T, Sawaguchi A, Groswasser J, Franco P, Scaillet S, Kelmanson I, Dan B. Sudden infant deaths: from epidemiology to physiology. Forensic Sci Int. 2002 Sep 14;130 Suppl:S8-20.	Laboratory Polysomnography (SIDS)	More obstructive and mixed apnea's in SIDS-victims (N= 40 SIDS, N control= 607) but apnea duration did not exceed 15 sec; greater cardiac sympathovagal balance (N=18 SIDS, Ncontrol=36) ;contribution of these findings for SIDS remains to be determined	Moderate: Review of large prospective cohort studies already published: more than 20 000 PSG (+-:combination of population of several studies not specified, use of different standards compared to most other studies)
	PSG-PM8-3	Ravet F, Francois G.[Follow-up of the premature infant: prevention of severe diseases and sudden death. Role of polysomnography] Arch Pediatr. 1998 Apr;5(4):435-41.	PSG in prematures	Prematures and low-birthweight infants are at increased risk of SIDS. They also exhibit an elevated number of apnea's. Especially very small prematures can exhibit apnea's on PSG which were not visible clinically. Henderson-Smart DJ (1983) showed an increase in conduction time for auditory evoked potentials in prematures with as compared to prematures without apnea's. Infants that exhibit respiratory symptoms or apnea's on the NICU, should have a PSG before they leave the hospital and go home with home monitoring; other infants with birthweight below 1700 gr should undergo PSG at 46-48 weeks PMA. However, PSG cannot predict SIDS.	Weak: literature review without mentioning search strategy; and expert opinions.

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
	PSG-PM8-4	Shoemaker M, Ellis M, Meadows S, Gannons M. Clinical inquiries. Should home apnea monitoring be recommended to prevent SIDS? J Fam Pract. 2004 May;53(5):418-9.	home monitoring in prematures	apnea of prematures has never been shown to be a risk factor for or a precursor to SIDS. Based on this literature evidence, home monitoring for the purpose of preventing SIDS cannot be recommended. Based on expert opinion, neonates with significant neurologic or pulmonary disease may benefit from home monitoring.	Weak: restricted literature review (9 references) without mentioning search strategy
	PSG-PM8-5	Poets CF. Apparent life-threatening events and sudden infant death on a monitor. Paediatr Respir Rev. 2004;5 Suppl A:S383-6.	home monitoring in ALTE by cardiorespiratory monitoring and oxymetry	In unexplained ALTE episodes of prolonged apnea or bradycardia are found in only a minority. Early detection of hypoxemia (on oxymetry) rather than of apnea or bradycardia might be more relevant. Relevance with regard to SIDS remains to be determined.	Weak: observational study; idiopathic ALTE: N=12 (-: small study)

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
PSG-PubMed-9	PSG-PM9-1	Cote A, Hum C, Brouillette RT, Themens M. Frequency and timing of recurrent events in infants using home cardiorespiratory monitors. J Pediatr. 1998 May;132(5):783-9.	Relationship PSG-home monitoring (ALTE, prematures, SIDS-siblings, anxious parents)	1.PSG in ALTE predictive of later life-threatening events as documented by event-recording home monitor- 2.For the whole group: 36% of infants had significant event recorded- bradycardia with irregular breathing movements (45% of events) more prevalent than other forms of bradycardia or central apnea; 87% of infants with significant events had first event in first month of monitoring; in the first month of recording significantly more events in prematures than in others; poor correlation between parental report of events and registrated events	Weak: 1. ALTE: retrospective case-control study (26 ALTE-cases with abnormal PSG, 47 ALTE-controls without abnormal PSG)-2. observational study (N=147) (-: retrospective, no blinding of investigator)
	PSG-PM9-2	Barrington KJ, Finer N, Li D. Predischarge respiratory recordings in very low birth weight newborn infants.J Pediatr. 1996 Dec;129(6):934-40.	PSG in very low birth weight infants (VLBW: BW less than 1251gr) at discharge NICU	1.Apnea (more than 12 sec) on PSG, mostly obstructive, occurs in 91% of 187 VLBW (including 79 with bronchopulmonary dysplasia) without clinical signs of apnea. No relationship between apnea's on PSG and subsequent ALTE as reported by parents. 2.If readmission after discharge because of respiratory infection (N=15), infants showing clinical apnea's at that time (N=8) had significantly more apnea's on PSG at discharge from NICU	1. Good: observational study (N=187) 2. Weak: prospective cohort study (N=15)(+-: no clear definition of apnea at time of respiratory infection, small numbers)

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
	PSG-PM9-3	Al-Nashash H, Lvov B. Sudden infant death syndrome detector.Technol Health Care. 1997 Dec;5(6):461-9.	New technological device to prevent SIDS	When cardiorespiratory pauze, infant receives mild push, if no reaction, audiovisual alarm is given. Tested in 6 students and 3 infants	Weak: small test sample (3 infants)
PSG-PubMed-12	PSG-PM12-1	Tester DJ, Ackerman MJ. Sudden infant death syndrome: how significant are the cardiac channelopathies?Cardiovasc Res. 2005 Aug 15;67(3):388-96.	heritable arrhythmia syndromes in SIDS	Review: heritable arrhythmia syndromes like Long QT syndrome (which counts several variants), Short QT syndrome, Brugada syndrome, caused by cardiac channel mutation accounts for about 5% of SIDS among whites (5 channel genes screened)	Moderate:1.Schwartz PJ 1998: large prospective cohort study of ECG in neonates (N=34.442; SIDS-cases 24) (+-: QT-length cut off of 440ms statistical, not absolute) Good: 2. Ackerman MJ 2003: genetic testing of 5 genes in 93 SIDS-victims
	PSG-PM12-2	Plant LD, Bowers PN, Liu Q, Morgan T, Zhang T, State MW, Chen W, Kittles RA, Goldstein SA.A common cardiac sodium channel variant associated with sudden infant death in African Americans, SCN5A S1103Y.J Clin Invest. 2006 Feb;116(2):430-5.	heritable arrhythmia syndromes in SIDS	1. population-based study of 133 autopsy SIDS-cases confirms 24-fold increase of homozygosy (3 cases) for the SCN5A mutation of a cardiac ion-channel gene in SIDS-cases 2. In vitro study of cells with the above mutation causes abnormal Natrium-flow only when the cell was exposed to acidosis	Good: population-based study autopsy SIDS-cases (N=133)

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
	PSG-PM12-3	Baird TM.Clinical correlates, natural history and outcome of neonatal apnoea.Semin Neonatol. 2004 Jun;9(3):205-11.	review: neonatal apnea, SIDS and home monitoring	Neonatal cardiorespiratory events occur in both preterm and full term infants and decrease over time; beyond the period of 3-4 weeks postterm age no difference exists in frequency between preterms and terms. If persisting events, preterms can be discharged home with home monitoring until 3-4 weeks postterm age. There is no direct relationship of these events to SIDS. Home monitoring did not decrease the incidence of SIDS. The relationship between neonatal apnea and developmental outcome is unclear.	Weak: literature review without mentioning search strategy.

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
	PSG-PM12-4	Fleming PJ, Blair PS.Sudden unexpected deaths after discharge from the neonatal intensive care unit.Semin Neonatol. 2003 Apr;8(2):159-67.	SIDS in preterms/dysmatures	Persistence of apnea/bradycardia in preterms/dysmatures is not associated with an increased risk of SIDS, and ceases spontaneously at the age of 2-3 monts postterm. Preterms (except BPD, bronchopulmonary dysplasia) can be discharged home without home monitor if they are 8 days free of apnea. General recommendations to prevent SIDS should be advised moreover since the potential benefit is proportionally greater than for term infants. The use of home monitors did not decrease the SIDS rate. Infants with BPD should have regular oximetry, and should be discharged with home oxygen if their baseline shows basic hypoxia.	Weak: literature review without mentioning search strategy.
PSG-PubMed-13	PSG-PM13-1	Hoppenbrouwers T, Hodgman JE, Arakawa K, Durand M, Cabal LA.Polygraphy after discharge in preterm infants with and without apnea in the nursery.Neuropediatrics. 1992 Apr;23(2):75-81.	PSG in former prematures with/without apnea	In otherwise healthy prematures with apnea's of prematurity (apnea of more than 20 sec at 32-36 weeks PMA) PSG was performed at 44 weeks which did not differ from controls without apnea's.	Moderate: case-control study (N=8 cases; N=9 controls) (+-:no report of blinding of investigators)

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
	PSG-PM13-2	Abreu e Silva FA, MacFadyen UM, Williams A, Simpson H.Sleep apnoea in infancy.J R Soc Med. 1985 Dec;78(12):1005-8.	Serial PSG in 33 mildly sick infants, 24 SIDS-siblings, 29 ALTE infants and 11 normal controls	Obstructive apnea mainly in mildly sick infants or ALTE, prolonged central apnea in some ALTE-infants	Weak: Case-control study (N total= 86)(-: no clear inclusion criteria, no age of infants at PSG, groups vary, no information on blinding)
PSG-PubMed-15	PSG-PM15-1	Skinner JR.Is there a relation between SIDS and long QT syndrome?Arch Dis Child. 2005 May;90(5):445-9.	Long QT-syndrome in SIDS	same references as Tester DJ 2005 (PSG-PM12-1)	See PSG-PM12-1
	PSG-PM15-2	Rahilly PM.Pneumographic studies: predictors of future apnoeas but not sudden infant death in asymptomatic infants.Aust Paediatr J. 1989 Aug;25(4):211-4.	Pneumocardiography in asymptomatic infants	In 401 asymptomatic infants, it was found that infants of anxious parents (N=26) had less abnormalities than siblings of SIDS (N=322). Abnormalities were defined for apnea's of more than 10 sec or bradycardia compared to average for age. However, all 8 infants who subsequently died, had normal pneumograms.	Moderate: Prospective cohort study (N=401) (+-: no blinding of investigator)

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
PSG-PubMed-18	PSG-PM18-1	Poets CF, Samuels MP, Southall DP. Epidemiology and pathophysiology of apnoea of prematurity. Biol Neonate. 1994;65(3-4):211-9.	Pneumography and oxymetry in preterms	A.16 preterms with inexplicable cyanosis were evaluated by PSG, apart from central or obstructive apnea's hypoxeamia without breathing abnormalities or bradycardia was noted as well. B.160 other preterms at time of discharge from NICU were evaluated for the frequency of apnea/desaturation. In 53 of them, apnea and desaturation at 2-7 weeks postterm age was compared to controls, no differences were observed; outlying values all normalized.	A.Weak: Small observational case-series B.Good: prospective cohort study (N=53)
PSG-PubMed-21	PSG-PM21-1	Oren J, Kelly DH, Shannon DC. Familial occurrence of sudden infant death syndrome and apnea of infancy. Pediatrics. 1987 Sep;80(3):355-8.	PSG and home monitoring in SIDS-siblings	in families with multiple SIDS, PSG not predictive of later SIDS; home monitor not protective	Weak: Prospective cohort study (N=73) (-: not always autopsy to diagnose SIDS)
	PSG-PM21-2	Oren J, Kelly DH, Shannon DC. Pneumogram recordings in infants resuscitated for apnea of infancy. Pediatrics. 1989 Mar;83(3):364-8.	PSG, home monitor in ALTE	PSG in ALTE not predictive of later SIDS; home monitor does not prevent SIDS	Weak: Prospective cohort study (N=51) (-: small groups, no clear matching for 10 infants)
	PSG-PM21-3	Kahn A, Sottiaux M, Appelboom-Fondu J, Blum D, Rebuffat E, Levitt J.Long-term development of children monitored as infants for an apparent life-threatening event during sleep: a 10-year follow-up study.Pediatrics. 1989 May;83(5):668-73.	Long Term Development after ALTE	At 7 years, children that as an infant suffered from ALTE (for which 8/26 needed resuscitation), did not show significant differences as compared with normal controls regarding behaviour or IQ.	Good: observational study (N=26)

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
	PSG-PM21-4	Keens TG, Ward SL, Gates EP, Hart LD, Basile A, Tinajero LA, Lau AM, Chan CE.A comparison of pneumogram recordings in infants in the hospital and at home.Pediatr Pulmonol. 1986 Nov-Dec;2(6):373-7.	Validity of pneumocardiogram	No difference between pneumocardiogram in hospital and at home for infants with preterm or infancy apnea and SIDS-siblings	Moderate: case-control study (N=64) (+-: no information regarding blinding of investigator)
PSG-PubMed-23	PSG-PM23-1	Rahilly PM.The pneumographic and medical investigation of infants suffering apparent life threatening episodes.J Paediatr Child Health. 1991 Dec;27(6):349-53.	Pneumocardiography in ALTE: predictive of future events	340 ALTE, of which 51 without known etiology (17 infants with airway pathology), 135 infants without airway pathology were sent home with home monitoring. Of these, infants with normal pneumocardiography (N=109) in 27 cases had apnea's with reflux whereas infants with abnormal pneumocardiography (N=26) in 20 cases had apnea's that were always without reflux. Eye-ball pressure in 65 infants with white apnea at first presentation had a sensitivity of 81% and a specificity of 74% in predicting further white events. Pneumocardiography can predict future central apnea's in ALTE	Weak: case-control study (N total= 340)(-: no clear inclusion criteria and mixing of groups with different pathologies)

