BUILDING THE EVIDENCE BASE FOR DISINVESTMENT FROM INEFFECTIVE HEALTH CARE PRACTICES:

A CASE STUDY IN OBSTRUCTIVE SLEEP APNOEA SYNDROME

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For my mother, father, sister and brother

~ From little things big things grow ~

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Thesis Summary

In the early 1990s claims were made that in all areas of health care, "30-40% of patients do not receive treatments of proven effectiveness",[1] and, "20-25% of patients have treatments that are unnecessary or potentially harmful".[2] Many such practices were diffused prior to the acceptance of modern evidence-based standards of clinical- and cost-effectiveness. I define *disinvestment* in the context of health care as the processes of withdrawing (partially or completely) resources from any existing health care practices, procedures, technologies or pharmaceuticals that are deemed to deliver little or no health gain relative to their cost, and thus are not efficient health resource allocations. Arguably disinvestment has been central to Evidence-Based Medicine (EBM) for well over a decade yet despite general advances in EBM, this topic remains relatively unexplored. This thesis examines the ongoing challenges that exist within the Australian context relating to effective disinvestment. Upper airway surgical procedures for the treatment of adult Obstructive Sleep Apnoea Syndrome (OSA) are used as a case study to contextualise these challenges. This thesis has six sections:

1. A review of the literature outlines developments in EBM broadly and provides a detailed background to OSA, including the numerous treatment options for the condition. This review examines evidence that highlights the importance of 'highly effective treatment' over 'sub-therapeutic treatment' as a necessity to confer improved health outcomes in OSA. It is argued that claims of surgical success inherent in most published results of surgery effectiveness fail to assimilate contemporary evidence for clinically significant indicators of success.

2. Section two comprises the first reported meta-analysis in this area. It presents the pooled success rates of surgery according to various definitions. Specifically, when the traditional 'surgical' definition of success is applied the pooled success rate for Phase I (i.e. soft palate) surgical procedures is 55% (that is 45% fail). However, using a more stringent definition (endorsed by the peak international sleep medicine body), success is reduced to 13% (that is 87% fail). Similarly for Phase II (i.e. hard palate) procedures success rates decrease from 86% to 43% respectively when moving from a surgical to a medical definition of success. That various medical specialties differentially define treatment success, I argue, creates uncertainty for observers and non-clinical participants in this debate (eg policy stakeholders and patients). This represents a barrier to disinvestment decisions.

3. Results are presented from a clinical audit of surgical cases conducted as a component of this thesis. Both clinical effectiveness and procedural variability of surgery are reported. A unique methodology was utilised to capture data from multiple centres. It is the first time such a methodology has been reported to measure procedural variability alongside clinical effectiveness (inclusive of a comparative treatment arm). The observed cohort (n=94) received 41 varying combinations of surgery in an attempt to treat OSA. Results on effectiveness demonstrate an overall physiological success rate of 13% (according to the most stringent definition; phases I and II combined). This demonstration of procedural variability combined with limited effectiveness highlights clinical uncertainty in the application of surgical procedures.

4. Section four outlines how a qualitative phase of enquiry, directed at exploring the perspectives and experiences of surgery recipients, was approved by three independent research ethics review boards but was not supported by a small group of surgeons, resulting in the project being canceled. Potential consequences of this for impeding health services research (HSR) are discussed.

5. Two sets of results are reported from a qualitative phase of enquiry (semi-structured interviews) involving senior Australian health policy stakeholders. The first results are of policy stakeholders' perspectives on the surgical meta-analysis and clinical audit studies in 2 and 3 above. The second results are from an extended series of questions relating to challenges and direction for effecting disinvestment mechanisms in Australia. Stakeholder responses highlight that Australia currently has limited formal systems in place to support disinvestment. Themes include how defining and proving inferiority of health care practices is not only conceptually difficult but also is limited by data availability and interpretation. Also, as with any policy endeavour there is the ever-present need to balance multiple interests. Stakeholders pointed to a need, and a role, for health services and policy research to build methodological capacity and decision support tools to underpin disinvestment.

6. A final discussion piece is presented that builds on all previous sections and summarises the specific challenges that exist for disinvestment, including those methodological in nature. The thesis concludes with potential solutions to address these challenges within the Australian and international context. Systematic policy approaches to disinvestment represent one measure to further improve equity, efficiency, quality of care, as well as sustainability of resource allocation.

Manuscripts Contributing to This Thesis

Published
Elshaug AG, Moss JR, Southcott A and Hiller JE. Redefining success in airway surgery for Obstructive Sleep Apnea: A meta analysis and synthesis of the evidence. **Sleep** 2007;30(4):461-467. [2006 ISI Impact Factor: 5.126]

Published
Elshaug AG, Moss JR, Southcott A and Hiller JE. An analysis of the evidence-practice continuum: Is surgery for Obstructive Sleep Apnoea contraindicated? **Journal of Evaluation In Clinical Practice** 2007;13(1):3-9. [2006 ISI Impact Factor: 1.263]

Letter of acceptance received July 19, 2007 (see appendix four)
Elshaug AG, Moss JR, Maddern GJ and Hiller JE. Upper airway surgery should not be first-line therapy for adult obstructive sleep apnoea (OSA)**. British Medical Journal** Accepted July 19, 2007. [2006 ISI Impact Factor: 9.245]

To be submitted to the Journal of Evaluation in Clinical Practice
Elshaug AG, Hiller JE and Moss JR. Exploring policymakers' perspectives on a clinical controversy: airway surgery for adult OSA. To be submitted to the **Journal of Evaluation In Clinical Practice** [2006 ISI Impact Factor: 1.263]

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Statements

This PhD 'by publication' thesis exceeds the minimum standards for PhD by publication set down in the Academic Program rules outlined in the Adelaide Graduate Centre's postgraduate program rules (PhD rule 9.3 and specifications), and the Faculty of Health Sciences PhD by publication guidelines specific to the School of Population Health and Clinical Practice, available at (web link accessed October 1, 2007):
http://www.adelaide.edu.au/health/research/higher/code/SchoolPhDThesisbypublicationGuidelines.pdf

Choice of journals in this portfolio of publications is justified as follows. *SLEEP* is the official journal of the American Academy of Sleep Medicine (AASM) and is considered the leading international subject-specific journal in sleep medicine (2006 impact factor: 5.126). The *Journal of Evaluation in Clinical Practice* (2006 impact factor: 1.263) was the first and preferred choice for the second research publication (clinical audit), and for the report of stakeholder perspectives on the surgery case study (to be submitted). This is a highly regarded international journal of health policy, evidence based medicine and health services research. The *BMJ* (impact: 9.245) paper was commissioned by a BMJ editor (based on an idea by A/Prof John Moss and a subsequent submission by me) for their *Change Page* series. The *International Journal of Technology Assessment in Health Care* (impact: 1.151) was a strategic choice (first and preferred choice) for publication of the stakeholder engagement regarding disinvestment. It is the official journal of Health Technology Assessment International (HTAi) and addresses a diverse international audience of health care providers, decision makers in government, industry and health care organisations as well as diverse scholarly disciplines such as economics, psychology, ethics, sociology, law etc. Finally, *Australia and New Zealand Health Policy* (no impact factor) also represents a strategic choice for the final publication. It is a BioMed Central, peer-review, online open access journal aiming to improve interaction between policy practitioners and academics, and to promote debate and understanding about contemporary health policy developments in the Australasian region, while maintaining international relevance. There are no other Australian or New Zealand journals with this focus.

Signed:	_____	_____	_____
	Adam Elshaug (Candidate)	A/Prof John Moss (Principal Supervisor)	Prof Janet Hiller (Co-Supervisor)
Dated:	_____	_____	_____

This work contains no material which has been accepted for the award of any other degree or diploma in any university or other tertiary institution and, to the best of my knowledge and belief, contains no material previously published or written by another person, except where due reference has been made in the text.

I give consent for this copy of my thesis, when deposited in the University of Adelaide Library, being available for loan or photocopying.