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
PSG-PubMed-25	PSG-PM25-1	Crowell DH, Kulp TD, Kapuniai LE, Hunt CE, Brooks LJ, Weese-Mayer DE, Silvestri J, Ward SD, Corwin M, Tinsley L, Peucker M; CHIME Study Group.Infant polysomnography: reliability and validity of infant arousal assessment.J Clin Neurophysiol. 2002 Oct;19(5):469-83.	PSG: validity	Reliability (intra- and interrater kappa with 95% confidence interval) for evaluation of PSG sleep stages in infants: kappa moderate before, excellent after training of evaluators	Good: validity study
PSG-PubMed-27	PSG-PM27-1	Baroni MA.Apparent life-threatening events during infancy: a follow-up study of subsequent growth and development. J Dev Behav Pediatr. 1991 Jun;12(3):154-61.	Development at follow-up after ALTE	51 ALTE, of which 30 without specific etiology, were home-monitired and evaluated at 10-14 months (retrospective birth chart evaluation, growth, Bayley Scales of Infant Development, grading of home environment (HOME Scale), apnea on pneumocardiography immediately after ALTE): apnea negative correlation with all other outcome measures, including developmental outcome. This remained significant even after correction for gestational age and birth complications.	Weak: Retrospective cohort study (N=51) (-: mixed etiological group)

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
	PSG-PM27-2	Milioti S, Einspieler C.The long-term outcome of infantile apparent life-threatening event (ALTE): a follow-up study until midpuberty.Neuropediatrics. 2005 Feb;36(1):1-5.	Development at follow-up after ALTE	14 unexplained ALTE and 10 controls were followed until puberty. Alte's had more minor neurological abnormalities like associated movements, but motor performance test and behaviour checklist were not different from controls. One ALTE had severe motor and learning disabilities	Moderate: Prospective cohort study (N cases= 14, N controls= 10)(+-: no information on blinding of assessors)

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
	PSG-PM27-3	Kahn A; European Society for the Study and Prevention of Infant Death.Recommended clinical evaluation of infants with an apparent life-threatening event. Consensus document of the European Society for the Study and Prevention of Infant Death, 2003.Eur J Pediatr. 2004 Feb;163(2):108-15.	ALTE: diagnostic investigation, home monitoring (review)	PSG is not required in most infants with ALTE. In some cases PSG has an added value to the diagnostic procedure. For monitoring, cardiorespiratory event recorders are preferred as they contribute to the significance of the reported events. None of the currently available home memory monitors will reliably detect obstructive apnea's. Oxygenation monitors have been recommended as alternative. There are at present no universally accepted criteria to determine which infant should be monitored at home. The effectiveness of home monitoring in reducing the rate of SIDS has not been established for any risk group, including preterms, SIDSsiblings and ALTE. When the decision is taken to monitor an infant, continuous medical, psychological and technical support should be available, including staff always being available for direct or telephone consult (expert opinion).	Moderate: extensive literature review (55 references) (+-: no search strategy reported)

PubMed Search ID	PubMed article ID	PubMed article: Title, Reference	PubMed article: Topic	PubMed article: Content	Evaluation (Dutch Cochrane Library Checklists) +: good +-: moderate -: weak
	PSG-PM27-4	McGovern MC, Smith MB.Causes of apparent life threatening events in infants: a systematic review.Arch Dis Child. 2004 Nov;89(11):1043-8.	systematic review on causes of ALTE	Incidence of ALTE was 0.6-0.8% of all emergency visits of infants under 1 year, and 0.6/1000 live births; recurrences varied between 0 and 24%. Most common diagnoses were gastro-oesophageal reflux, followed by seizures and respiratory tract infection. 23% were idiopathic (no cause found). Death rate in the ALTE group was 8/1000, but some of these infants suffered from congenital metabolic disorders.	Moderate: literature review (+-: clear methodology but only non-randomized studies found), 8 studies retained after quality selection
	PSG-Embase-4	Hsu CH, Chow JC. Design and clinical monitoring of a newly developed non-attached infant apnea monitor. Biomedical Engineering-Applications, Basis and Communications 2005 17(3):126-134.	New technological device to prevent SIDS	New device detecting temperature changes by thermo sensors induced by breathing, without skin contact so avoiding skin irritations	Weak (-: only tested on 10 clinical cases)

3 APPENDIX TO CHAPTER IV

3.1 FILTERS AND DATABASES

Date of Search	Database	Platform/URL	Search Terms	Limitations	# Hits
June 15th 2006	PubMed	www.pubmed.gov	(PSG or polysomnography or somnography or (monitoring and sleep) or (monitoring and ambulatory)) AND (SID* or cot death or crib death or infant death) AND (Technology Assessment or cost*)	NA*	44
	Embase	Licensed product Embase		NA	34
	CRD (York University: DARE, NHS EED, HTA)	www.york.ac.uk	(PSG or polysomnography or somnography or (monitoring and sleep) or (monitoring and ambulatory)) AND (SID* or cot death or crib death or infant death)	NA	4
*"NA": no (other than default) limitations apply					

3.2 EVIDENCE SHEETS

3.2.1 Economic evaluations summary sheet: Waite 1993

Author	**Waite A., Emery J., Taylor E., Carpenter R., Gardner A.** [1]
Country	**United Kingdom**
Design	**Cost-"utility"("the parents expressed equal satisfaction"): multicentre comparison of two randomly assigned groups**
Perspective	Implicitly societal
Time window	Follow-up until 26th week or 2 months beyond the age of death of the sibling
Interventions	Not event-recording (?) monitor versus weighing scales NB: several types of monitors seem to be used.
Population	2 groups of each 50 families, corresponding to 1 baby (sibling) each
Assumptions	Not applicable (e.g. absence of stated protocol, no definition of monitor alarm, etc.)
Data source for costs	Not specified. The (few) presented valuations of cost items seem to refer to nation-wide (NHS-related?) costs.
Cost items included	Implicit costs for health services utilisation: Health Visitor, GP, Paediatrician, Hospital (days as an inpatient), Clinic (outpatient programs) Equipment Cost: 1) capital costs of the apnoea monitor and "other" monitor expenses: 6-month period 2) cost for electronic scales (period not clear)
Data source for outcomes	Primary research (survey was sent to parents and care-takers to assess patient (i.e. parent) satisfaction)
Discounting	Not applicable (short time frame)
Costs	Capital cost of the apnoea monitors: £300-£350 each, "other expenses depending on the type of monitor" of between £10 and £30 Electronic scales: "cost £180 each and require one battery every six months, which lasts several years" (sic) Other identified cost items ("professional time and bed use") remained unquantified
Outcomes	Parent "satisfaction": reported to be "equal", but not quantified, nor extensively elucidated.
Cost-effectiveness	"The cost of care for families allocated apnoea monitors was greater in professional time, bed use and equipment costs."
Sensitivity analysis	Not performed.
Conclusions	"The results show that both groups of parents seemed equally satisfied with their programme of support, and regularly weighing the infant does not appear to cause increased anxiety to parents or unnecessary use of services. The findings show that when the two groups are compared, the cost of care for families allocated apnoea monitors was greater in professional time, bed use and equipment cost."
Remarks	Interventions are performed with the sole purpose of offering mental support to parents. In fact, the parents are patients in this study. It seems odd that a study analyzing "comparative costs" does not undertake a detailed cost analysis for all explicitly stated cost items, nor calculates absolute and relative overall costs for respective interventions .As a result, sensitivity analyses cannot be performed. Furthermore, the main effect "satisfaction" remains unquantified or at least not specified in the paper. Several key elements remain unclear (monitors with or without event recording, life time of batteries…)

3.2.2 Economic evaluations summary sheet: Steinschneider 1995

Author	**Steinschneider A., Stantos V., Freed G.** [2]
Country	**United States**
Design	**Descriptive Cost Analysis (Comparative design)**
Perspective	Implicity societal, limited to direct monetary ("billed") costs
Time window	Primary data were obtained from 155 subjects during the first 20 weeks of life (no right censoring).
Interventions	Diagnostic intervention 1: home monitoring relying on parental reporting Diagnostic intervention 2: home monitoring relying on event recording
Population	155 subsequent sudden infant death syndrome siblings, based on preceding prospective research [3]
Assumptions	Definition of an episode: 1) Event-recorded protocol: >= 20 seconds per apnea AND >= 80 beats per minute (bpm) for bradycardia during at least 10 seconds 2) parental report protocol: report of an apnea alarm during sleep (as confirmed by event-recording of apnea) OR physiologic alarm associated with skin colour change or resuscitative intervention Clinical protocol: each infant is monitored until there are no "episodes" for 16 consecutive weeks. No diagnostic or screening tests are done before monitoring starts. If an episode occurred during the first 16 weeks, a 12-channel PSG would be performed. No child receives more than 1 PSG. Repeat episodes would, however, lengthen the home monitoring period. No other diagnostic interventions would stem from confirmed episodes.
Data source for costs	Based on fees structure at the American SIDS institute
Cost items included	4-week rental fee for home monitor: 350$ Interpreting waveforms during 4-week period: 180$ Administering and interpreting a polysomnogram: 600$
Data source for outcomes	Primary research (Steinschneider, Stantos, 1991)
Discounting	NA (limited time frame)
Costs	Cf. supra
Outcomes	Successful SID prevention (rationale: "although the National Institute of Health Consensus Development Report of 1987 questioned the necessity of monitoring subsequent siblings of SIDS victims, several research studies have demonstrated that this group is at increased risk to die of SIDS, and many practitioners choose to monitor these infants.")
Cost-~~effectiveness~~	Median per patient cost of home monitoring, exclusive of polysomnogram cost: 1.978$ (parental reporting) versus 2.120$ (event recording): difference not statistically significant (Wilcoxon's T, non-parametric test) Median per patient cost of home monitoring inclusive of polysomnogram cost: 2.578$ (parental reporting) versus 2.120$ (event recording): difference statistically significant (Wilcoxon's T, non-parametiric test)
Sensitivity analysis	Not performed: "Undoubtedly, employing other criteria would have changed the numerical data, but it is unlikely that the fundamental conclusion of this study would be altered." ("criteria" refers to cut-off values defining medically significant episodes).
Conclusions	"[…] incorporating event recordings as an integral component of home monitoring resulted in no significant difference in the average per patient treatment cost even when excluding the cost of any diagnostic tests. Adding the cost of only one diagnostic test resulted in a significantly lower per patient treatment cost when document monitoring was employed"
Remarks	Sensitivity analysis should have been performed regarding cut-off values for medically significant episodes given the lack of consensus on appropriate thresholds values. Targeted outcome is only briefly hinted at ("monitoring for monitoring' s sake" it would almost seem) It seems likely other diagnostic tests would result from observed episodes. These additional tests would only further corroborate the cost-effectiveness analysis (add extra cost to the parental report protocol). Would be interesting to compare with current "protocols" in Belgium: established criteria + current "algorithm" (for siblings): polysomnography – monitoring – further polysomnographies?

3.2.3 Economic evaluations summary sheet: Zupancic 2000

Author	**Zupancic J., Triedman J., Alexander M., Walsh E., Richardson K.,e.a. [4]**
Country	**Canada/United States**
Design	**Incremental Cost analysis comprising decision analytic (Markov state) modelling**
Perspective	Societal, limited to direct and monetary ("billed") costs
Time window	Maximum life span of subjects in the cohort considered: 75 years of age.
Interventions	Baseline, "no screening": Promotion of supine/side-sleeping, pediatric guidance and health maintenance counselling. Comparator 1 "Universal Screening" 12-lead ECG on third day of life, if QTc interval > 440 millisec → repeat ECG and cardiology consultation. All children testing positive receive medication, monitoring and follow-up during first year. Comparator 2 "High-Risk Screening" Same screening algorithm as described for first comparator, but limited to hypothetical portion (high risk neonates) of population (odds-ratio SID = 5).
Population	Hypothetical cohort of healthy term infants: (incremental cost is derived per single live birth) Based on primary data by Schwartz [5] , 1998 (covering 34.442 Italian children between 1976 and 1994)
Assumptions	Pathophysiologic assumptions are presented and calculated as three separate scenarios: 1) "SIDS/LQTS": screening finds infants with LQTS, antiadrenergic therapy and monitoring constitute an effective treatment 2) "SIDS": screening finds children with developmental cardiac instability not intrinsically related to LQTS (temporary symptoms), antiadrenergic therapy and monitoring constitute an effective treatment 3) "Autonomic": screening would find autonomic dysregulation linked to brain stem development. SIDS would hence occur through non-cardiac events and therefore antiadrenergic therapy yields no beneficial effect. Decision analytic model: Time horizon of 75 years and three states: death, LQTS without pacemaker and LQTS with pacemaker. annual cycles are run according to state transitions probabilities derived from two prospective studies covering both general and paediatric populations. Successful identification of high-risk neonates is feasible. Successful treatment by means of bètablockers and/or home monitoring is possible for non-autonomous (brain stem) aetiology.
Data source for costs	Unit costs and professional fees: actual data for Children's Hospital, Boston Medicare charge-center-specific costs-to-charges ratios Going market prices for other services and goods.
Cost items included	Unit costs Market prices for professional services Hospital stay costs Drugs (Bèta-blockers, …) Home monitoring Current and future direct medical costs related to either SIDS or to LQTS, such as adverse events related to Bètablocker-treatment
Data source for outcomes	Sensitivity and specificity of neonatal ECG screening were calculated based on previous prospective research [5]
Discounting	Annual discount rate for both costs and outcomes was set at 3%
Costs	Not explicitly stated per cost item. Baseline cost is estimated at 7,93$ per life birth. The cost for an additional ECG can be derived: 90$ per ECG administered to a