Signed: _____
Adam Elshaug (Candidate)

Dated: _____

Conference Presentations Arising Out of This Thesis

Forthcoming - Abstract accepted for oral presentation: 5th Health Services Research Association of Australia and New Zealand (HSRAANZ) conference. December 2-5, 2007, Auckland, New Zealand. **Elshaug AG**, Hiller JE and Moss JR. *Exploring Policymakers' Perspectives on Disinvestment from Ineffective Health Care Practices.*

Forthcoming - Abstract accepted for oral presentation: The Menzies Centre for Health Policy, Emerging Health Policy Research Conference. Canberra, Australia, October 12, 2007. **Elshaug AG**, Hiller JE and Moss JR. *Disinvestment from Ineffective Health Care Practices: An Exploration of Policymakers' Perspectives.*

Invited panel member (one of four), Surgery for OSA. 2-hour symposium. WorldSleep07 – International Congress of the World Federation of Sleep Research and Sleep Medicine Societies (WFSRSMS), Cairns, Australia, September 6, 2007.

The Menzies Centre for Health Policy, Emerging Health Policy Research Conference. Sydney, Australia, October 13, 2006. **Elshaug AG**, Hiller JE and Moss JR. *Stuck with the old and overwhelmed by the new: challenges in the policy process for the disinvestment of non-efficacious health care practices in Australia.*

The 6th International Conference on Priorities in Health Care. Toronto, Canada, September 20-22, 2006. Abstract #P-01, page 49. **Elshaug AG,** Hiller JE, Southcott AM and Moss JR. *When clinical practices vary, best practice is one of multiple variations: need for a formal mechanism to monitor effectiveness.*

Canadian Association for Health Services and Policy Research (CAHSPR) Annual Conference, Vancouver, Canada, September 17-19, 2006. **Elshaug AG**, Hiller JE, Southcott AM, Moss JR. '*When clinical practices vary, best practice is one of multiple variations: a practical analysis of the 'know-do-gap'.*

Health Technology Assessment International (HTAi) 2006 Annual Meeting, Adelaide, Australia, July 2-5, 2006. **Elshaug AG**, Hiller JE, Southcott AM, Moss JR. '*An Audit of Surgical Intervention for Obstructive Sleep Apnoea: Questions of Efficacy and Improved Health Outcomes'.*

4th Health Services and Policy Research Conference (Health Services Research Association of Australia and New Zealand). Canberra, Australia 2005. **Elshaug AG,** Hiller JE and Moss JR. '*When clinical practices vary, best practice is only one of multiple variations: need for a formal mechanism to monitor effectiveness'.*

Public Health Association of Australia, Australasian Faculty of Public Health Medicine and Australian Health Promotion Association joint conference 'Public Health in the Community', Adelaide, Australia, October 22, 2005. **Elshaug AG,** Hiller JE and Moss JR. '*The need for a formal mechanism to monitor effectiveness in health care: questions of efficacy and improved health outcomes'.*

Invited Addresses Arising out of This Thesis

Government/Policy - International

4-hour Round Table Forum on *Disinvestment* (with 8 senior government health policy advisors). Strategic Policy and Research Branch, Ministry of Health, Victoria, British Columbia, Canada. September 15, 2006.

Strategic Policy and Research Branch, Ministry of Health, Victoria, British Columbia, Canada. *Stuck with the old and overwhelmed by the new: challenges in the policy process for the disinvestment of non-efficacious health care practices in Australia*. September 15, 2006.

Institute of Health Economics, Edmonton, Alberta, Canada. *Disinvestment from ineffective health care: challenges for Australian health policy processes*. September, 2006.

Government/Policy - Australia

Australian Safety and Efficacy Register of New Interventional Procedures – Surgical (ASERNIP-S), Australasian College of Surgeons. *Building the evidence base for disinvestment from ineffective or inappropriately applied health care practices.* September 27, 2007.

TRACsa Trauma and Injury Recovery, Government of South Australia. *Disinvestment from ineffective health care: barriers, carriers and stakeholder engagement*. July 31, 2007.

Adelaide Health Technology Assessment (AHTA) Retreat and Planning Day. "Disinvestment from ineffective health care: barriers, carriers and stakeholder engagement". July 27, 2007.

South Australian Government Department of Health, Adelaide, Australia. *Stuck with the old and overwhelmed by the new: challenges in Australian policy processes for the disinvestment of ineffective health care practices*. May 14, 2007.

National Institute of Clinical Studies (NICS), Melbourne, Australia. *Stuck with the old and overwhelmed by the new: challenges in Australian policy processes for the disinvestment of ineffective health care practices*. February 13, 2007.

Invited discussion with advisors to the Australian Medical Services Advisory Committee (MSAC). Australian Department of Health, Canberra, Australia. November, 2005.

Clinical Medicine

Forthcoming: Medical Grand Round presentation. The Repatriation General Hospital, Adelaide. *Reporting on a clinical controversy: upper airway surgery for obstructive sleep apnoea (OSA) – results from a meta analysis and South Australian based surgical audit*. October 10, 2007.

Royal Adelaide Hospital multidisciplinary meeting (FRACP/FRACS), Adelaide. *Utilizing patient perspectives in the planning of clinical programs and health policy: a research study*. 2006.

Royal Adelaide Hospital multidisciplinary meeting (FRACP/FRACS), Adelaide. *The fourth hurdle: Cost-effectiveness and efficacy in the regulation of medical services.* 2006.

Invited Addresses Arising out of This Thesis, Continued

Academia

Forthcoming – Discipline of Public Health, The University of Adelaide. *Towards quality and sustainability: building the evidence base for disinvestment from ineffective health care.* Invited Seminar Series Presentation.

Centre for Health Economics Research and Evaluation (CHERE), University of Technology, Sydney. *Health services research and the proliferation of sleep medicine: questions of resource allocation, treatment efficacy & health outcomes?* 2006.

Department of Public Health, The University of Adelaide. *A clinical and policy investigation to support the disinvestment of ineffective health care services in Australia*. PhD progress presentation. 2005.

Department of Public Health, The University of Adelaide. *A clinical and policy investigation to support the disinvestment of ineffective health care services in Australia*. PhD establishment presentation. 2004.

Awards and Merits Arising Out of This Thesis

Merit Award from the World Federation of Sleep Research and Sleep Medicine Societies (WFSRSMS). $1,500 prize, invited address and symposia panellist at WorldSleep07, the 5th World Congress of the WFSRSMS, 2007.

The University of Adelaide (Faculty of Health Sciences) Travelling Fellowship Award (competitive). $2,000 travel award for international conferences and institutional visits. 2006 (Canada)

Best Presentation by a New or Emerging Researcher: 4th Health Services and Policy Research Conference (Health Services Research Association of Australia and New Zealand). Canberra, Australia, 2005

Best Presentation (Respondent's Prize): 'Public Health in The Community' Joint Conference of the Public Health Association of Australia, Australasian Faculty of Public Health Medicine and Australian Health Promotion Association. Adelaide, Australia, 2005

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"*Free should the scholar be – free and brave.*"

Ralph Waldo Emerson

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Finally, I dedicate this thesis to my family, who I moved from to attend University. As the first and only family member to have done so, this submission represents an achievement for us all. To my father, who sold personal belongings to pay for my first-ever semester fees, and who sat and helped me choose my first semester subjects – I'm not sure who was more daunted? To my beautiful mother, whose inner force, love and encouragement have strengthened my development from child to adult. To my big brother, my oldest friend. As a long-distance truck driver travelling through Adelaide he would stop in for a cuppa and to shoot the breeze. Later that night, as he would be half way home on a dark country road I would happen upon a twenty dollar note stuffed hastily into my toothpaste jar, or under my pillow, 'just to see me through'. And to my baby sister, whose tear-filled eyes I still remember gazing back at me from the car as they drove away, leaving me for the first time, alone in a new city. If successful with this submission I will become the first Dr Elshaug in Australia. I now have two nieces and three nephews - may this accomplishment light their way.

Glossary of Acronyms

AHI	apnoea/hypopnoea index (Note US spelling: apnea/hypopnea)
AI	apnoea index
ART	assisted reproductive technologies
BMI	body mass index
BP	blood pressure
CI	confidence interval
CPAP	continuous positive airway pressure
CVD	cardiovascular disease
EBM	evidence-based medicine
ENT	ear, nose and throat
GA	genioglossus advancement
GAHM	genioglossal advancement with hyoid myotomy and suspension
GBAT	genial bone advancement trephine
HREC	Human Research Ethics Committee
HRQL	health related quality of life
HRQoL	health related quality of life
HS	hyoid suspension
HSR	health services research
HTA	health technology assessment
ICU	intensive care unit
IVF	in vitro fertilisation
LAUP	laser-assisted uvulopalatoplasty
MAD/S	mandibular advancement device/splint
MI	myocardial infarction
MMA	maxillomandibular advancement
mmHg	millimetres of mercury
MSAC	medical services advisory committee
NHMRC	national health and medical research council
NICE	national institute for clinical excellence
NICS	national institute for clinical studies
OA	oral appliance
OSA	obstructive sleep apnoea syndrome (Note US spelling: apnea)
PBAC	pharmaceutical benefits advisory committee
PBMA	Program Budgeting and Marginal Analysis
PSG	polysomnography
RCT	randomised controlled trial
RFVR	radiofrequency volume reduction of soft tissue
SaO2	oxyhaemoglobin saturation
SAQLI	sleep apnoea quality of life index
SD	standard deviation
SDB	sleep disordered breathing
SE	standard error
SF-36	short-form – 36
SAHS	sleep apnoea hypopnoea syndrome
TCRF	temperature-controlled radiofrequency volumetric reduction
UK	united kingdom
UPPP	uvulopalatopharyngoplasty
US/USA	united states of america
WMD	weighted mean difference

Chapter One

Introduction and Literature Review

None of the arts theorise about individual cases. Medicine, for instance, does not theorise about what will help to cure Socrates or Callias, but only about what will help to cure any or all of a given class of patients. This alone is business: individual cases are so infinitely various that no systematic knowledge of them is possible.

Aristotle. *Rhetoric*. book I, chapter 2: 1356b

1. Introduction

In the area of health services delivery the translation of research evidence into clinical practice can often be a lengthy, fractured and imprecise process. The complexity involved in effectively translating evidence into practice has seen the science of evidence-based medicine (EBM) develop as a central theme in health services research (HSR). The overarching premise of EBM purports that patient care and outcomes, as well as efficiencies in health service provision, could be significantly improved if the knowledge gained from research was better translated into practice.

In this thesis Obstructive Sleep Apnoea Syndrome (OSA) will be used as a case study to explore appropriateness of care and associated policy approaches if deficiencies are identified in optimal clinically effective care. OSA is a condition in which repeated upper airway obstructions during sleep lead to hypoxia, repeated sympathetic discharge, increased cardiac load, and repeated neurological arousal. Over the past 30 years, increasing recognition of the short and long-term impacts of this disorder has been paralleled by increased demand on various clinical services to diagnose and treat not only OSA itself, but also health conditions and outcomes that are independently associated with OSA.

OSA has therefore been selected for case-study investigation based on a number of important factors, including the prevalence amongst Australia's ageing population, the severity of its adverse consequences and the costs to third party payers. Furthermore, OSA is a health condition that reciprocally influences, and is influenced by co-morbid health states. Consequently, numerous medical specialties come into contact with OSA sufferers. This occurs not only in the diagnosis and treatment of known or suspected cases but also somewhat unknowingly when associated health risks, sequelae and outcomes (such as obesity and cardiovascular disease [CVD]) are managed in undiagnosed cases. Contact potentially occurs through General Practitioners; Respiratory Physicians; Cardiologists; Endocrinologists; Ear, Nose and Throat Surgeons; Obesity Surgeons; Orofaciomaxillary Surgeons and increasingly, Dentists and associated dental specialists. OSA therefore represents a health condition that is potentially serviced by a fractured care model, perhaps to a limited degree in diagnosis but certainly and

increasingly with regard to treatment 'options'. It is therefore important to ensure that treatments are in accord with best evidence for effectiveness, safety and cost-effectiveness. Currently, this cannot be categorically claimed in relation to OSA.

Building on this case study analysis, the primary impediments to the effective translation of evidence into practice in this particular field will be examined. That is, why is there a lack of guidelines and a limited push for them? Is there a genuine lack of good evidence or conflicting evidence? Is there difficulty in overcoming complex operational paradigms? These may relate to prevailing clinical autonomy, disincentives or to perverse incentives related to payment structures among other factors.

These factors will be explored within a framework of disinvestment. That is, what policy mechanisms exist to address the existence of potentially ineffective, less than effective, or inappropriately applied health care practices or technologies? The current project will investigate the policy and clinical practice implications associated with fractured care and produce research, practice and policy recommendations based on these findings. In an era of rising chronic disease prevalence, the complex nature of co-morbid health management suggests that there is increased potential for fractured or inappropriate care to occur. The need for comprehensive assessment of specific areas in healthcare will formally identify evidence-practice gaps, reasons why such gaps might exist, and guide the translation process to improve policy approaches for the effective and efficient delivery of healthcare.

2. Evidence-Based Medicine and Health Services Research

Recent issues of the *Medical Journal of Australia* [2004; 180(6 suppl)] and the *British Medical Journal* [2004; 328(7438)] re-visited in considerable detail a question that continues to raise high levels of interest and controversy. That is, 'how much mainstream medical care is based on the best available scientific knowledge about what does or doesn't work?' This question is not a new one and has influenced the life work of influential members of the scientific community, notably Kerr White, (a US physician who pioneered the discipline of HSR; Archie Cochrane, (the UK physician and medical researcher whose work led to the development of the Cochrane Collaboration, which has set up agreed methods for the systematic review of medical and health related treatments); and David Sackett, a pioneer of EBM. EBM and HSR have since developed into fundamental disciplines within modern healthcare.

It is increasingly acknowledged that the translation of research evidence into clinical practice can often be a lengthy, fragmented and imprecise process. This is particularly pertinent given research findings that suggest 30-40% of patients do not receive treatments of proven effectiveness and that 20-25% of patients have treatments that are unnecessary or potentially harmful [3] [4]. If accurate then these figures are indeed alarming. The Dartmouth Atlas of Health Care project in the United States of America documents serious defects in the quality of care now provided in the fee-for-service medical system. One member of this collaboration has stated:

> *There is substantial overuse, under use, and misuse of medical care in the United States. Interventions that are of little value are commonly overused; care that is effective is commonly underused; and care that is of unproved value is frequently misused. Spending on medical interventions continues to increase without evidence that doing more results in better outcomes or better patient satisfaction* [5].

In light of this situation, certain questions arise such as, what impedes the translation of evidence into practice and how can this situation be overcome? Attempts to address these seemingly fundamental questions now occupy a central theme for a significant portion of the healthcare community (clinical, academic and bureaucratic), either directly or indirectly. However, as one

might expect from this seemingly intractable scenario the answers are complex, and perhaps initially require the questions to be broken down into components, which include but are not limited to the following (in no specific order):

1) Is there a lack of quality evidence from which to base decisions that will effect change?
2) Is there conflicting evidence making the decision process problematic?
3) Is there sound evidence but ineffective means of communicating that evidence to the necessary audience (be it clinicians, policy makers or patients)?
4) Are prevailing practices (including those that are less than effective) occurring as a function of clinical autonomy and therefore difficult to overcome?
5) Are prevailing practices (including those that are less than effective) occurring as a function of clinical autonomy and therefore undesirable to overcome? (Indicating a calculated reluctance by policy makers etc to impinge on clinical judgement or the doctor/patient relationship)
6) Do change agents face difficulty in getting their ideas onto the professional agenda?
7) Are established medical practitioners trapped within an existing Kuhnian paradigm [6] that perhaps only new entrants to the field can escape?
8) What is the role of incentives and/or disincentives (including perverse incentives) in relation to the payment structure(s) for particular medical services?
9) Are patients allowed, and evoking the right to choose between various treatment options (of varying effectiveness)?
10) From (7) what is the role of Supplier-Induced Demand in this choice?

Considerable ground has been made in answering the questions listed above. Indeed, countless books, dedicated journals and journal articles have contributed a wealth of research and understanding in this field. Importantly, there is no simple answer for any one of these questions in isolation. The process depends on many variables related to the health condition under examination and factors that surround it, including the risk profile associated with the condition, prevalence and incidence rates, health outcomes of treating versus not treating and, how expensive and effective are the associated health technologies. Obstructive Sleep Apnoea

Syndrome (OSA) represents an interesting and highly relevant case for investigation based on many of these variables, as will be discussed in later segments of this chapter. Within the Australian context the Chief Executive Officer of the National Institute for Clinical Studies (NICS) has suggested,

> *We do not know how much of the total healthcare Australians receive is based on the best available evidence; studies of a number of specific conditions show that there are gaps between what is known and what happens in practice* [7].