Author	**Zupancic J., Triedman J., Alexander M., Walsh E., Richardson K.,e.a. [4]**
Country	**Canada/United States**
Design	**Incremental Cost analysis comprising decision analytic (Markov state) modelling**
	newly-born.
Outcomes	Outcomes regard lives saved, which are transformed into life years saved. Quality of life was not accounted for
Cost-effectiveness	Incremental CE (Base case): 1) SIDS/LQTS: High-risk screening: 3.403$ per Life Year saved Universal screening: 18.465$ per Life Year Saved 2) SIDS: High-risk screening: 3.016$ per Life Year saved Universal screening: 18.863$ per Life Year Saved 3) Autonomic: Universal screening: 309.527$ per Life Year Saved (should be considered as cost for preventing death by LQTS and not by SID)
Sensitivity analysis	Univariate analyses: 1) Cost items: no change in general conclusions 2) Higher QTc threshold value: cost-effectiveness deteriorating 3) Ability to identify high risk group: cost-effectiveness high risk screening deteriorating 4) SID incidence: broadly stable 5) Probability of death in LQTS: broadly stable 6) Adverse event rate bètablockers: broadly stable
Conclusions	"The results of our analysis are well within the range for other accepted health interventions such as hepatitis B immunization and neonatal intensive care, particularly if a high risk subgroup can be identified" → It is by no means clear how a subgroup would be identified.
	"The appealing cost-effectiveness ratios reported here are contingent upon the efficacy of bètablocker treatment for true-positive test results." → Specific source for assumption of efficacy not clearly referenced. → This parameter is not formally included in the sensitivity analyses (base case assumes 100% success rate), although the adverse event rate for Bètablockers is accounted for and clearly referenced and varied in a separate sensitivity analysis.
	"Until further longitudinal studies that clearly demonstrate the clinical value and statistical accuracy of GT prolongation as a marker of SIDS are performed, we believe that mass routine ECG screening of all neonates is not yet warranted."
	"[…] we may be reasonably confident that any pronouncement on the cost-effectiveness of screening for prolonged QT interval must await clarification of the underlying SIDS mechanism(s)."
Remarks	As ECG is a part of formal PSG-testing, this paper indirectly deals with our research scope. When focusing exclusively on QTc-screening, it should be clear that ECG-screening by definition would be more cost-effective than comprehensive PSG-screening. Evidently, we assume ECG characteristics to be identical.
	The hypothetical cohort is based on population traits in previous prospective research [5]. As a result, the paper deals with "healthy, term" children. Paradoxically, a key component in the model's build-up consists in the identification of high-risk neonates. This high-risk population would make up about 5% of the cohort. The real-life situation, however, does involve a considerable proportion of preterm infants as well, invalidating the cost-effectiveness analysis for wider real-life screening.
	The conclusions draw on a manifold of secondary data (transition probabilities) and their firmness is precluded by the uncertainty of key assumptions (e.g. identifying high-risk neo-nates and efficacy of SID-treatment).

4 APPENDIX TO CHAPTER V

4.1 DATA SOURCES

SOURCE	NAME	CONTENT	YEAR(S)/DATE DATA APPLY TO	TABLE	FIGURE
RIZIV/INAMI [16]	"N DOCUMENTS"	Billing codes for all Belgian residents, by financial year and actual year of intervention, covering reimbursed cases and amounts (€)	1995-2005	5	8,10,11
	"DOC HOSP"	Billing codes for all Belgian residents **by distinct hospitals**, by year of intervention, covering reimbursed cases and amounts (€)	1995-2003		See Appendix
	"ANNUAL REPORT CONVENTION FOR CARDIORESPIRATORY HOME MONITORING"	Patient numbers, number of monitored days, indication of patient subgroups and duration of monitoring. Base data are collected among the 12 acknowledged treatment centres and aggregate data tables are then enclosed to annual reports by RIZIV-INAMI	2003-2004	2	17,18
	NUMBER OF GENERAL PHYSICIANS AND PAEDIATRICIANS	Number of general physicians and paediatricians by contact address (RIZIV/INAMI related contact address)	2002	7	16
TCT [72] (TECHNISCHE CEL/CELLULE TECHNIQUE)	MKG-MFG/RCM-RFM STATISTICS	Data for Belgian hospital stays: (among others): ranked by main DRG and Degree of Severity: average Lenght of Stay, Average Overall Cost, Average Cost per Item (pharmaceuticals, physicians' fees, …)	2003	7	
BELGIAN MINISTRY OF HEALTH [83]	MKG/RCM STATISTICS	Belgian Database on Hospital Stays, containing medical - and other- patient data. Through ICD9 procedure codes 8917 ("polysomnographic hospital stays with sleep-related diagnostics were requested: frequencies (of hospital stays) by anonymized hospitals, by patients' place of residence, age and main DRG. and residency data.	2003,2004	7	11,12,13,14,15,19
	DATA ON NUMBER OF	Number of general physicians by region	31/12/2003		16 (cross-checking

SOURCE	NAME	CONTENT	YEAR(S)/DATE DATA APPLY TO	TABLE	FIGURE
	GENERAL PHYSICIANS	("arrondissement"): RIZIV/INAMI recognized physicians and non-recognized physicians			RIZIV/INAMI data)
NIS/INS (National Institute of Statistics) [94]	POPULATION DATA	Belgian Demographic Database by Region (lowest level: community) aggregated by age groups of 5 years + median/mean income per rgeion	2004		16
MARSHALL ET AL[10]	INTERNATIONAL DATA ON NUMBER OF PSGs	International data for PSG utilisation rates in pre-published paper and further details for paediatric PSGs in Australia from personal communication with authors.	2004	4	
UNMS/NVSM [116]	AFFILIATES DATABASE	Database for large Belgian sickness fund (covering around 30% of all Belgian residents). We requested utilisation data on (poly)somnographic interventions for outpatients/inpatients by billing codes 474563, 474552, 474541, 474530	2004-2005	6,7	
CM/MC [12]	AFFILIATES DATABASE	Database for large Belgian sickness fund (covering around 43% of all Belgian residents). We requested utilisation data on (poly)somnographic and infant home monitoring interventions for outpatients/inpatients by billing codes 474563, 474552, 474541, 474530	2004-2005	3,6,7	

4.2 METHODOLOGICAL NOTES

4.2.1 Figure 9: related trend regression

The REG Procedure (SAS 9.0)

Model: Linear_Regression_Model

Dependent Variable: GEVALLEN

Number of Observations Read	10
Number of Observations Used	10

Analysis of Variance

Source	DF	Sum of Squares	Mean Square	F Value	Pr > F
Model	1	116376488	116376488	749.36	<.0001
Error	8	1242403	155300		
Corrected Total	9	117618890			

Root MSE	394.08166	R-Square	0.9894
Dependent Mean	25675	Adj R-Sq	0.9881
Coeff Var	1.53486		

Parameter Estimates

Variable	Label	DF	Parameter Estimate	Standard Error	t Value	Pr > |t|
Intercept	Intercept	1	2400475	86752	27.67	<.0001
Prestatiejaar	Prestatiejaar	1	-1187.69697	43.38696	-27.37	<.0001

4.2.2 Distribution by Hospital: formulae en software procedures

We applied three different measures to assess the distribution of PSG-stays between 1995 and 2004:

- The Gini coefficient (GINI), ranging from 0 (completely unequal distribution) to 1 (fully equal distribution).
- The coefficient of variation (CV): a relative measure derived from taking the ratio of standard deviation to mean.
- The Herfindahl-Hirschman Index, ranging from 0 (completely equal) to 10.000 (completely inequal distribution).

Gini Coefficifient

Gini coefficient for ordered data (frequencies by ascending order):

$$G = \frac{\sum_{i=1}^{n} (2i - n - 1) x_i'}{n^2 \mu}$$

With n = number of observations (i to n, ranked in ascending order by frequencies); X is frequency value, μ = average frequency.

SAS procedure for GINI-coefficient: based on P. Dixon [13], further developed in SAS EnterpriseGuide, leveraging SAS®9.0

HHI

$$H = \sum_{i=1}^{n} (s_i^2)$$

With s = share of each hospital (i to n)

Analysis

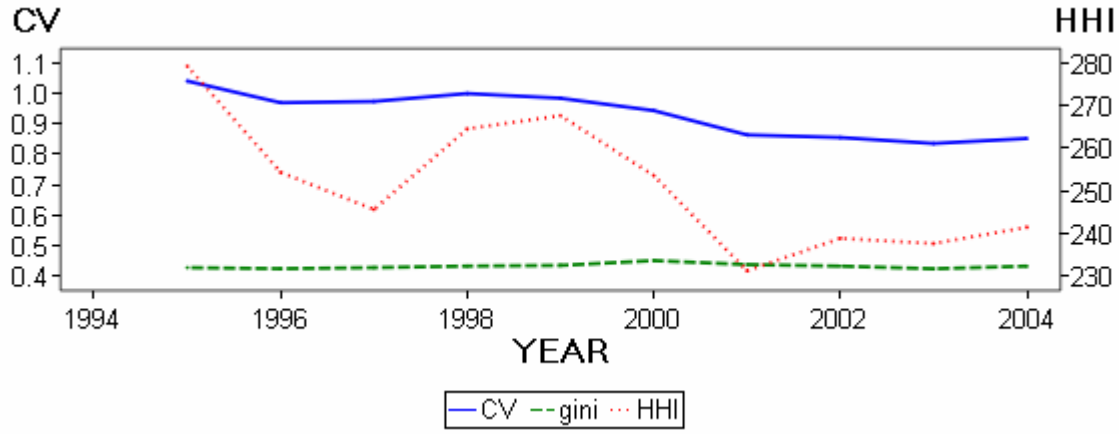
At first sight these different measures lead to conflicting conclusions:

- going by the GINI the dispersion would appear to have remained relatively stable throughout the analyzed period;
- judging by the CV the distribution would appear to have become more concentrated;
- following the HHI it would seem the distribution has evened out.

This paradoxical observation, however, can be explained by pointing out the mathematical characteristics of the varying measures. It is reported [14, 15] that the GINI is more sensitive to changes around the median, whereas the CV and HHI are respectively more sensitive to changes around the upper and lower part of the frequency domain.

The data seem to confirm this: most of the effect can be attributed to the (relative) redistribution from the observed frequency of the highest ranking hospital in our sample (dropping back from 3.227 PSG stays in 1995 to 1.290 stays in 2004) to lower ranking hospitals. As a result, some of the distribution weight was shifted from the upper end to the lower end of the distribution. Geometrically, this situation corresponds to the Lorenz curves[c] of 1995 and 2004 crossing each other: lower end hospitals have gained in share whereas upper end hospitals have lost in share.

Figure: Overnight PSGs 1995-2004: Concentration by Hospital



The table below depicts the overall stakes PSG-stays for patients below one year represent within their respective MDC-groups per hospital (number of hospital stays with main DRG coming under their respective MDC-groups for 2004). PSG-stays for patients below to the age of one make up 0,55% of all hospital stays in 2004.

[c] Cumulative frequency distribution with frequencies set as percentage.

Strikingly, polysomnographies performed during a hospital stay with indicated main DRG 862 account for over 20% of stays coming under the MDC 862 "Other Factors Influencing Health". Moreover, the coefficients of variations (CV) indicate that this average percentage is more concentrated than comparable values for other PSG-stays.

Table : Percentages Accounted for by PSGs in MDC-groups per Hospital (2004)

DRG	MDC		% PSG STAYS FOR ALL HOSPITALS AVERAGE	CV
058	01	Nervous System	1,94%	154%
144	04	Respiratory System	1,28%	218%
243	06	Digestive System	0,24%	142%
580-640	15	Neonates-Perinatal Disorders	1,87%	166%
862	23	Other Factors Influencing Health	20,54%	81%
ALL HOSPITAL STAYS			0,55%	110%

4.2.3 Geographical Distribution: exploratory association analyses

4.2.3.1 Admission Rate (AR) and Number of Physicians/MedianMean Income

PEDIATERS = number of paediatricians

HUISARTSEN = number of general physicians

GR04 = population aged 0-4 years

TOTALPOP = total population

Pearson Correlation Coefficients Prob > |r| under H0: Rho=0 Number of Observations	AR GR04 PER THOUSAND CAPITA
PEDIATERS/GR04	-0.02635
PEDIATERS/GR04	0.8685
	42
PEDIATERS/TOTALPOP	0.01157
PEDIATERS/TOTALPOP	0.9420
	42
HUISARTSEN/POP	0.33465
HUISARTSEN/POP	0.0283
	43
HUISARTSEN/GR04	0.14738
HUISARTSEN/GR04	0.3456
	43
AANTAL ERKENDE HUISARTSEN/GR04	0.14823
AANTAL ERKENDE HUISARTSEN/GR04	0.3428
	43
AANTAL ERKENDE HUISARTSEN/TOT	0.34116

Pearson Correlation Coefficients
Prob > |r| under H0: Rho=0
Number of Observations

	AR GR04 PER THOUSAND CAPITA
AANTAL ERKENDE HUISARTSEN/TOT	0.0252
	43
AANTAL NIET ERKENDE HUISARTSEN/G	-0.08915
AANTAL NIET ERKENDE HUISARTSEN/G	0.5697
	43
AANTAL NIET ERKENDE HUISARTSEN/T	-0.03369
AANTAL NIET ERKENDE HUISARTSEN/T	0.8302
	43
AANTAL HUISARTSEN (31_12_2003)/G	0.06283
AANTAL HUISARTSEN (31_12_2003)/G	0.6890
	43
AANTAL HUISARTSEN (31_12_2003)/T	0.21394
AANTAL HUISARTSEN (31_12_2003)/T	0.1683
	43
MEAN INCOME 2001	-0.12495
MEAN INCOME 2001	0.4247
	43
MEDIAN INCOME 2001	-0.20711
MEDIAN INCOME 2001	0.1827
	43

4.2.3.2 Admission Rate (AR) and Hospital Beds

E = number of beds in paediatric wards.