Underlying this is the question: to what degree should policy directives guide clinical practice (based on best available evidence) versus to what degree should clinicians 'on the ground' be left to effect change if and when they see fit, either individually or collectively? That is, should clinicians be responsible for recognising, adopting and translating research evidence into practice or should there be policy guidance? Ferlie and Shortell [8] have suggested four levels at which interventions to improve the quality of healthcare might operate:

- The individual health professional
- Healthcare groups or teams
- Healthcare organisations
- The larger healthcare system or environment in which individual organisations are embedded (policy directives)

Historically, the dissemination of research evidence into healthcare has largely relied on publication in peer-reviewed journals, and on presentation at conference seminars and associated medical education programs (aimed at the individual health professional or healthcare group). However, the effectiveness of these approaches is continually questioned given the overwhelming proliferation of research evidence available. It has been suggested that 10, 000 new randomised trials are included in MEDLINE every year [9] and 350, 000 trials can be identified in the Cochrane Collaboration (Cochrane Collaboration). In this environment there is clear potential for research and receiver capacities to be compromised, and indeed Buchan [7] has suggested 'bridging the evidence gap will not be achieved simply by informing clinicians about the evidence'.

Debate therefore continues as to whether or not research evidence dissemination and implementation mechanisms should be the domain of policy-makers or left to clinicians. Currently in Australia it could be said that a combination of approaches prevails. Concern has been expressed worldwide (predominantly by clinicians) that too much policy direction will erode and compromise clinical autonomy and the doctor/patient relationship [10]. This issue will continue to be debated however in the present context this matter is immediately relevant as clinical practices related to OSA will be under the spotlight. If, as expected, research findings highlight that certain practices related to the treatment of OSA are in fact ineffective and/or inefficient, then questions will arise over the judicious translation and implementation of research evidence and the use of health resource allocations in this context.

2.1 Economic Considerations in EBM and HSR

As presented, the primary goals of EBM are to provide safe and effective healthcare for the community. However, in EBM and HSR the goals of expenditure control, equity and efficiency are also central considerations that maintain a fundamental role. Internationally, there is emerging consensus that reimbursement in public and private health care systems should be informed by evidence of the cost-effectiveness of certain technologies [11 12]. It is suggested that evaluation is an essential component of this process, yet in many areas, including those related to health conditions of burgeoning prominence, evaluation remains as essential as it is rare [13].

The potential over-utilisation of less than effective clinical practices (treatments) and the potential under-utilisation of effective clinical practices not only results in the deprivation of optimal care but also fragmented and inefficient resource allocation. It is argued that such outcomes are not only inefficient, but from a population perspective represent opportunity costs and unethical practice. Health service researchers and advocates of EBM utilise tools such as the Cochrane Collaboration's recommendations for changing professional behaviour as a means of delivering effective, safe and efficient healthcare to the community. Measuring the degree to which this occurs, and developing the means of improving the process continues as a challenge.

2.2 Translation of Research Evidence into Clinical Practice

Barriers and Incentives

Numerous theories and models of evidence translation point to factors that may affect the successful implementation of evidence. However, the evidence for the value of these factors in the field is still limited. In general, implementation research and theory is a much less well-developed area than evaluation practice. Most knowledge of barriers to and incentives for change are not derived from well-designed prospective studies, but rather from retrospective studies and theoretical reflections. Most of the theories have common characteristics, and most are not supported by scientific research on their ability to facilitate change in clinical practice [14]. Nevertheless, this field is progressing rapidly and what does exist is useful for identifying potential barriers and promoters for change.

In attempting to categorise the determinants for change, two complementary approaches have been used extensively, the first focusing on characteristics of individual professionals and the second on interpersonal factors and system characteristics, or paradigm characteristics (also necessarily inclusive of individual professionals).

Individual Professionals

In order for individual professionals to effectively implement change, they need to be informed, motivated and perhaps trained in the process. In a comprehensive review of 76 studies on barriers to guideline adherence, Cabana and co-workers [15] used a "professional development model" in which they identified salient factors as barriers to translation. These included a lack of awareness, lack of familiarity, lack of agreement, lack of self-efficacy (i.e. the belief in one's ability to perform a behaviour), low expectancy of favourable outcomes, inertia/lack of motivation, and perceived external barriers beyond the control of individuals. Empirical data showed that lack of awareness and motivation, as well as perceived external factors, were particularly important barriers to adopting guidelines.

Other models describe a 'stepwise change process' that individuals need to undergo in order to modify their behaviour and facilitate the translation process. "Stages-of-change" theories have

been used to distinguish between patients with different degrees of motivation to adopt better lifestyles [16], but are increasingly being used in research of implementation strategies [17]. A recent systematic review of stage-based interventions has found only limited evidence for their effectiveness [18].

Structural/Paradigm Characteristics

Healthcare professionals operate in specific social, organisational and structural environments, involving factors at different levels that may support or impede the effective translation of evidence into practice. The "PRECEDE–PROCEED" model of knowledge facilitation and evidence translation [19 20 17] makes a significant distinction between "predisposing factors" (eg, knowledge and attitudes in the target group), "enabling factors" (eg, capacity, resources, availability of services) and "reinforcing factors" (eg, opinions and behaviour of others). Systematic reviews [21 22] of studies on effective implementation of evidence and guidelines indicate that strategies that take into account factors at all three levels (predisposing, enabling and reinforcing) are the most successful.

Numerous quantitative and qualitative studies have shown that failure to translate evidence into practice involves factors at different levels of the healthcare system (including characteristics of professionals and patients; team functioning; influence of colleagues; organisation of care processes; available time, staff and resources; policymaking and leadership etc) [23 24]. Yet despite these efforts, we still lack the information on how to effectively tailor interventions to produce change. Grol and Wensing [25] highlight the paradigm elements[a] that contribute to resistance to change. These elements are central to advances in evidence to practice translation:

[a] Use of the word 'paradigm' in this context is deliberate as the conceptual model purported by Grol and Wensing builds on the model presented by Kuhn (6. Kuhn TS. *The Structure of Scientific Revolution.* 2 ed. Chicago: University of Chicago Press, 1970.). That is, advancing the processes for evidence to practice translation requires a revolution from an existing, dominant paradigm(s).

Table One (of Chapter 1): Barriers to and incentives for change at different levels of healthcare

NOTE: This table is included on page 10 of the print copy of the thesis held in the University of Adelaide Library.

Source: Grol and Wensing, 2004 [25]

These complexities are highlighted in research that sought to investigate perceived barriers to implementing guidelines on diabetes care, from a survey of physicians in general hospitals in the Netherlands [26]:

Cognitive factors	**Proportion of respondents citing reason (n = 96; 91% response rate)**
Guideline will not be read	44%
Insufficient evidence base	35%
Lack of knowledge of complications	34%
Attitude of physicians	
Guideline too rigid	56%
Use of guideline costs too much time	54%
Don't like imposed activities	50%
Social and organisational context	
No support by management	44%
Disagreement among physicians	35%
Heavy workload of physicians	81%
Lack of necessary staff	46%
Economic context	
No financial compensation	57%

These factors are interrelated and complex yet this overview provides an interesting insight. Of notable absence in this research and indeed much research in this field is the question of perverse incentives associated with payment structures for certain practices, and how they may influence behaviour. Clearly, when planning complex changes in medical practice, potential barriers at various levels need to be addressed. Commentators in this field acknowledge that planning needs to take into account the innovations available, characteristics of the professionals and patients involved, and the social, organisational, economic and political context. However, limited attention appears to have been paid to specifics of the health condition under question and complexities that are associated with it. This is particularly, and increasingly relevant in the context of chronic disease and co-morbid health management, as will be discussed further.

3. Case Study: Introduction to Obstructive Sleep Apnoea Syndrome

Obstructive Sleep Apnoea Syndrome (OSA) is a condition in which repeated upper airway obstructions during sleep lead to acute adverse effects, including hypoxia, repeated sympathetic discharges, cortical (neurological) arousal, increased cardiac load and significant sleep fragmentation. Over the last 30 years there has been a growing recognition of the widespread short and long-term impacts of this disorder, linking OSA to cognitive, behavioural, cardiovascular and cerebrovascular morbidities [27] [28] [29]. This recognition has been paralleled by increased demand on clinical services to diagnose and treat OSA. The public health importance of OSA arises from the disease burden among our aging population, from the severity of its adverse consequences, and from the extent of the costs to third party payers.