M = number of beds in maternity homes

Totalerk = total number of hospital beds

GR04 = population aged 0-4 years

TOTALPOP = total population

Pearson Correlation Coefficients Prob > |r| under H0: Rho=0 Number of Observations	AR GR04
E/GR04	0.18213
E/GR04	0.2738
	38
M/GR04	-0.02683
M/GR04	0.8712
	39
TOTALERK/TOTALPOP	-0.17946
TOTALERK/TOTALPOP	0.2616
	41
M/TOTALPOP	0.04254
M/TOTALPOP	0.7971
	39

4.3 CONTACTED INTERNATIONAL EXPERTS

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4.4 AUSTRALIAN DATA FOR PSG UTILISATION

4.4.1 Billing Codes for PSGs: descriptions

Category 2 - DIAGNOSTIC PROCEDURES AND INVESTIGATIONS
12203 OVERNIGHT INVESTIGATION FOR SLEEP APNOEA FOR A PERIOD OF AT LEAST 8 HOURS DURATION, FOR AN ADULT AGED 18 YEARS AND OVER WHERE: a) continuous monitoring of oxygen saturation and breathing using a multi-channel polygraph, and recording of EEG, EOG, submental EMG, anterior tibial EMG, respiratory movement, airflow, oxygen saturation and ECG are performed; b) a technician is in continuous attendance under the supervision of a qualified sleep medicine practitioner; c) the patient is referred by a medical practitioner; d) the necessity for the investigation is determined by a qualified adult sleep medicine practitioner prior to the investigation; e) polygraphic records are analysed (for assessment of sleep stage, arousals, respiratory events and assessment of clinically significant alterations in heart rate and limb movement) with manual scoring, or manual correction of computerised scoring in epochs of not more than 1 minute, and stored for interpretation and preparation of report ; and f) interpretation and report are provided by a qualified adult sleep medicine practitioner based on reviewing the direct original recording of polygraphic data from the patient - payable only in relation to each of the first 3 occasions the investigation is performed in any 12 month period.

Source: [16]

Category 2 - DIAGNOSTIC PROCEDURES AND INVESTIGATIONS
12207 OVERNIGHT INVESTIGATION FOR SLEEP APNOEA FOR A PERIOD OF AT LEAST 8 HOURS DURATION, FOR AN ADULT AGED 18 YEARS AND OVER WHERE: a) continuous monitoring of oxygen saturation and breathing using a multi-channel polygraph, and recordings of EEG, EOG, submental EMG, anterior tibial EMG, respiratory movement, airflow, oxygen saturation and ECG are performed; b) a technician is in continuous attendance under the supervision of a qualified sleep medicine practitioner; c) the patient is referred by a medical practitioner; d) the necessity for the investigation is determined by a qualified adult sleep medicine practitioner prior to the investigation; e) polygraphic records are analysed (for assessment of sleep stage, arousals, respiratory events and assessment of clinically significant alterations in heart rate and limb movement) with manual scoring, or manual correction of computerised scoring in epochs of not more than 1 minute, and stored for interpretation and preparation of report; and f) interpretation and report are provided by a qualified adult sleep medicine practitioner based on reviewing the direct original recording of polygraphic data from the patient *where it can be demonstrated that a further investigation is indicated in the same 12 month period to which item 12203 applies* for the adjustment and/or testing of the effectiveness of a *positive pressure ventilatory support device* (other than nasal continuous positive airway pressure) in sleep, in a *patient with severe cardio-respiratory failure,* and where previous studies have demonstrated failure of continuous positive airway pressure or oxygen - each additional investigation

Source: [16]

Category 2 - DIAGNOSTIC PROCEDURES AND INVESTIGATIONS
12210 OVERNIGHT PAEDIATRIC INVESTIGATION FOR A PERIOD OF AT LEAST 8 HOURS DURATION FOR A CHILD AGED 0 - 12 YEARS, WHERE: a) continuous monitoring of oxygen saturation and breathing using a multi-channel polygraph, and recording of EEG (minimum of 4 EEG leads with facility to increase to 6 in selected investigations), EOG, EMG submental +/- diaphragm, respiratory movement must include rib and abdomen (+/- sum) airflow detection, measurement of CO2 either end-tidal or transcutaneous, oxygen saturation and ECG are performed; b) a technician or registered nurse with sleep technology training is in continuous attendance under the supervision of a qualified paediatric sleep medicine practitioner; c) the patient is referred by a medical practitioner; d) the necessity for the investigation is determined by a qualified paediatric sleep medicine practitioner prior to the investigation; e) polygraphic records are analysed (for assessment of sleep stage, and maturation of sleep indices, arousals, respiratory events and the assessment of clinically significant alterations in heart rate and body movement) with manual scoring, or manual correction of computerised scoring in epochs of not more than 1 minute, and stored for interpretation and preparation of report; f) the interpretation and report to be provided by a qualified paediatric sleep medicine practitioner based on reviewing the direct original recording of polygraphic data from the patient. payable only in relation to the first 3 occasions the investigation is performed in a 12 month period.

Source: [16]

Category 2 - DIAGNOSTIC PROCEDURES AND INVESTIGATIONS
12213 OVERNIGHT PAEDIATRIC INVESTIGATION FOR A PERIOD OF AT LEAST 8 HOURS DURATION FOR A CHILD AGED BETWEEN 12 AND 18 YEARS, WHERE: a) continuous monitoring of oxygen saturation and breathing using a multi-channel polygraph, and recording of EEG (minimum of 4 EEG leads with facility to increase to 6 in selected investigations), EOG, EMG submental +/- diaphragm, respiratory movement must include rib and abdomen (+/- sum) airflow detection, measurement of CO2 either end-tidal or transcutaneous, oxygen saturation and ECG are performed; b) a technician or registered nurse with sleep technology training is in continuous attendance under the supervision of a qualified sleep medicine practitioner; c) the patient is referred by a medical practitioner; d) the necessity for the investigation is determined by a qualified sleep medicine practitioner prior to the investigation; e) polygraphic records are analysed (for assessment of sleep stage, and maturation of sleep indices, arousals, respiratory events and the assessment of clinically significant alterations in heart rate and body movement) with manual scoring, or manual correction of computerised scoring in epochs of not more than 1 minute, and stored for interpretation and preparation of report; f) the interpretation and report to be provided by a qualified sleep medicine practitioner based on reviewing the direct original recording of polygraphic data from the patient. payable only in relation to the first 3 occasions the investigation is performed in a 12 month period.

Source: [16]

Category 2 - DIAGNOSTIC PROCEDURES AND INVESTIGATIONS
12215 OVERNIGHT PAEDIATRIC INVESTIGATION FOR A PERIOD OF AT LEAST 8 HOURS DURATION FOR CHILDREN AGED 0 - 12 YEARS, WHERE: a) continuous monitoring of oxygen saturation and breathing using a multi-channel polygraph, and recording of EEG (minimum of 4 EEG leads with facility to increase to 6 in selected investigations), EOG, EMG submental +/- diaphragm, respiratory movement must include rib and abdomen (+/- sum) airflow detection, measurement of CO2 either end-tidal or transcutaneous, oxygen saturation and ECG are performed; b) a technician or registered nurse with sleep technology training is in continuous attendance under the supervision of a qualified paediatric sleep medicine practitioner; c) the patient is referred by a medical practitioner; d) the necessity for the investigation is determined by a qualified paediatric sleep medicine practitioner prior to the investigation; e) polygraphic records are analysed (for assessment of sleep stage, and maturation of sleep indices, arousals, respiratory events and the assessment of clinically significant alterations in heart rate and body movement) with manual scoring, or manual correction of computerised scoring in epochs of not more than 1 minute, and stored for interpretation and preparation of report; f) the interpretation and report to be provided by a qualified paediatric sleep medicine practitioner based on reviewing the direct original recording of polygraphic data from the patient. where it can be demonstrated that a further investigation is indicated in the same 12 month period to which item 12210 applies, for therapy with Continuous Positive Airway Pressure (CPAP), bilevel pressure support and/or ventilation is instigated or in the presence of recurring hypoxia and supplemental oxygen is required - each additional investigation.

Source: [16]

Category 2 - DIAGNOSTIC PROCEDURES AND INVESTIGATIONS
12217 OVERNIGHT PAEDIATRIC INVESTIGATION FOR A PERIOD OF AT LEAST 8 HOURS DURATION FOR CHILDREN AGED BETWEEN 12 AND 18 YEARS, WHERE: a) continuous monitoring of oxygen saturation and breathing using a multi-channel polygraph, and recording of EEG (minimum of 4 EEG leads with facility to increase to 6 in selected investigations), EOG, EMG submental +/- diaphragm, respiratory movement must include rib and abdomen (+/- sum) airflow detection, measurement of CO2 either end-tidal or transcutaneous, oxygen saturation and ECG are performed; b) a technician or registered nurse with sleep technology training is in continuous attendance under the supervision of a qualified sleep medicine practitioner; c) the patient is referred by a medical practitioner; d) the necessity for the investigation is determined by a qualified sleep medicine practitioner prior to the investigation; e) polygraphic records are analysed (for assessment of sleep stage, and maturation of sleep indices, arousals, respiratory events and the assessment of clinically significant alterations in heart rate and body movement) with manual scoring, or manual correction of computerised scoring in epochs of not more than 1 minute, and stored for interpretation and preparation of report; f) the interpretation and report to be provided by a qualified sleep medicine practitioner based on reviewing the direct original recording of polygraphic data from the patient. where it can be demonstrated that a further investigation is indicated in the same 12 month period to which item 12213 applies, for therapy with Continuous Positive Airway Pressure (CPAP), bilevel pressure support and/or ventilation is instigated or in the presence of recurring hypoxia and supplemental oxygen is required - each additional investigation.

Source: [16]

4.4.2 Further Analysis

The Australian data pertain to billing codes 12203, 12207 (adult population) and 12210, 12213, 12215, 12217 (paediatric population) from the Australian Medicare Benefits Scheme. As readers can verify above, the content of these interventions corresponds well to the scope of our report: overnight examination during sleep of respiratory, cardiologic and neurological physiologic signs.

Authors state[10] that their data apply to "more than 85-90% of total numbers of PSGs performed in Australia". Assuming that

- 15% of PSGs in Australia were not taken into account by the authors and (in all about 11.000 PSGs)
- those 15% exclusively regard children between the ages of 0 and 4 years and

We can derive a corresponding relative probability (corrected for size of population) of more than three (odds for Belgian patient between 0 and 4 years compared to similar Australian patient).

Table: PSG utilisation in general Belgian population

Billing Code	Description	# Cases
474563	Paediatric Inpatient PSG	20.637
477234	Paediatric Outpatient PSG	7.766
477374	Adult Outpatient PSG	1.870
477385	Adult Inpatient PSG	21.502

4.5 ACCREDITED CENTRES

4.5.1 Name and Location

Table: Accredited Centres

CENTRE	
Het Koningin Paola Kinderziekenhuis	Antwerpen
L'Hôpital Universitaire des Enfants Reine Fabiola	Bruxelles
Het A.Z.-V.U.B.	Brussel (Jette)
Het U.Z. Gasthuisberg	Leuven
Het U.Z. Antwerpen	Edegem
Le C.H.N.D.R.F. Clinique Notre-Dame	Charleroi
Het A.Z. Sint-Jan	Brugge
Het Universitair Ziekenhuis Gent	Gent
Les Cliniques Universitaires Saint-Luc	Bruxelles
Het Virga Jesse Ziekenhuis	Hasselt
Le Centre de Référence Namurois du Sommeil pour Nourrissons	Namur
Le Centre Hospitalier Régional de la Citadelle	Liège

Table: Number of Home Monitoring Centres per Province

BELGIAN PROVINCES AND CAPITAL		NUMBER OF CENTRES IN 2006		POPULATION AGED 0-4 YEARS IN 2004		SQUARED KM
FRENCH DENOMINATION	DUTCH DENOMINATION					
Anvers	Antwerpen	2		87.846		2.867
Bruxelles-Capitale	Brussels Hoofdstedelijk gewest	3	4	143.346	67.551	162
Brabant flamande	Vlaams-Brabant	1			54.704	2.106
Brabant wallon	Waals-Brabant	0			21.091	1.091
Flandre occidentale	West-Vlaanderen	1		54.624		3.144
Flandre orientale	Oost-Vlaanderen	1		70.602		2.982
Limbourg	Limburg	1		40.138		2.422
Hainaut	Henegouwen	1		74.361		3.786
Liège	Luik	1		57.361		3.862
Luxembourg	Luxemburg	0		16.374		4.440
Namur	Namen	1		26.722		3.666
SUM		12		571.374		30.528

4.5.2 Data Transmitted by 12 Centres

The table below resumes the reported patient numbers for 31st December 2004 reported per centre and per category. It is clear from the matrix that frequencies vary considerably both along the "category" and "centre axis". It would seem logical we would obtain the most reliable measures of average duration for matrix cells belonging to the largest centres (2, 3, 6, 7 and 9) and the largest patient category (category 6).

Table: number of patients by category and centre (reference date: 31/12/2004)

	CENTRES												
CATEGORIES	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**	**10**	**11**	**12**	**SUM**
ALTE (PSG REQUIRED)	2	16	27	26	3	23	43	23	21	5	21	13	223
SID SIBLING (PSG REQUIRED)	2	8	7	9	5	15	6	5	7	4	5	7	80
PRETERM (SEVERE, NO PSG REQUIRED)	0	36	17	6	4	32	23	11	52	12	8	14	215
DRUG ABUSE MATERNITY (NO PSG REQUIRED)	0	6	0	0	0	3	1	0	2	0	3	2	17
PRTEREM (LESS SEVERE, PSG REQUIRED)	0	110	18	9	25	30	11	3	15	1	12	9	243
OTHER (PSG REQUIRED)	0	*157*	*78*	4	6	*54*	*33*	53	*80*	3	40	18	**526**
SUM	4	**333**	**147**	54	43	**157**	**117**	95	**177**	25	89	63	1.304

In the following table depicting the average duration we select the average duration for the matrix cells of which we assume they offer the most reliable indication of average duration. Bear in mind there is no information on the actual number of patients these averages relate to. We assume that the various group sizes will be proportional to those in the previous table.

Table: Indicated duration (number of monitoring days) by category and centre (applying to cohort of babies who started and finished monitoring during 2004)

	CATEGORIES					
CENTRE	**ALTE (PSG REQUIRED)**	**SID SIBLING (PSG REQUIRED)**	**PRETERM (SEVERE, NO PSG REQUIRED)**	**DRUG ABUSE MATERNITY (NO PSG REQUIRED)**	**PRTEREM (LESS SEVERE, PSG REQUIRED)**	**OTHER (PSG REQUIRED)**
1	122	275	114		99	76
2	110	204	153		159	*134*
3	183	290	183	214	217	*220*
4	104	238	150	153	116	119
5	278	226	89	58	125	217
6	121	215	111	76	100	*114*
7	214	275	76	61	214	*244*
8	226	125	174		163	220
9	162	164	134	81	114	*164*
10	85	137	85	70	119	64
11	172	156	144	111	205	171
12	102	244	111	89	121	132

In the below table we set the average duration for selected centres in category 6 and the percentage of patients monitored exclusively with memory-enabled monitors in those centres next to each other. Based on our literature review we would expect to find an indication that higher average duration occurs in centres with a higher share of memory-enabled monitors. The few data we gathered here certainly do not seem to point in that direction, nor do they point unequivocally in the opposite direction (longer duration for centres with more memory-enabled monitors).

Table: Indicated duration (number of monitoring days) by category and centre (applying to cohort of babies who started and finished monitoring during 2004)

	REPORTED AVERAGE DURATION FOR PATIENT COHORT IN		**PATIENTS EQUIPED EXCLUSIVELY WITH MEMORY-ENABLED MONITORS**	
CENTRE	**#DAYS**	**RANK**	**% PATIENTS**	**RANK**
2	134	4	0%	5
3	220	2	45%	3
6	114	5	72%	2
7	244	1	100%	1
9	164	3	25%	4

4.6 COST ALLOCATION MODEL (TABLE 7 OF REPORT)

Calculations (steps):

- We estimated the number of patients for each subgroup by splitting the overall number of PSGs in 2003 (RCM/MKG data) based on the P(S)G/capita data in table 5.
- Next, we estimated the number of monitoring days (2003, RIZIV/INAMI reported) for each subgroup by assuming around 10% of monitoring days were performed for patients who did not undergo a PSG-test (as was the case for the supplementary data we obtained with the LCM sickness fund).
- Then, in applying the proportion of relative duration we obtained from the LCM data, we divided the number of monitoring days and estimated an average duration for subgroups of monitored patients.
- Finally, we multiplied PSG and Monitoring Days per group with the (estimated) unit cost.

5 APPENDIX TO CHAPTER VI

5.1 RESEARCH PROTOCOL FOR INTERVIEWS (FRENCH VERSION)

Objectifs des entretiens :

Professionnels :

- Identifier les interlocuteurs des professionnels de santé concernant la prévention et le dépistage de MSN
- Identifier les interlocuteurs des parents dans la prévention de la MSN
- Identifier l'évolution des messages de prévention diffusés / reçus dans la prévention de la MSN – dont les outils
- Identifier les messages diffusés / reçus concernant la PSG dans le dépistage de la MSN – dont les outils
- Identifier les données disponibles en rapport avec les PSG
- Identifier les connaissances actuelles et passées sur l'utilité des PSG (enfants < 1an) dans le dépistage des risques de MSN et dans d'autres contextes
- Collecter les avis sur les monitorings non-prescrits (type angelcare)

Parents

Situer les parents d'un point de vue sociodémographique (age de la mère, du père, état civil, nombre d'enfants, age au premier enfant) + niveau d'étude / profession / statut emploi – statut tabagique - assurance hospitalisation ?

Age de l'enfant examiné

Milieu d'accueil de l'enfant

- Identification des connaissances relatives a la MSN
- Identification des sources d'information
- Identification des messages reçus (pratiques de prévention / dépistage)
- Identification des pratiques de prévention appliquées
- Identification des connaissances sur la PSG (objectives)
- Identification des attentes vis-à-vis de la PSG
- Identification des motivations de recours à une PSG
- Identification du processus de décision de recours a la PSG (qui a décidé)
- Identification des connaissances quant à la suite d'une PSG
- Identification des connaissances relatives au monitoring

Méthodologie :

1/ Entretiens auprès des professionnels :

ONE

K&G

Association de pédiatres belges

Vereniging van de Belgische kinderartsen

1/ Entretiens auprès des parents

Seize interviews sont programmées (8 francophones / 8 néerlandophones) dans 4 hôpitaux

Elles auront lieu au moment du test PSG

Les entretiens sont programmés en concertation avec le médecin directeur de l'unité et après obtention de l'accord des parents par celui-ci.

Avant le début de l'entretien, les parents reçoivent une information orale et écrite sur qui nous sommes et dans quel cadre et a lieu la recherche.

Cette note comporte aussi un volet garantissant le traitement confidentiel et anonyme des entretiens ainsi que les coordonnées de l'équipe de recherche

L'enquêteur leur demande l'autorisation d'enregistrer l'interview.

A la fin de l'entretien, l'enquêteur demandera au parent l'autorisation de les recontacter

1/ pour prendre des nouvelles de l'enfant (issue du test)

2/ pour soumettre la synthèse de l'entretien pour validation

3/ pour envoi du rapport final

Un petit cadeau sera offert au parent qui aura participé.

Aucune information de type médicale ne sera fournie par l'enquêteur.

En cas de demande des parents, il référera au médecin responsable.

Si une interview concernait un enfant subissant une PSG pour une raison spécifique qui n'attrait pas à sa survie, un autre enfant sera recherché.

5.2 RESEARCH PROTOCOL FOR INTERVIEWS (ENGLISH VERSION)

Objectives of the interviews:

Professionals:

- To identify interlocutors among health professionals with regard of the prevention and detection of SIDS
- To identify interlocutors among parents with regard to the prevention of SIDS
- To identify the evolution of broadcasted / received SIDS prevention messages – including possible media/tools.
- To identify the broadcasted / received SIDS prevention messages – including possible media/tools.
- To identify available data on the topic of the PSG.
- To identify current and past knowledge on the usefulness of the PSG (infants <1 year) and in different contexts
- To document opinions on non-prescribed monitoring devices (e.g. angelcare)

Parents

To identify parents' socio-demographic background (age of mother, age of father, marital status, number of children, age of the first child) + educational level / profession / unemployment / smoking habits / complementary insurance

Infant's environment:

- To identify knowledge on SIDS
- To identify sources of information
- To identify the received messages (prevention measures/SIDS detection)
- To identify applied prevention measures
- To identify knowledge on the PSG (objectives)
- To identify expectations vis-à-vis the PSG
- To identify reasons for undergoing a PSG
- To identify the decision process leading up to a PSG (who decided)
- To identify knowledge on possible aftermath of a PSG test
- To identify knowledge on monitoring

Methods:

1) Interviews with health professionals:

ONE

Kind & Gezin

Belgian paediatric association

2) Interviews with parents

16 interviews are planned (8 French-speaking, 8 Dutch-speaking) in 4 hospitals.

These interviews are held at the time the PSG test is taken.

The interviews are planned in agreement with the chief physician of the department and after parental consent communicated to the chief physician.

Before the interview starts, parents receive oral and written information on the identity of the KCE team member and context of our research.

This written document also encompasses a guarantee of confidential and anonymous processing of all interviews. Also, KCE contact details are featured.

The interviewer asks the authorisation to tape-record the interview.

Before the end of the interview, the interviewer asks the parent(s) the authorisation, to contact them (*note by authors: this part of the protocol was altered later on for reasons of patient anonymity, eventually patients were given the necessary contact details of a KCE member for further contact exclusively at the patient's initiative*):

1) to find about the infant (follow-up of test)

2) to present the summary of the interview for parental validation

3) to send the final report

A small gift will be presented to participating parents.

No medical information will be prompted by the interviewer.

If parents have medical questions, the interviewer will refer to the local physician in charge.

If an interview would regard a child undergoing a PSG for a specific reason not relating to its survival, this interview will be barred from further analysis and another infant's parents will be contacted.

5.3 GUIDE INTERVIEWS WITH HEALTH PROFESIONALS (DUTCH VERSION)

Gebruik van PSG in de opsporing van risicofactoren bij wiegendood

Gesprekspartner :

Datum :

Plaats :

Aanvangsuur : Einde :

Opmerkingen :

Momenteel voert het KCE onderzoek naar de diagnostische meerwaarde van polysomnografische testen bij **zuigelingen tot één jaar oud.** Een ruime literatuurstudie over diagnostiche indicaties van deze test werd reeds uitgevoerd. Verder zal een evaluatie van het gebruik ervan in andere Westerse landen volgen. Dit project omvat daarenboven ook een "kwalitatief" luik. Enerzijds trachten we hierbij de redenen te identificeren waarom ouders een polysomnografie laten uitvoeren. Anderzijds wensen we een verantwoordelijke van elke beroepsvereniging te onmoeten om hun inschatting te kennen van de plaats die polysomnografieën (en bij uitbreiding (thuis)monitoring voor wiegendood) innemen binnen het huidige zorgkader voor zuigelingen..

Kan u bondig omschrijven wie u bent:

Welke instelling vertegenwoordigt u?

Wat is uw rol binnen die instelling?

LUIK WIEGENDOOD

Welke zijn de activiteiten van uw instelling en/of uw persoonlijke activiteiten die verband houden met wiegendood?

Wie zijn naar uw mening de voornaamste gesprekspartners voor neonatale geneeskundigen in verband met wiegendood?

En de gesprekspartners voor de ouders ?

Hoe zijn de kennis, informatiecampagnes en overige activiteiten met betrekking tot wiegendood geëvolueerd volgens u?

In het algemeen

In uw instelling

Welk type materiaal is beschikbaar op dit vlak ?

Voor de professionals (flyers, website, etc)

Voor de ouders

LUIK PSG

Welke is naar uw mening de rol en het nut van het gebruik van een PSG (vóór de leeftijd van 1 jaar)

- Inzake wiegendood - Overige indicaties

Waarin verschilt deze visie, indien deze verschilt, van (uw) vroegere visies?

Timing

Ideeën

Wat is de houding van uw instelling met betrekking tot polysomnografische test bij de preventie van wiegendood?

Aan wie wordt deze boodschap overgemaakt (ouders? professionals ?)

Hoe wordt deze boodschap overgemaakt ?

Beschikbare middelen ?

Heeft u de indruk dat met betrekking tot dit punt er een consensus heerst onder professionals?

Hoe zijn de verhoudingen met

K&G en/of ONE (in verband met wiegendood)

Verenigingen van pediaters / huisartsen

LUIK Monitoring

Welke is volgens u de rol en het nut van thuismonitoring voor een kind in zijn eerste levensjaar?

Welke houding geldt er in uw instelling ten opzichte van thuismonitoring in het kader van wiegendoodpreventie?

Aan wie wordt deze boodschap overgemaakt ? (ouders ? professionals ?)

Hoe wordt deze boodschap overgemaakt ?

- Beschikbare middelen ?

Heeft u de indruk er een consensus heerst onder professionals over dit punt ?

Hoe ervaren ouders volgens u de thuismonitoring van hun kind ?

Indien van toepassing, wat kan er volgens u gedaan worden om de huidige situatie te verbeteren?

Bent u op de hoogte van het bestaan van monitors (of materiaal van het type "angel care" – niet zelf aanhalen!) die zouden verhandeld worden en gebruikt worden door ouders zonder voorschrift?

Indien dit het geval is, welk materiaal betreft het ?

Welke voordelen ziet u in het bestaan of het gebruik van dit type materiaal?

Welke nadelen ziet u in het bestaan of gebruik van dit type materiaal?

Indien niet aangehaald: welke gevaren kan dit inhouden volgens u?

LUIK Gegevens

Welke gegevens verzamelt u in verband met wiegendood, PSG en monitoring?

Met welke regelmaat

Voor welke doelstelling

Worden deze gegevens gebruikt ?

Door wie ?

Zijn ze beschikbaar ?

Wenst u verdere opmerkingen of aanvullingen te maken wat betreft het gebruik van PSG of monitoring bij de preventie van wiegendood?