OSA Prevalence

According to numerous cross-sectional studies, the prevalence rates for OSA among adults range from 2-4%, increasing with age [30]. Some controversy has been documented over prevalence estimates, based largely on the variance between clinical parameters used to define a positive case of OSA. More recent prevalence estimates based on larger samples have produced larger estimates. In 1993, Young et al [30] reported that in the USA, undiagnosed sleep-disordered breathing is present in 9% of women and 24% of men of middle age (i.e. age 30-65 years). Olsen

et al [31] found similar results in Australia. Interestingly, comparison of the male to female ratio in diagnosed OSA patient populations (8:1) varies considerably to that in undiagnosed OSA from population studies (2:1) [32]. This points to strong selection bias (towards males) when it comes to evaluation and diagnosis of the condition.

3.1 OSA: Risk Factors

There is a positive correlation between having OSA and increasing body mass index (BMI: weight/height in metres squared), neck circumference and waist-to-hip ratio. There is a high prevalence of OSA in obese individuals and a high prevalence of obesity in patients with OSA. OSA also occurs in non-obese individuals however excess weight is an independent causal factor [33]. A cohort analysis (4 years) demonstrated that a 10% increase in body weight was associated with a 6-fold greater risk of developing OSA in persons previously free of the condition [34]. Similarly, several small studies have demonstrated consistent and substantial reductions in OSA severity following surgical and/or dietary/behavioural weight loss interventions [32].

Cranio-facial and upper airway structure have also been identified as playing a role in the risk profile for OSA development, with evidence suggesting this is of particular significance for Asian populations [35] [36]. Furthermore, ongoing research seeks to identify subgroups of patients for whom skeletal or soft tissue abnormalities play a role in the development of the condition. These abnormalities include dysmorphisms related to mandibular or maxillary size and position, narrowed nasal cavities, and tonsillar hypertrophy. The population prevalence of these conditions is unknown. Similarly, there is uncertainty as to whether enlarged adenoids and tonsils during childhood may cause abnormal growth in the lower face and jaw and hence predispose one to OSA in later life. Of even greater controversy is whether surgical intervention can adequately correct these conditions and therefore successfully treat OSA [37]. This matter will be addressed further.

Research is also developing in the area of hormone and gender differences in relation to OSA prevalence, with most epidemiologic studies in this area focusing on the critical role of the menopause. In an analysis of mid-life women in the population based Wisconsin Sleep Cohort Study, postmenopausal women had 3 times the odds of having moderate or worse OSA

compared with pre-menopausal women, independent of age, body mass index, and other potential confounding factors [38]. In a Pennsylvania population-based cohort of 1000 women, a 4-fold greater risk of OSA was found in postmenopausal women not using hormone therapy vs. pre-menopausal women. In this sample, OSA risk increased with duration of menopause up to 5 years post menopause [39]. These findings support the hormone depletion hypothesis, suggesting hormone therapy may be protective for OSA in postmenopausal women. Findings from the Sleep Heart Health Study of 2994 women aged 50 years or older showed hormone therapy users compared with nonusers had half the odds of OSA [40]. However, in a blinded randomised trial involving postmenopausal women, Polo-Kantola et al [41] found only a weak effect of hormone therapy in reducing OSA. Research in this area is continuing.

Finally, acute risk factors for OSA include alcohol and/or sedative medication consumption whereby the relaxant effect on muscle tone promotes further collapsibility of the airway while asleep or while undergoing anesthesia-induced surgery where there is considerable peri-operative risk. The effects of long-term consumption of alcohol and sedative substances as a risk factor for OSA are unknown [42]. Finally, sleep in the supine position (on back) contributes to OSA in many sufferers as the force of gravity increases airway collapsibility.

3.2 Health Outcomes / Co-morbid Conditions

OSA has been associated with diabetes, hypertension, cardiovascular disease, myocardial infarction, congestive heart failure, and stroke and is independently associated with an increased risk of mortality [27] [28] [29]. From retrospective and matched control studies, mortality appears to correlate with the severity of sleep disordered breathing and is highly influenced by other co-morbidities [43] [44] [45]. Sleep-disordered breathing been identified as an independent risk factor for the development of co-morbidities such as pulmonary and systemic hypertension [46] [47] [48] [49] and cardiovascular events [50] [51]. In a comprehensive report, Young et al [32] found that people with untreated OSA carry a five-fold risk of hypertension, a four-fold risk of myocardial infarction (with an odds ratio as high as 23.3 for severe OSA sufferers), and a ten-fold risk of stroke. Untreated OSA also impairs neuropsychological performance (global intellectual dysfunction, deficits in vigilance, alertness, concentration, short- and long-term memory, and executive and

motor function), and hence significantly reduces Health Related Quality of Life (HRQL) as measured by the SF-36 and the Sleep Apnoea Quality of Life Index (SAQLI) [52].

The correlates of OSA, including excess body weight and hypertension, overlap with those of diabetes mellitus. Reports that OSA is associated with insulin resistance and other factors related to the metabolic syndrome are increasing however the time-ordering remains unclear [53]. Therefore, whether or not there is a causative role of OSA in the metabolic syndrome is uncertain, and at present important research in this area is being undertaken.

Other associations that have been documented include gastro-oesophageal reflux disease, depression, impotence and increased peri-operative anaesthetic risk (collapsing airway during surgery). Untreated sleep disordered breathing has also been associated with increased risk of motor vehicle accidents [54] [55]. A case-control study conducted in Spain demonstrated that the odds ratio of having a traffic accident for OSA sufferers was 6.3 compared to those without the condition [56].

In summary, OSA is associated with diabetes, hypertension, coronary artery disease, myocardial infarction, congestive heart failure, and stroke. The associations may be due in part to risk factors common to all these conditions; they may also reflect a role of OSA in the aetiology of these conditions.

Health Service Utilisation

In a medical records study it was found that, compared to matched controls, patients with undiagnosed OSA consume twice the health resources (hospitalisation stays, physician costs, mean annual medical costs etc) in the 10-years prior to diagnosis [57] [58]. Bahammam [59] subsequently reported that this trend is reversed following successful treatment. For example, physician costs fell by 33% and duration of hospital stays for OSA patients decreased from 1.27 days ± 0.25(SE) per patient per year, one year before diagnosis to 0.54 ± 0.13 per patient per year following treatment (p=0.01). Importantly, these differences were only significant in patients who adhered to what was termed, ‘effective’ treatment (specifically, Continuous Positive Airway Pressure: CPAP). These figures demonstrate the complex but significant co-

morbid nature of OSA as a health condition, and the potential for improved health outcomes and efficiency in many areas of healthcare following the effective treatment of the condition.

4. OSA: Evaluation and Diagnosis

Polysomnography (PSG) is the best laboratory procedure for studying sleep and its dysfunctions, including OSA. PSG involves a wire-up procedure that requires the patient to stay overnight in a sleep laboratory where ranges of neurological and cardio-respiratory variables are monitored. The diagnosis of OSA is based on the number of breathing abnormalities that occur per hour of sleep (known as the apnoea/hypopnoea index: AHI). Apnoea represents the full cessation in breathing whereas a hypopnoea is a partial cessation. The scoring of events takes into account the degree of oxygen desaturation, and associated respiratory-related arousals from sleep that occur. According to the American Academy of Sleep Medicine (AASM), the peak international sleep medicine body, the AHI cut points of 5+ (mild), 15+ (moderate), and 30+ (severe) are used to indicate OSA severity [60 61]. An AHI of 5 or less is deemed within normal limits and confers a negative diagnosis.