Bijvoorbeeld :Zouden, volgens u, sommige pediaters bepaalde PSG-tests of andere technische ingrepen kunnen misbruiken uit financiële overwegingen?

We danken u voor uw medewerking..

We stellen voor u de samenvatting van ons interview toe te sturen voor eventuele correctie en uw akkoord.

Uiteraard houden wij u op de hoogte van de publicatie van ons uiteindelijke rapport (voorzien eind 2006, begin 2007).

5.4 GUIDE INTERVIEWS WITH HEALTH PROFESIONALS (FRENCH VERSION)

Guide d'entretien Professionnels de la santé : Utilisation des PSG dans le dépistage des risques de mort subite du nourrisson

Interlocuteur :

Date :

Lieu :

Heure début : heure fin :

Commentaires :

Pour le moment, le KCE est en train de réaliser une étude sur l'apport diagnostic de la polysomnographie **chez le nourrisson de moins d'un an**. Jusqu'ici nous avons déjà réalisé une revue de la littérature assez large relative aux indications diagnostiques de ce test. Ensuite nous comptons mener une étude comparative sur l'usage des polysomnographies dans d'autres pays occidentaux et enfin, ce projet comporte une partie 'qualitative'. D'une part, nous essayerons d'identifier les raisons qui amènent les parents à faire passer une polysomnographie. D'autre part, nous souhaitons rencontrer un responsable de chaque organisme professionnel afin d'appréhender leur opinion sur la place des polysomnographie (et ensuite des monioring pour la MSN) dans le contexte actuel des soins aux nourrissons.

En quelques mots, de manière générale, qui êtes-vous :

À quelle institution appartenez-vous ?

Quel est votre rôle dans cette institution ?

VOLET MSN

Quelles sont les activités de l'institution et/ou les vôtres en relation avec la Mort Subite du Nourrisson ?

D'après vous, quels sont les principaux interlocuteurs des professionnels de la petite enfance par rapport à la MSN ?

Et les interlocuteurs pour les parents ?

D'après vous, comment ont évolué les connaissances, les campagnes, les activités relatives à la prévention de la MSN ?

En général

Dans votre institution

Quel type de matériel est disponible dans ce domaine ?

Pour les professionnels (flyers, website, etc)

Pour les parents

VOLET PSG

D'après vous, quelle est la place et l'utilité du recours a une PSG (avant 1 an) ?

- Pour la MSN
- Autres indications

En quoi cette vision diffère-t-elle, le cas échéant, d'avant ?

Timing

Idées

Quel discours tient votre institution par rapport à la PSG dans le contexte de la prévention de la MSN ?

A qui ce message est-il diffusé (parents ? professionnels ?)

Comment le message est-il diffusé ?

Outils disponibles ?

Avez-vous l'impression qu'il existe un consensus des professionnels par rapport à ce point ?

De quelle nature sont les liens/concertation avec :

K&G et/ou ONE (en rapport avec la MSN) ?

Associations de pédiatres / médecins généralistes

VOLET Monitoring

D'après vous, quelles sont la place et l'utilité de la mise sous monitoring d'un enfant de moins d'un an ?

Quel discours tient votre institution par rapport à la mise sous monitoring dans le contexte de la prévention de la MSN ?

A qui ce message est-il diffusé (parents ? professionnels ?)

Comment le message est-il diffusé ?

- Outils disponibles ?

Avez-vous l'impression qu'il existe un consensus des professionnels par rapport à ce point ?

D'après vous, comment les parents vivent-ils la mise sous monitoring de leur enfant ?

D'après vous, que peut-on faire pour améliorer la situation le cas échéant ?

Avez-vous connaissance de l'existence de monitoring (ou matériel de ce type 'angel care'- ne pas citer) qui circulerait et serait utilisé par les parents sans prescription médicale ?

Si oui de quel matériel s'agit-il ?

Quels avantages voyez-vous dans l'existence ou l'utilisation de ce type de matériel ?

Quels désavantages voyez-vous dans l'existence ou l'utilisation de ce type de matériel ?

Si pas cité : quels danger cela peut-il représenter à vos yeux ?

VOLET Données

Quelles données collectez-vous en rapport avec la MSN, la PSG, le monitoring ?

A quel rythme

Pour quelle finalité

Les données sont-elles utilisées ?

Par qui ?

Sont-elles disponibles ?

Par rapport à l'usage de PSG ou de monitoring dans la prévention de la MSN, souhaiteriez-vous ajouter quelque chose, aborder un autre thème ?

Par exemple, selon vous, les PSG ou des actes techniques pourraient-ils être utilisés à des fins de financement pour certains pédiatres ?

Nous vous remercions pour votre aide.

Je vous propose de vous envoyer la synthèse de notre entrevue pour correction éventuelle et accord.

Nous vous tiendrons informé de la sortie de notre rapport final (prévue pour fin décembre 2006)

5.5 GUIDE INTERVIEWS PARENTS (DUTCH VERSION)

Avant de faire passer l'entretien, lire le consentement informé, en faire signer un exemplaire (à conserver) et laisser le deuxième aux parents – Si OK lancer l'enregistrement. – après l'interview remettre le cadeau.

Gids onderhoud Ouders: Gebruik van PSG

Gesprekspartner : vader / moeder / overige: _____

Datum :

Plaats :

Uur van aanvang : Uur van beëindiging:

Opmerkingen :

Luik PSG

1/ Laten we het over uw kind hebben : Wat is zijn/haar naam, Zijn/haar leeftijd, Zijn er broertjes en zusjes (welke leeftijd),

Wie houdt zich met uw kind bezig overdag?

Werkt u? Welk soort werk?

Werkt de vader? Welk soort werk?

2/ Hoe stelt uw kind het in het algemeen?

3/ Waarom bent u hier vandaag gekomen met uw kind?

4/ Hoe bent u de hoogte geraakt van de slaaptest?

- ONE / K&G
- pediater / huisarts
- ouders

- vrienden

5/ Wie heeft u aangeraden de test te laten doen?

-ouder

- pediater /geneesheer

-vrienden

6/ Wat/wie heeft u overtuigd de test te doen?

7/ Waar dient deze test voor volgens u ?

8/ Wat hoopt u te leren uit deze test / wat zal er na de test gebeuren volgens u ?

bevestiging van vermoeden

weerlegging van vermoeden

advies tot thuismonitoring

Ons onderzoek richt zich in het bijzonder op wiegendood, we hadden graag enkele vragen gesteld over dit onderwerp.

9/ Wat weet u over wiegendood?

10/ Wat kan volgens u gedaan worden om wiegendood te vermijden?

11/ Vanwaar heeft u die informatie?

12/ Zijn de adviezen over preventie voor het publiek voldoende duidelijk volgens u?

13/ Is er een verandering in die adviezen door de tijd heen, indien ja, welke?

14/ Neemt u speciale voorzorgsmaatregelen?

15/ Rookt u? (vader/moeder)

Op welke plek?

In het bijzijn van uw kind?

INDIEN UIT HET INTERVIEW HET GEBRUIK VAN THUISMONITORING NAAR VOREN KWAM

16/ Wat is volgens u het nut van een zuigeling tot één jaar aan een monitor te leggen ?

17/ Had u hier graag iets aan toegevoegd in verband met de slaaptest, wiegendood (of thuismonitoring)?

5.6 GUIDE INTERVIEWS PARENTS (FRENCH VERSION)

Guide d'entretien Parents : Utilisation des PSG

Interlocuteur : père / mère / autre : _____

Date :

Lieu :

Heure début : heure fin :

Commentaires :

VOLET PSG

1/ Parlez-moi de votre enfant : Comment s'appelle-t-il, Quel âge a-t-il, A-t-il de frères et sœurs (quel age),

Qui s'occupe de lui en journée ?

Vous travaillez ? Quel type d'emploi

Le papa travaille-t-il ? Quel type d'emploi

2/ De manière générale, comment va votre enfant ?

3/ Pourquoi souhaitiez-vous faire le test du sommeil ?

4/ Comment avez-vous entendu parlé de ce test?

- ONE / K&G
- pédiatre / médecin de famille
- parents
- amis

5/ Qui vous a conseillé de le faire ?

-parent
- pédiatre /médecin
-amis

6/ Qu'est-ce qui/qui vous a décidé à le faire ?

7/ D'après vous, à quoi sert ce test ?

8/ Qu'espérez-vous apprendre à l'issue de ce test / que se passera-t-il d'après vous ?

confirmation doute

infirmation doute

mise sous monitoring

Notre étude s'intéresse particulièrement à la mort subite du nourrisson, nous aimerions vous posez quelques questions sur ce sujet.

9/ Que connaissez-vous de la MSN ?

10/ Que peut-on faire d'après vous pour l'éviter ?

11/ D'où tenez-vous ces informations ?

12/ D'après vous, les messages de prévention diffusés dans le public sont-ils clairs ?

13/ Ont-ils évolué au cours du temps, si oui comment ?

14/ Et vous, que faites-vous Que faites-vous pour la prévenir ?

15/ fumez-vous ? (père / mère)

A quel endroit ?

En présence de votre enfant ?

SI LA MISE SOUS MONITEUR A ETE EVOQUEE DANS L'ENTRETIEN

16/ Quelle est à votre avis l'utilité de la mise sous moniteur d'un enfant de moins d'un an ?

17/ Souhaiteriez-vous ajouter quelque chose, que ce soit en rapport avec le test du sommeil, la mort subite du nourrisson (ou le monitoring) ?

5.7 ANALYSIS ROSTER INTERVIEWS PARENTS (DUTCH/FRENCH)

	Unités d'enregistrement								
Unités d'analyse	**Lien de parenté**	**hôpital**	**langue**	**Sexe de l'enfant**	**Age de l'enfant**	**Fratrie**	**Travail de la mère**	**Travail du père**	**Test avant**
LAK1	parents	1	francophone	F	5 mois	-	Employée dans secrétariat	Employé dans le secteur de la vente	2ème test (contrôle)
LAK2	mère	1	francophone	F			Employée chez un traiteur	Enseignant (enseignement supérieur)	2ème test (contrôle)
LAK3	père	1	francophone	G	3 mois	2 sœurs (10 et 12 ans)	dentiste	Business international	2ème test (1er pas clair)
LAK4	mère	1	francophone	G	7 semaines	2 frères (6 ans sous monito jusqu'à 9 mois et 18 mois- sous monito jusqu'à 6 mois)	Employée dans le secteur bancaire	Employé dans une société photocopieurs	1er test
LAK5	mère	1	francophone	F	4 mois	-	Éducatrice	mécanicien	1er test
LAK6	mère	2	francophone	F	2 mois	-	Aide-ménagère	Employé dans le secteur de l'alimentation	1er test
LAK7	parents	2	francophone	F	3 mois	1 sœur (3ans)	Coiffeuse –aide ménagère	Employé dans la sécurité	1er test
LAK8	parents	2	francophone	F	3 semaines	-	Sans emploi (étudiante aide soignante)	Sans emploi (maroquinerie) - handicapé	1er test
LAK9	mère	2	francophone	G	10 jours	-	Coiffeuse en Algérie	technicien	1er test

	Unités d'enregistrement								
Unités d'analyse	**Lien de parenté**	**hôpital**	**langue**	**Sexe de l'enfant**	**Age de l'enfant**	**Fratrie**	**Travail de la mère**	**Travail du père**	**Test avant**
MAE1	Beide ouders	3	Nederlandstalig	J	1 maand	1 broer (5 jaar)	verpleegster	bediende	1ste test
MAE2	Beide ouders	3	Nederlandstalig	J	3.5 maand	1 broer (20 maand)	Ambtenaar	Schilder	1ste test
DIV1	Beide ouders	4	Nederlandstalig		8 weken	-	verpleegkundige	Voedingssector	1ste test
DIV2	Beide ouders	4	Nederlandstalig	J	9 weken	-	Zelfstandig HORECA	Zelfstandig HORECA	1ste test
DIV3	Beide ouders	4	Nederlandstalig	M	10 weken	-	Bediende	bouwsector	1ste test
DIV4	Beide ouders	4	Nederlandstalig	M	10 weken	1 broer (2.5 jaar)	Huisvrouw	schrijnwerker	1ste test
DIV5	Beide ouders	4	Nederlanstalig	M	4 maand	1 broer (4.5 jaar)	Onthaalmoeder	ambtenaar	2de test (controle)

	Unités d'enregistrement						
Unités d'analyse	**Etat général de l'enfant**	**Motivation pour faire passer le test**	**Source(s) d'information relative(s) au test**	**Personne qui a conseillé de faire le test**	**Personne qui a décidé du test**	**Utilité perçue du test**	**Suite envisagée / attendue du test**
LAK1	Excellent Aucun problème à la naissance	Par précaution – pour dépister risque de MSN	Via la kiné post-natale qui a parlé d'une maman qui avait perdu son enfant : une autre maman a alors parlé du test + via le cousin de la maman qui avait passe un test et été mis sous monitoring 5 ans auparavant + via les amis déjà parents	Une fois que les mamans savent que le test existe, elles veulent le faire	Les parents, les pédiatres n'était pas contraire et n'a pas essayé de dissuader les parents de le faire	Voir si apnées trop longues ou si risque de décès	1er test pour vérifier qu'elle n'était pas à risque - après le 2ème test, veulent savoir si le système nerveux à maturation et plus besoin de monitoring
LAK2	Hypotonie à la naissance – dystrophie musculaire → EXCLU DE L'ANALYSE						
LAK3	Excellent sauf problèmes de respiration pendant la nuit	Impression d'étouffement pendant le sommeil – fait beaucoup de bruit en dormant → Mise au point avant de le mettre dans sa propre chambre (+ demande de la maman indépendamment de toute source d'inquiétude)	Une sœur aînée a eu le test Via la maman	Demande des parents via le pédiatre	Pas clair pour le répondant : les parents ou le pédiatre	Contrôler le nombre et la durée des apnées obstructives	S'attendait à ce que tout soit normal. 2ème test pour vérifier parce que 3 apnées obstructives donc doute
	Unités d'enregistrement						
Unités d'analyse	**Etat général de l'enfant**	**Motivation pour faire passer le test**	**Source(s) d'information relative(s) au test**	**Personne qui a conseillé de faire le test**	**Personne qui a décidé du test**	**Utilité perçue du test**	**Suite envisagée / attendue du test**
LAK4	excellent	L'aîné a passé le test parce que 2 cas de décès dans la famille (1 cousin et 1 tante) – juste pour vérifier mais pas de réelle inquiétude de la part de la maman	Par des amis Dans la famille enfants sous monitoring	Le pédiatre pour rassurer la maman	La maman le fait d'office pour ses enfants (comme le fait le reste de la famille)	Détecter les apnées obstructives et les bradycardies	S'attend à ce que soit positif car a beaucoup plus de symptômes que ses aînés
LAK5	excellent	Par prévention de la MSN– aucun symptôme – le test est accessible alors a demandé au pédiatre	Bouche à oreilles de parents qui l'ont fait – connaissance s qui connaissent quelqu'un qui a perdu un enfant A la kiné post-natale aussi A posé la question a K&G	Les parents ont demandé au pédiatre qui a fait la prescription	Les parents	Voir oxygène dans le sang, respiration correcte, importance des apnées, transpiration, pas de problème de sommeil	Espère apprendre que tout va bien, que pas d'apnées et que respire bien

			qui a renvoyé vers une responsabilité parentale				
LAK6	excellent	Pour être tranquille la nuit et la mettre dans sa chambre. Pas de soucis particulier, le bébé se réveille facilement - souhaite être rassurée	Amie qui a fait le test car son enfant avait un sommeil profond : l'amie avait acheté un matelas d'apnée – comme il sonnait souvent avait fait le test. A la TV : émission sur les prématurés et activités d'un service de pédiatrie	La maman a demandé au pédiatre qui a dit que ce n'était pas une obligation mais que ce n'était pas plus mal de faire le test	Les parents	Pour voir si le sommeil est profond. N'a pas reçu d'explications	Espère apprendre que tout va bien mais comme ne sait pas à quoi sert le test…s'attend à être rassurée

	Unités d'enregistrement						
Unités d'analyse	**Etat général de l'enfant**	**Motivation pour faire passer le test**	**Source(s) d'information relative(s) au test**	**Personne qui a conseillé de faire le test**	**Personne qui a décidé du test**	**Utilité perçue du test**	**Suite envisagée / attendue du test**
LAK7	Problème de laryngomalacie → EXCLU DE L'ANALYSE						
LAK8	excellent	Fait de petites apnées (mais se reprend toujours) – aurait fait le test par précaution de toutes façons à 2 mois	Pendant les études de la maman, Par la famille (enfants qui ont aussi fait le test)	Le pédiatre à la demande des parents	Le pédiatre à la demande des parents	Contrôler la respiration et le rythme cardiaque de l'enfant + le temps des apnées	Ne sait pas ce qui va se passer – espère apprendre que pas trop de risque de MSN – que si elle a des apnées, elles se reprend toute seule
LAK9	Bonne santé	Inquiétude – fait des apnées et pleurs – ont amené aux urgences parce que le papa était inquiet	Aux urgences	Le médecin aux urgences pour être sure	Le médecin aux urgences	Ne sait pas – était trop paniquée et ne comprend pas bien le français	Pense que le résultat va montrer qu'il va être handicapé ou qu'il va mourir demain. Le papa n'était pas inquiet du résultat
MAE1	Nog opgenomen in ziekenhuis - premature→ EXCLU DE L'ANALYSE						

	Unités d'enregistrement						
Unités d'analyse	**Etat général de l'enfant**	**Motivation pour faire passer le test**	**Source(s) d'information relative(s) au test**	**Personne qui a conseillé de faire le test**	**Personne qui a décidé du test**	**Utilité perçue du test**	**Suite envisagée / attendue du test**
MAE2	Heel gezond	Voor wiegendood	twee neefjes met hoog risico voor wiegendood (met "matje" + oudste zon getest	met twee pediaters gesproken. Beiden hebben gezegd "dat er geen indicaties waren, maar dat de test gedaan werd indien	zelf	Functies meten: oa ademhaling.	Weet dat test geen absolute zekerheid biedt Hoop dat alles OK is. Indien test niet OK is, dacht in eerste instantie aan "matje"
DIV1	in het algemeen goed	Gerusstelling door test	Via vrienden en familie + werk van de vrouw (iedereen heeft afgeradenà	Pediater heeft voorgeschreven op vraag van de ouders	wiegendood in de familie van de vrouw	Ademhaling meten Hartslag meten Hersenactiviteit meten (via brochure van ziekenhuis)	Dat hun kind geen risico loopt voor wiegendood
DIV2	In het algemeen stelt het goed	Veiligheid, zekerheid	Zuster van een ouder heeft 2 kinderen, allebeide getest.	Uit eigen beweging gevraagd aan pediater		Controle van: hart, ademhaling, zuurstof in bloed, hersenactiviteit, spieractiviteit.	Dat hun kind geen problemen heeft. Zekerheid bieden. Momenteel zijn beide ouders niet bijzonder ongerust

	Unités d'enregistrement						
Unités d'analyse	**Etat général de l'enfant**	**Motivation pour faire passer le test**	**Source(s) d'information relative(s) au test**	**Personne qui a conseillé de faire le test**	**Personne qui a décidé du test**	**Utilité perçue du test**	**Suite envisagée / attendue du test**
DIV3	In het algemeen stelt K het goed	SID-geval in familie + vader heeft slaapstoornissen: Schrik voor familiale antecedenten	via haar werk (verzekeringssector). Test zit in hospitalisatieverzekeringen.	Ze hebben het test aangekaart bij pediater	Pediater schreef voor omdat er familiale antecedenten zijn en ouders ongerust werden	Om na te gaan of er een risico is voor wiegendood. Test meet hart, longen, hersenen.	Hoop dat er geen risico aangetoond wordt. Indien risico aangetoond wordt, verwacht O1 monitor mee naar huis te krijgen.
DIV4	stelt het goed overdag, maar slaapt onrustig	Geruststelling. K was in ziekenhuis al 2 maal gestopt met ademhalen. Monitor werd meegegeven met ouder	Verpleegster had monitor aangeraden voor geruststelling	Pediater heeft geadviseerd de test te doen	Pediater heeft voorgeschreven.	Long, hart, hersenen, bewegingen	Hoop dat alles in orde blijkt. Indien positieve test monitor "tot de leeftijd van 2 jaar of zo".
DIV5							

	Unités d'enregistrement						
Unités d'analyse	**Connaissance sur la MSN**	**Moyens perçus pour éviter la MSN**	**Source(s) d'information relative(s) la prévention de la MSN**	**Clarté perçue des informations relatives à la MSN**	**Evolution au cours du temps perçue des messages de prévention de la MSN**	**Mesures de préventions de la MSN appliqués**	**Tabagisme / réaction face à l'environnement enfumé**
LAK1	Le bébé oublie de respirer durant son sommeil	Rester près du bébé chaque fois qu'il s'endort Température de la pièce Ne pas coucher sur le ventre	Température : au labo sommeil lors du 1er test Position couchage : maternité lors des séances d'allaitement mais pas pendant le séjour Pas de souvenir que le pédiatre en ai parlé (ni privé, ni à la maternité, ni à l'ONE)	Pas vraiment d'information, seulement si on pose des questions	Pendant les séances d'info sur l'allaitement, on a dit que la position de couchage recommandée changeait et que pour le moment c'est sur le dos	Dort sur le dos la nuit et sur le ventre en journée Sac de couchage Mesure de la température de la pièce Dort dans la chambre des parents	Père ex-fumeur Ne vont pas au restaurant à cause de la fumée (en attente de la loi) Visiteurs fument sur la terrasse

	Unités d'enregistrement						
Unités d'analyse	**Connaissance sur la MSN**	**Moyens perçus pour éviter la MSN**	**Source(s) d'information relative(s) la prévention de la MSN**	**Clarté perçue des informations relatives à la MSN**	**Evolution au cours du temps perçue des messages de prévention de la MSN**	**Mesures de préventions de la MSN appliqués**	**Tabagisme / réaction face à l'environnement enfumé**
LAK3	La MSN touche un certain pourcentage d'enfant C'est la cause de mortalité infantile des plus importantes Les raisons ne sont pas claires (génétiques t autres) L'enfant s'arrête de respirer pendant la nuit et décède. Conditions augmentant le risque : tabagisme dans la famille, dormir sur le ventre, trop habillé, famille	Dort sur le dos Température de la chambre tabagisme	Internet Médecins Enfants précédents lectures		La position de couchage change tous les 4 ou 5 ans / 2 ou 3 fois tous les 10/20 ans	Dort sur le dos Pas de tabac Attentifs à la température de la chambre (mais pas mesurée) Pyjama grenouillère Sac de couchage pas trop épais	Plutôt 'anti' donc très attentifs à l'environnement sans fumée

	Unités d'enregistrement						
Unités d'analyse	**Connaissance sur la MSN**	**Moyens perçus pour éviter la MSN**	**Source(s) d'information relative(s) la prévention de la MSN**	**Clarté perçue des informations relatives à la MSN**	**Evolution au cours du temps perçue des messages de prévention de la MSN**	**Mesures de préventions de la MSN appliqués**	**Tabagisme / réaction face à l'environnement enfumé**
LAK4	L'enfant meurt dans son sommeil	Le test – il devrait être proposé à tous les parents L'hygiène de vie autour de l'enfant Pas de cigarette Température Eviter les médicaments Position sur le dos	Pédiatre et labo sommeil La position et l'hygiène de vie sont des messages diffusés souvent à la TV, dans les livres	Si on tombe sur une message, il est clair mais c'est rare – n'a rien entendu les dernières années	Pas d'évolution Pas assez d message	Test Aère la chambre Pas de tabac position sur le dos Pas trop couvert N'arrive pas à respecter la température	Personne ne fume chez eux
LAK5	L'enfant oublie de respirer – une apnée très importante entraîne la mort- causes possible : température trop élevée de la chambre – parents fumeurs – dort sur le ventre	Chambre entre 18-20°C Enfant pas trop couvert Coucher sur le dos Allaiter Bien aérer Sac de couchage Pas de tabac	Carnet de l'ONE Pédiatre (conseil de température pendant l'été) Livres sur la grossesse et la naissance 'c'est toujours dans l'information générale'	Les messages sont clairs	Moins de MSN grâce au test et aux messages de prévention mais la position change tous les 10 ans	Coucher sur le dos Pas trop chaud dans la chambre (mesure la température – thermomètre demandé comme cadeau de naissance) Ne fume pas	Demande aux parents d'aller fumer ailleurs quand l'enfant est présent, amis fument sur la terrasse Évite les endroits enfumés

	Unités d'enregistrement						
Unités d'analyse	**Connaissance sur la MSN**	**Moyens perçus pour éviter la MSN**	**Source(s) d'information relative(s) la prévention de la MSN**	**Clarté perçue des informations relatives à la MSN**	**Evolution au cours du temps perçue des messages de prévention de la MSN**	**Mesures de préventions de la MSN appliqués**	**Tabagisme / réaction face à l'environnement enfumé**
LAK6	Le sommeil est trop profond, c'est une question de respiration quand ils s'endorment	Le test Matelas bien dur Pas d'oreiller Pas de peluches Pas trop chauffer la chambre	Livre sur la grossesse Pédiatre (matelas) Internet Pas d'info venant de la maternité Magazine spécialisé ('9 mois' ou 'parents') + maman de la répondante	On n'en parle pas assez, n'a pas vu de messages dans le public		Matelas Chambre entre 18-20C° (mesure la température) Coucher sur le dos Attend 20-30 minutes après le repas avant de coucher l'enfant Couvertures pas trop hautes	Mère non-fumeuse, papa fume dehors (pas dans la voiture ni en présence de l'enfant)
LAK7							
LAK8	Les bébés meurent pendant leur sommeil, oublient de respirer – arrive souvent aux enfants de moins d'1 an	Température de la chambre +/- 18°C Ne pas trop couvrir l'enfant Couvertures pas trop hautes Coucher sur le dos Tétine Vêtements pas trop serrés	On le répète assez souvent, les infirmières aussi À 'école Dans les magazines	Messages clairs	La position a changé : sur le ventre puis sur le côté puis sur le dos	Chambre fraîche Coucher sur le dos Redescend les couvertures Dort à côté de la maman Vêtements pas serrant	Maman : A essayé sans succès d'arrêter de fumer pendant la grossesse (3à 5 cigarettes par jour). A arrêté depuis la naissance Papa non-fumeur Demande aux gens de fumer ailleurs