4.1 OSA: Clinically Important Endpoints

At this point it is necessary to focus briefly on the AHI as it represents a clinically important endpoint in terms of treatment effectiveness. An increasing number of contemporary research findings now demonstrate the importance of reducing the AHI to near or below five events per hour of sleep (and therefore controlling OSA) in order to improve numerous physiological, health outcome and quality of life measures [62-77]. These findings are the result of numerous large-scale cohort studies from which long-term follow up is only now providing meaningful information to help define treatment effectiveness. This point cannot be understated as to date, research into the ‘effectiveness’ of various treatment modalities has concentrated on varying degrees of reduction in AHI. Importantly however, research now indicates that a significant reduction in AHI (albeit a statistically significant reduction) does not necessarily confer improved health outcomes unless the reductions achieved are of the extent that OSA is adequately controlled. Evidence of this will be presented in the following sections on treatment effectiveness.

4.2 OSA: Treatment Modalities

Modern sleep medicine has been in existence for only 25 years and may therefore be regarded as a comparatively recent field of specialisation. For this reason it is not surprising that there are numerous new developments concerning the treatment of OSA. Treatment modalities for OSA fall into two broad categories: 1) weight loss related, either by dietary/behavioural, pharmacological or surgical intervention; 2) medical intervention, including CPAP, Oral Appliances (OA) such as the Mandibular Advancement Splint/Device (MAS/D) and surgical procedures of the soft palate/upper airway, such as the uvulopalatopharyngoplasty [78]. Generally, surgery for weight loss occurs independently of OSA whereas CPAP, oral appliances and surgery of the airway are specific and direct treatments for OSA. The following overview will therefore concentrate on those specific treatment modalities.

Continuous Positive Airway Pressure

Prior to 1981, the only effective treatment for OSA was tracheostomy, a highly invasive medical procedure [79]. However in 1981, an Australian physician developed and implemented a non-invasive positive air pressure delivery interface that effectively treats OSA. CPAP delivers a continuous stream of positive air pressure via tubing to a soft gel nasal, or oronasal mask. This process effectively acts as a pneumatic splint that prevents collapsing of the airway, maintaining airway patency. CPAP thus represents an ongoing treatment for OSA; it is not a cure but a treatment modality that must be worn throughout every sleep period for OSA to be controlled.

Before CPAP use can commence, a process of titration must occur whereby various pressures are trialed in order to identify the optimal treatment pressure required to eliminate respiratory disturbances. CPAP must therefore be titrated during sleep and this requires simultaneous polysomnography. A full-night diagnostic study and a full-night CPAP titration study have generally been the accepted standard of practice. This has led to considerable demand on polysomnographic clinical services to diagnose and treat the condition, and over the last decade

the number of these clinical services has increased markedly in Australia, from 14,308 in 1994 to 66,134 in 2006[b].

This problem of increasingly stretched diagnostic and treatment services is compounded due to the requirement for pressure re-assessment or re-titration. That is, existing CPAP users return periodically for a full night of assessment to gauge the effectiveness of their prescribed CPAP setting. At present this is an important area of potential inefficiency as the guidelines for who returns, when, and for what reasons are vague.

Despite the growing demand for, and provision of this service, a consulting firm recently estimated that in Australia only 10-20% of OSA sufferers have currently been diagnosed and treated for the condition [80]. This estimation was presented along with the statement, "*the current Australian sleep service landscape is fragmented and under-resourced*" (pg. 18) and seeks, amongst other things, to justify calls for increased funding of this sector. This author submits that in the first instance this estimation is poorly qualified with no referential support or direct evidence provided for this claim. And secondly, even if this estimation is accurate, the report says little about potential efficiency gains, and evidence-based directives that exist but are yet to be explored within the sector.

Oral Appliances

In addition to CPAP, oral appliances (OAs) that modify the pharyngeal spaces have also been offered to OSA cases (OAs are similar in construction to joined upper- and lower-jaw mouth guards). During sleep, muscle tone decreases, leading to increased collapsibility of the pharyngeal tissues (upper airway), mandibular opening and posterior displacement of the tongue. These changes result in narrowing and/or occlusion of the oropharyngeal and hypo pharyngeal airway [81], hence OSA. A variety of OAs are available whose primary actions are to advance the mandible or tongue and thus enhance airway patency. Another, less accepted theory explaining

[b] (Data available from: http://www.medicareaustralia.gov.au/statistics/dyn_mbs/forms/mbs_tab4.shtml (Accessed 11/07/2007)

their mode of action, is that OAs cause stretch-induced activation of the pharyngeal motor system, thus reducing soft tissue laxity and airway collapse [82].

Side effects have been reported with the use of OAs including discomfort in the temporo-mandibular joint, teeth or facial musculature, bite change, excessive salivation or dryness of the mouth [83]. These devices are custom-made and hence primarily fitted by orofaciomaxillary surgeons, dentists and dental specialists after a mould of the teeth/jaw is taken.

Surgical Procedures

There is an array of surgical procedures employed either concurrently or stepwise over multiple operations. Surgical treatments for OSA aim to relieve the obstruction by increasing the surface area, to bypass the pharyngeal airway, or to remove a specific pathological lesion. The principal interventions were briefly described by the American Sleep Disorders Association in 1996 [84] and are expanded on here by Li [85] to include:

1. Tracheostomy (which bypasses the pharyngeal airway-used primarily in acute emergencies).
2. Uvulopalatopharyngoplasty (UPPP), which is intended to increase the area of the retropalatal airway by removal of the posterior portion of the uvula and palate and tonsillectomy - an increasingly common modification is laser UPPP. Referred to as laser-assisted (LAUP) when laser is used.
3. Tonsillectomy – primarily for the treatment of OSA in children.
4. Inferior sagittal mandibular osteotomy and genioglossal advancement with hyoid myotomy and suspension (GAHM), aiming to create an enlarged retrolingual airway.
5. Laser midline glossectomy and lingualplasty also aiming to create an enlarged retrolingual airway.
6. Maxillo-mandibular osteotomy and advancement to enlarge both the retrolingual and retropalatal airway.
7. Epiglottoplasty for selected cases of laryngomalacia.
8. Removal of local specific obstructing pathological lesions ie, hypertrophy of the tonsils.
9. Temperature-controlled radio frequency tissue volume ablation (TCRAFTA) – applies energy to the base of the tongue and/or the soft palate.
10. Epiglottoplasty for selected cases of laryngomalacia.

Adding to the list, the Genial Bone Advancement Trephine (GBAT) system is a relatively new, one-step system that allows for isolation and advancement of the genioglossus muscle via a guided trephine system. In Australia, so-called phase I category procedures (UPPP, hyoid

myotomy, palatal surgery, and/or genioglossus advancement) and phase II category surgery (osteotomies) are both widespread (occurring in all states) and increasing year on year. Australian Medicare data allows tracking of procedures provided through private surgical practice (i.e. private surgical clinics and hospitals, not public hospitals). Currently there are over 26 individual Medicare reimbursement item numbers identifying all procedural variants. A search of this database revealed just over 3,500 procedures were performed Australia-wide for the 2006 calendar year. This is an increase from the 3,000 performed in 2004. The UPPP (traditional or laser assisted) remains the most common surgical procedure for the treatment of OSA, accounting for over one-third of all procedures subsidised by Medicare[c]. Note that Medicare does not track procedures performed in state-run public hospitals.

Ear, Nose and Throat (ENT) and/or orofaciomaxillary surgeons carry out all surgical procedures. A recent systematic review of 48 studies (4 randomised trials, 17 prospective designs, 23 retrospective reviews of consecutive patients, 4 of unspecified design) found persistent adverse effects reported in up to 62% of surgery recipients[86]. Specifically, difficulty in swallowing, including spontaneous nasal regurgitation, was introduced (i.e. new after surgery) in up to 29%, globus sensation in up to 36%, voice changes in up to 14%, taste disturbances in up to 7%, smell disturbances in up to 8%, and persistent dry throat in up to 56%. In that systematic review up to 22% regretted surgery however this has been reported elsewhere in up to 61% of recipients[87]. These rates of side effects coupled with the regret rates have implications for patients adhering (i.e. potential non-adherence) to multiple operations (so-called stepwise approach to surgery) as is increasingly recommended by the surgery community as standard practice[88]. It has also been demonstrated that UPPP compromises subsequent CPAP therapy by increasing mouth air leak and reducing CPAP pressure tolerance (non-randomised trial, n=26)[89].