	Unités d'enregistrement						
Unités d'analyse	**Connaissance sur la MSN**	**Moyens perçus pour éviter la MSN**	**Source(s) d'information relative(s) la prévention de la MSN**	**Clarté perçue des informations relatives à la MSN**	**Evolution au cours du temps perçue des messages de prévention de la MSN**	**Mesures de préventions de la MSN appliqués**	**Tabagisme / réaction face à l'environnement enfumé**
LAK9	N'a jamais entendu parler de la MSN	Ne sait pas				Température de la chambre et du bain. A demandé elle-même à la maternité comment coucher son bébé	Non fumeurs Evitent les restaurants enfumés et les invités ne fument pas chez eux
MAE1							
MAE2	200 overleden per jaar in Belgie	Temperatuur Verluchting Geen rook Rugligging Niet te warm toedekken	Lectuur Internet K&G	vrij duid. Men moet wel zelf op zoek en wat rondkijken elijk	"ik hoor dat zeggen"		Beiden gestopt met roken, enkele jaren ervoor. Bezoek rookt ook niet in buurt van baby.
DIV1	stoppen met ademen, gevolgd door dood	Thuismonitor Temperatuur Rugligging Niet roken Goed verluchte kamer Geen dekbedje	Boeken van K&G	Advies van K&G is voldoende helder	Denkt dat het niet veranderd is	Kamer verluchten. Donsdeken weggehaald Temperatuur controleren. Roken vermijden	Vader rookt altijd buiten In het algemeen: rook vermijden

	Unités d'enregistrement						
Unités d'analyse	**Connaissance sur la MSN**	**Moyens perçus pour éviter la MSN**	**Source(s) d'information relative(s) la prévention de la MSN**	**Clarté perçue des informations relatives à la MSN**	**Evolution au cours du temps perçue des messages de prévention de la MSN**	**Mesures de préventions de la MSN appliqués**	**Tabagisme / réaction face à l'environnement enfumé**
DIV2	verstikking tijdens slaap	Rugligging Opletten bij Indekken (slaapzak) Temperatuur (18-20 graden) Geen pluchen beestjes Geen huisdieren in bed laten	Uit brochure gekregen op moment van afspraak in ziekenhuis Wel veel brochures (K& G,..), maar nog niet doorgenomen. Brochure van pediater.	Voldoende duidelijk	heeft de indruk dat de test relatief nieuw is	Rugligging Temperatuur Geen huisdieren Geen speelgoed	Beiden roken, maar niet in buurt van hun kind
DIV3	Stoppen met ademen, in diepe slaap raken en doodgaan	Rugligging. Temperatuur (gemeten met thermometer) Geen speeltjes in bed.	Brochures van K&G. + in materniteit brochure gekregen (NIET brochure van K&G) over PSG-test en monitors.	voldoende duidelijk		Rugligging. Temperatuur Geen speeltjes in bed. AngelCare	Geen van beiden rookt. Geen rook in de buurt van hun kind
DIV4	Het kind "vergeet te ademen" door te diepe slaap, waarop de dood volgt	Niet te warm indekken. Monitor. Geen rook	Via K&G + zus + K&G	"Brochures zijn niet altijd even duidelijk"	nee	"Niet te warm ingedekt" (18-20 graden)	Geen van beiden rookt Gasten gaan buiten roken.
DIV5							

	Unités d'enregistrement						
Unités d'analyse	**Utilité du moniteur**	**ajoutes**	**ajoutes**	**ajoutes**	**ajoutes**	**ajoutes**	
LAK1	Pour savoir s'il faut ranimer	La gardienne prône le test systématique Les répondants prônent le test systématique	Avaient consulté pour inquiétude parentale mais ont du repartir avec un moniteur	N'ont aucune idée du prix du test	Rassurés et habitués par le moniteur – pas de problème s'ils doivent le garder	Gestes de réanimation devraient être mieux expliqués aux parents	-
LAK3	Via Internet ou les copains : pas idéal pour les parents qui se réveillent tout le temps et peur puis problème pour arrêter le monitoring à 6 mois	Pourquoi laisser parti sans monitoring pour 3 semaines s'il y a un doute : étaient prêt à financer eux-mêmes le monitoring	Aucune cause de risque de MSN chez le bébé	Ne pense pas que le test soit fait dans d'autres pays	D'un point de vue psychologique, l'existence d'un test est rassurante pour les parents	Le fait qu'il ait eu des apnées pendant le test sans qu'il fasse du bruit alors que bruit d'habitude a angoissé le papa	-
LAK4	Détecter les apnées trop longues Sécuriser et rassurer les parents – c'est quand on sait qu'on est à risque que ça commence vraiment	Analogie avec la ceinture de sécurité : on la met mais on e se dit pas qu'on va avoir un accident – le test on le fait mais on ne pense pas que l'enfant va mourir de MSN	Ne sait pas combien ça coûte mais ça n'a pas d'importance	Une amie a fait le test – négatif – mais a quand même acheté un moniteur (1500€) Elle, elle fait confiance au milieu médical	-		
LAK5	Non abordé	-	-	-	-	-	

	Unités d'enregistrement						
Unités d'analyse	**Utilité du moniteur**	**ajoutes**	**ajoutes**	**ajoutes**	**ajoutes**	**ajoutes**	
LAK6		On devrait informer les parents sur le test et la position	Avait pensé à utiliser un matelas d'apnées mais le médecin l'a dissuadée et a alors conseillé plutôt le test	Aucune idée du prix – aurait pu mettre n'importe quel prix	Pas d'information sur la suite si le test était positif	On devrait faire des séances sur ce qu'il faut faire et ne pas faire, comme les séances sur l'allaitement	-
LAK7							
LAK8	Non abordé	Ne sait rien sur comment le test se passe	-				
LAK9	Non abordé						
MAE1							
MAE2	Weet niets extra rond	Vrij weinig mensen die die test doen, veel mensen die niet op de hoogte zijn van bestaan					
DIV1	Alarm bij ademhaling. "Omdat we denken dat wiegendood te wijten is aan ademhaling" Verwittiging om in te kunnen grijpen: wakker schudden, naar dokter bellen,...						

	Unités d'enregistrement						
Unités d'analyse	**Utilité du moniteur**	**ajoutes**					
DIV2		heeft vragen rond definitie van wiegendood	Test zou spontaner aangeboden moeten worden aan ouders of verplicht worden	Terugbetaling: niet volledig			
DIV3	Ongeveer zelfde als PSG-test: ademhaling, hart testen en alarm in geval van nood	heeft momenteel AngelCare-dekentje.	raadt test aan andere ouders aan, hoewel 100% zekerheid niet bestaat.				
DIV4		Folders moeten beter uitleggen + preventiemaatregelen duidelijker benadrukken	Tamelijk dikke boek van K&G" nodigt niet uit tot lezen	monitor zou alleen (retroactief) terugbetaald worden als PSG positief is.			
DIV5							

5.8 LITERATURE REVIEW: EFFECT OF HOME APNEA MONITOR – PARENT ISSUES

#	Search History MEDLINE (1996-2006) via OVID
1	exp Sudden Infant Death/
2	monitor$.mp.
3	exp Parents/
4	2 and 3
5	exp Stress/
6	5 and 4
7	home.mp.
8	(home adj3 monitor$).mp.
9	6 or 8
10	limit 9 to "all infant (birth to 23 months)"
11	Sleep Apnea Syndromes/
12	10 and 11
13	from 12 keep 2, 8-10, 14, 17, 31, 33...
14	13
15	limit 14 to yr="1996 - 2006"

#	Search History PSYCHINFO (1996-2006) via Ovid
1	exp Sudden Infant Death/
2	monitor$.mp.
3	exp Parents/
4	2 and 3
5	exp Stress/
6	5 and 4
7	home.mp.
8	(home adj3 monitor$).mp.
9	limit 8 to 100 childhood <birth to age 12 yrs>
10	9
11	limit 10 to yr="1996 - 2006"
12	from 11 keep 11, 16, 18-19

Embase

alarm monitor' OR **'alarm monitoring'**/exp OR **'apnea monitor'** OR **'home monitoring'** AND [infant]/lim AND [embase]/lim AND [1996-2006]/py

Sociological abstract:

monitor*

Table of evidence

Author, date, country	Type of study (sample/ methodology)	Focus	Results/outcomes	Discussion	Comment
Abendroth[17], 1999, USA	Quasi-experimental repeated mesure design – parents with HM from neonatal intensive care (n=52) and without (n=52) Interviews	To compare emotional distress and family functioning of parents whose infant use home monitor with parents who do not use it and to describe parental experience with home apnea monitor	Emotional distress= anxiety, depression and hostility scores measures by the Multiple Affect Adjective Checklist (MAACL) Family functioning= Family Assesment Device Depression increased 2 weeks after discharge in the group with HM and decreased in the other. Hostility increased in the group with HM where it remained stable in the other. Both emotions decreased after. No change occurred in family functioning	Small number of children Stress maybe results from multiple roles if the mother works – no information on it	
Baker, 2000[18], USA	Intervention of a medical social worker among 8 parents who were not compliant with the HM A-B-A design	To promote greater compliance with home monitoring	Intervention=Initial face-to-face, interviews providing the role of social worker in helping the use of the monitor, intervention of social worker to overcome barriers to compliance if needed + behavioural prompting (phone calls) Use of monitor higher during intervention (number of days and hours per day)		

Author, date, country	Type of study (sample/ methodology)	Focus	Results/outcomes	Discussion	Comment
Cabone, 2001[19],USA	39 premature, 13 siblings of SIDS, 16 ALTE of 2 months (corrected age) retrospective review of the first month's recording	Compliance with the use of cardiorespiratory moitoring at home based on the diagnostic indication of use	Parents of infants with ALTE or prematures were compliant (20h/day) Parents of siblings of SIDS used monitor less hours No effect of the SES	Different emotion in sids families than in others Confort of the low probability of a recuring SIDS More likely tohold and directly observe the infant in lieu of monitoring	Refrence to silvestri 1995
Kurz, 2002[20], Austria	90 questionnaire of IC families 70 questionnaires of control families	To evaluate the effects of an intensive counselling program on technical problems with handeling home monitor.	IC leads to a better use of monitor and decreases stress: less stressed and reacted less aggressively, use more often during sleep period reduce the use of monitor (6 weeks)	Parents with IC are more rapidly reassured + medical and psychological support. No change in anxiety because anxiety was pre-existent due to the circumstances that lead to monitor infant	Standard process: team explained the result of the PSG, he indication for monitoring + infant resuscitation + SIDS preventions measures IC: standard + trained and experience nurse called family 2 days after discharge.

Author, date, country	Type of study (sample/ methodology)	Focus	Results/outcomes	Discussion	Comment
McCaleb, 1996[21], USA	Descriptive study 22 children 8-24 months	To describe characteristics of families with HM, decribbe family function characteristics, describe the relationship between family functioning and coping patterns in these families	Families are high in adaptability and average in cohesion, perceive themselves as coping adequately. Correlation among family functioning score and coping pattern subscale scores		
Silvestri, 2005[22], USA	Infant in the CHIME study (>1 month+Siblings of SIDS, preterm, ALTE) - 5 sites - 2 cohorts (n=527 and n=248)	Frequency of monitor use Sociodemographic factors that influenec monitor use Validity of a model to predict use (information and use in the first week)	The use of the monitor during the first week is the best predictor of subsequent use	Adherence to recommendations should be assessed very early.	Compliance with monitor, no information on emotional distress or family consequences Recommendation to identify families who need more support to favour using monitor

Author, date, country	Type of study (sample/ methodology)	Focus	Results/outcomes	Discussion	Comment
Williams et al, 1999[23], USA	Comparative longitudinal study. 28 caregivers of preterm infants discharged with a home monitoring and 46 without. 3 times observation: at discharge – 1 week after- 1month after) + qualitative	To determine differences in fatigue in mothers with and without preterm infant under home monitor	Fatigue increased in the group of mothers with monitor while it decreased in the other group Infant under monitoring were more ill at birth Mothers of infants under monitor seems to possess more economic and social resources to cope with the preterm infant	Increasing fatigue is due to prolonged vigilance	
Zotter, 2003[24], Austria	26 heathy symptomeless infants	To rule out the false alarm and shorthen the duration of home monitoring	39/770 alams were classified as true of these, 30 were misinterpreted by parents as false. 15 were trues. On 283 oxygenation alarms, only 2 was true	Short-term event recording can clarify the significance of frequent alarms, reassure parents and shorten the duration of home monitoring	

6 APPENDIX TO CHAPTER VIII

6.1 PROPOSAL TO IMPROVE THE REPORTING SYSTEM FOR CARDIO-RESPIRATORY MONITORING BASED ON COHORT REPORTING

REPORT YEAR (ex 2006) …..

Table I. Number of children who started CER this year : (ex: 2006)

Indication	Monitor with memory	Monitor without memory	Total (A)
1			
2			
3			
4			
5			
6			
TOTAL			

Table II. For children who started CRM the preceding year (ex 2005) (ex: 2005; figures need to match those reported in column 'total (A) *the previous year*) : mean duration of monitoring

Indication	Total Children (Same as A)	Monitor with memory		Monitor without memory		With + without memory)	
		N children	Mean duration (days)	N children	Mean duration (days)	N children	Mean duration (days)
1							
2							
3							
4							
5							
6							
Total							

Explanations:

- Table I and table II do not refer to the same children.(I: started in 2006; II: started in 2005)
- The advantage of this approach is that duration of monitoring can be computed for all children who started during a given year (regardless of the year their finished their monitoring) . Those who started in 2005 are computed with 2005 even if they finished in 2006.

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