An issue worthy of note is that in Australia these surgical procedures can be carried out without the consultation of a Respiratory/Sleep physician and/or the utilisation of sleep laboratory services to either, 1) initially establish a positive diagnosis of OSA, or 2) subsequently refer on

[c] Medicare data available from: http://www.medicareaustralia.gov.au/statistics/dyn_mbs/forms/mbs_tab4.shtml (accessed July 16, 2007):

to these services post surgery for an 'effectiveness' assessment to be made. The choice to undertake either of these options remains with the treating ENT physician with influence also via the referral pathway from a general practitioner.

4.3 OSA: Documented Effectiveness of Available Treatments

The claim of treatment effectiveness for OSA is an area that invites critique, as it has become clear that the terms 'treatment success' and 'treatment effectiveness' have become loosely defined and are used interchangeably. Available evidence suggests the need for closer scrutiny in relation to term definition and differentiation. This is particularly pertinent when considering health outcomes associated with various treatment modalities.

CPAP

The original Cochrane Review to assess the effectiveness of CPAP in treating OSA was published in December, 2000.[90] This review was updated in 2006 [91]. In these reviews CPAP is overwhelmingly regarded as the gold-standard treatment modality for OSA and remains the most definitive medical therapy available, as it is still regarded as the most consistently efficacious and safe option. This is primarily due to the significant reduction (in most cases elimination) of apnoeic and hypopnoeic events that CPAP therapy produces [91]. Therefore, from a clinical perspective there is a substantive evidence base to support that CPAP has the potential to offer almost 100% effectiveness in almost 100% of cases [92-95].

However, the major disadvantage of CPAP, which impacts its efficacy, is that it does not confer a cure to the disorder and hence therapy is generally life-long with usual problems of treatment compliance (i.e. acceptance and adherence). This modality is accepted worldwide as the most reliable treatment regardless of anatomy and severity, and further, is recommended as the initial intervention protocol even if alternative therapies are sought [90]. Research suggests that long-term CPAP compliance ranges between 60-80% [96 97]. This factor is regarded as a major challenge going forward and considerable effort and expenditure is being invested to overcome this problem. In a recently reported RCT, two one-hour sessions of Cognitive Behaviour Therapy

(CBT) increased CPAP compliance (at 28 days) from 70% (a rate commonly reported) to 92% [98].

Importantly, contemporary research strongly indicates there are clinically significant reductions in cardiovascular [99 62] and endocrinological [72] sequelae related to OSA following CPAP treatment. Furthermore, the treatment of OSA with CPAP has a cost-effectiveness that is in line with that of other commonly funded treatments such as antihypertensive drugs [100].

The use of CPAP treatment has also been shown to significantly improve Health Related Quality of Life (HRQL) measures (global intellectual function, vigilance, alertness, concentration, short- and long-term memory, and executive and motor function) for users [76 70]. Interestingly, bed partners also show improved quality of life measures following their partner's initiation of CPAP treatment [77]. This occurs due to improved sleep; the bed partner is no longer subjected to snoring, snorting and the often-dramatic body movements associated with apnoeic recovery. These improved measures, for both the sufferer and their bed partner, have been observed over both the short (3 months) and long term (12+ months) as measured by the SF-36 and the Sleep Apnea Quality of Life Index (SAQLI).

From an evidence-based perspective, CPAP treatment therefore offers a highly effective outcome, not only in treatment outcome but also health-related outcomes, and cost-effectiveness. Not surprisingly therefore, CPAP is referred to in guidelines as the first-line therapy (with conservative weight and alcohol management) for the treatment of OSA [101 102].

Oral Appliances

Issues of CPAP tolerance and associated non-compliance have led to greater choice in treatment modalities for OSA. In light of this, one must ask how well these alternative modalities address similar criteria to those listed above. That is, clinical effectiveness (reducing AHI to the degree of controlling OSA), cost-effectiveness and improvements in health outcome, i.e. improved cardiovascular and endocrinological outcomes and improved quality of life measures.

The most recent Cochrane Review in this area was published in 2006 [103]. Sixteen studies (745 participants) met the inclusion criteria. All had some shortcomings, such as small sample size, under-reporting of methods and data, and lack of blinding. OA versus control appliances (six studies): OA reduced daytime sleepiness in two crossover trials (WMD -1.81 [scale 0-14]; 95% CI -2.72 to -0.90), and improved AHI (-10.78 [continuous scale]; 95% CI-15.53 to -6.03 parallel group data - five studies). OA versus CPAP (nine studies): OA were less effective than CPAP in reducing AHI (parallel group studies: WMD 13 (95% CI 7.63 to 18.36), two trials; crossover studies: WMD 7.97; (95% CI 6.38 to 9.56, seven trials). However, no significant difference was observed in symptom scores. CPAP was more effective at improving minimum arterial oxygen saturation (SaO2) during sleep compared with OA. In two small crossover studies, participants preferred OA therapy to CPAP. OA versus upper airway surgery (one study): Symptoms of daytime sleepiness were initially lower with surgery, but this difference disappeared at 12 months. AHI did not differ significantly initially, but did so after 12 months in favour of OA. The authors of the Cochrane review concluded [103]:

> *There is increasing evidence suggesting that OA improves subjective sleepiness and sleep disordered breathing compared with a control. CPAP appears to be more effective in improving sleep disordered breathing than OA. The difference in symptomatic response between these two treatments is not significant, although it is not possible to exclude an effect in favour of either therapy. Until there is more definitive evidence on the effectiveness of OA in relation to CPAP, with regard to symptoms and long-term complications, it would appear to be appropriate to recommend OA therapy to patients with mild symptomatic OSAH, and those patients who are unwilling or unable to tolerate CPAP therapy. Future research should recruit patients with more severe symptoms of sleepiness, to establish whether the response to therapy differs between subgroups in terms of quality of life, symptoms and persistence with usage. Long-term data on cardiovascular health are required.*

Surgical Procedures

The place of surgery in the treatment of OSA and the relative effectiveness of different interventions is controversial. In this area of invasive surgery there is a paucity of RCTs. Most studies recommending a particular surgery are based on evidence from case series. Historically, reviews of the surgical therapy for OSA are generally narrative, summarising the evidence provided by case series and uncontrolled observational studies [104]. In the 2005 Cochrane review

eight studies (n=412) met the inclusion criteria with data from seven assessed.[3] Data were not meta-analysed. Results were inconsistent, with statistically significant improvements in PSG outcomes (e.g AHI) reported in only three studies (with limited comment on clinical significance). Statistically significant improvements on validated measures of HRQoL were found in four. CPAP was superior overall, and in one report OA produced a significantly lower AHI than UPPP. The 2004 Cochrane review concluded;

> *In light of the current lack of good trial-based evidence, clinicians should consider restricting surgery for obstructive sleep apnoea to that carried out as part of clinical trials. Where practice is continued, patients should be informed of the experimental nature of the operations.*
>
> *Patients should be told that there is a lack of good trial based evidence of the efficacy of surgery for obstructive sleep apnoea, a course of action that may restrict the use of these operations. There is an urgent need for high quality randomised controlled trials to be carried out in the field of surgery for the treatment of OSA, as there is a complete deficiency of such work.* [37]

The 2005 compendium continues, "*The studies assembled in the review do not provide evidence to support the use of surgery in [OSA], as overall significant benefit has not been demonstrated...*".[89]

In the most recent reporting of surgical efficacy in the peer-review surgical literature, claims of treatment success, effectiveness and even 'cure' of OSA have been defined as a reduction in AHI of greater than 50% and to 20 respiratory events per hour or less [105] [106] [107]. Based on these criteria, Kim and co-workers, as well as Souter et al have found that surgical treatments vary considerably, from effectively treating up to 95% of recipients through to worsening OSA for others [108] [109]. This variability in definitions of treatment success is further apparent in the Cochrane review, where the authors refrain from including the phrase in their reports. Instead, they concentrate on statistically significant improvements with limited note of how these translate to clinically meaningful outcomes. The reporting of quality of life measures goes some way toward this but is very limited. Problematically, for the surgical studies reporting 'success', one important point to recognise is that a reduction in AHI to 20 or less still confers the status of mild to moderate OSA, and the number of recipients/patients who achieve this varies substantially. Therefore, following surgery to treat OSA, a highly variable number of cases

remain OSA positive, and in some cases are worse than prior to surgical intervention. This then has significant implications for health outcomes and furthermore, what may be deemed effective, safe and cost-effective (efficacious) treatment.

5. Potential Gaps in Evidence to Policy to Practice Translation

Sleep medicine is no longer the sole domain of respiratory and sleep physicians and associated support staff, but now includes a variety of alternative specialties who are involved primarily in treating the condition. CPAP is unequivocally regarded as the gold-standard treatment modality available. On the basis of published literature to the year 2000, White [90] suggested there were no studies available to show that CPAP effects the incidence or outcomes of other medical conditions associated with OSA, such as hypertension, cerebrovascular disease, cardiopulmonary disease, or road traffic accidents. In light of this it may have been considered that any treatment that reduced AHI was a good and worthwhile treatment. However, recent evidence strongly indicates this can no longer be claimed, and the implications for clinical policy and practice are considerable. Becker and co-workers provide one landmark example of this:

> *Apneas and hypopneas [AHI] were reduced by approximately 95% and 50% in the therapeutic and sub therapeutic groups, respectively. Mean arterial blood pressure decreased by 9.9+/-11.4 mm Hg with effective CPAP treatment, whereas no relevant change occurred with sub therapeutic CPAP (P=0.01). CONCLUSIONS: Effective CPAP treatment… leads to a substantial reduction in both day and night arterial blood pressure. The fact that a 50% reduction in the apnea-hypopnea index did not result in a decrease in blood pressure emphasizes the importance of highly effective treatment. The drop in mean blood pressure by 10 mm Hg would be predicted to reduce coronary heart disease event risk by 37% and stroke risk by 56%* [62].

As well as cardiovascular disease [63-68], similar results have also surfaced in areas related to heart failure [69,70], endocrinology [71-74], and health-related quality of life [75-77]. All indicate the importance of 'highly effective treatment' with a substantial decrease in the AHI over 'sub-therapeutic treatment' as a necessity to confer improved health outcomes. Recent meta-analyses pooling effects of CPAP on blood pressure show marginal overall effect but highly clinically significant for severe OSA [110 111].

Less so for dental devices but increasingly for surgical treatment modalities, overarching concerns include findings of limited effectiveness and, in addition, the somewhat spurious and misleading presentation of research evidence. Most notably, conclusions of treatment 'success'

and 'cure' are based on insufficient reductions in AHI, a key clinical endpoint. This is fundamentally important because contemporary research now points to a reduction in AHI that effectively eliminates OSA as being required to improve health outcomes such as quality of life, endocrine function and cardiovascular disease outcomes. A distinction must be drawn between clinical significance and statistical significance. Although a reduction in AHI of 50% is indeed significant (including statistically), such a reduction should be interpreted in relation to treatment and health outcomes. A statistically significant finding does not necessarily support claims of an effective or efficacious procedure. To call a procedure successful in this context, where significant symptoms, co-morbid disease and associated poor health may still be present, is potentially misleading and contrary to what may reasonably be considered efficacious clinical practice. This scenario potentially represents a significant gap in the translation of evidence into clinical practice.

6. Research Justification

Numerous reports recommend the restricted use of these surgical procedures for the treatment of OSA, including the original [37] and updated [89] Cochrane reviews, a report of the Scottish Intercollegiate Guidelines Network (SIGN) [102] and the Swedish Council on Technology Assessment in Health Care (SBU) HTA/Nordic Project on Sleep Apnea report [86]. Despite these recommendations, Australian Medicare data indicates the procedures are widespread and their use is increasing. In Australia there is limited policy guidance as to who should be offered various treatment options, under what circumstances, and by whom. These decisions occur according to individual assessments by physicians and/or surgeons. Moreover, we do not currently know how much collective evidence is considered in these decisions, and indeed what the collective outcomes are for many who seek out, or are induced into alternative therapies. The current project will investigate the policy and clinical practice implications associated with fractured care and produce recommendations based on available evidence. In an era of rising chronic disease prevalence, the complex nature of co-morbid health management suggests that there is increased potential for compartmentalised care to occur.

A wealth of research evidence has emerged from numerous specialties regarding the risk profile, health outcomes, treatment options and treatment outcomes for OSA. Overnight polysomnography (diagnosis) and CPAP (treatment) are currently considered gold-standard practices for diagnosis and treatment, respectively. Yet despite this, there is currently no cohesive, structured path for clinicians or patients to follow as an alternative to CPAP in managing this syndrome. There remains a paucity of high-level research evidence into the effectiveness and cost-effectiveness of surgical procedures. The overarching premise of EBM is that patient care and outcomes, as well as health service efficiency scales, could be significantly improved if the knowledge gained from research was better translated into practice.

Evidence suggests that OSA represents a health condition that is potentially serviced by a fractured care model, perhaps to a limited degree in diagnosis but certainly, and increasingly with regard to treatment 'options'. It is therefore important to ensure that treatment modalities are effective and offer tangible improvements in health outcomes, and that resource allocation is efficient. Despite the poor translation of evidence to practice calls continue for increased funding for treatment of OSA [80].

After highlighting the evidential complexities associated with surgery for OSA (as a case study) this thesis will then move on to the larger policy issue of what to do when a particular health care practice or technology is identified as having uncertain or questionable clinical and cost-effectiveness. This concept is increasingly known as obsolescence, referring to ineffective or inappropriately applied health care practices and technologies. Associated with obsolescence is the notion of disinvestment. To disinvest is the process of (partially or completely) withdrawing health resources from these *existing* (as distinct from *new* or *emerging*) health care practices, procedures, technologies or pharmaceuticals that are deemed to deliver little or no health gain for their cost, and thus do not represent efficient health resource allocation.

Considerable effort and resources have been invested in Australia, in developing well-defined criteria and evidence-based policy processes for assessing new and emerging health technologies, surgical procedures and pharmaceuticals to gauge their safety, effectiveness and cost-effectiveness [112 113]. Reimbursement approval (and therefore universal access through

Australia's Medicare system [114]) for these new services, as well as decisions to withdraw reimbursement for existing services rest with the Australian Government Minister for Health and Ageing, under advice from the Medical Services Advisory Committee (MSAC) and, for pharmaceuticals, the Pharmaceutical Benefits Advisory Committee (PBAC). The MSAC and the PBAC are supported by health technology assessment (HTA) groups, employing stringent review processes based on the existence, and quality, of data and evidence that are available at the time of assessment. Underpinning disinvestment, however, is recognition that these stringent assessment methods are relatively novel and that the processes focus overwhelmingly on technologies or practices with new applications for reimbursement/registration within particular jurisdictions and not on existing services (even though this is within the mandate of the MSAC). Australia therefore, like other countries, suffers from a legacy whereby many currently implemented health care interventions were diffused prior to well-defined standards of cost-effectiveness becoming a criterion for reimbursement.

The aim of this project is to apply an investigative, clinical epidemiological framework in healthcare to synthesise available evidence, identify gaps in policy and practice, and hence contribute to, and advance the translation of research evidence into (disinvestment) policy and practice within the Australian context. The project also aims to identify the limitations that currently exist in that process and how these may be transferable to other health conditions and states. Integration of evidence from various specialties to guide effective care (and policy) is increasingly important. The current research project may provide insight to address the complex needs of an ageing population, the rise of chronic disease prevalence and associated co-morbid health management, and the demand they place on the healthcare sector.

7. Research Questions

Does upper airway surgery for obstructive sleep apnoea syndrome represent clinically effective treatment? What are the possible explanations for any gap between evidence, policy and practice, and what are the potential resolutions? Does Australia have established policy mechanisms, including health care reimbursement structures, that can react to and influence existing health care practices with uncertain clinical and cost-effectiveness? Can these policy mechanisms adapt to the ever-changing nature of evidence?

Aims

The aim of this project is to conduct a systematic investigation in health care (using surgery for OSA as a case study) that will synthesise available evidence, identify gaps in and between policy and practice, and advance the evidence base to guide a formal disinvestment policy agenda. This project will offer insights to guide policy processes for disinvestment from ineffective, less effective or inappropriately applied health care practices.

Research is the only hope that the future will be different than the past

Daniel Mintz, MD

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8. Research Program

The literature review in this area highlights uncertainties in the application of surgery for the treatment of OSA. In order to address the specific research questions the following sequence of research projects has been carried out. Extending on from the literature review (chapter one) there are a further eight chapters of the thesis:

Chapter two comprises the first reported meta-analysis of surgery for the treatment of OSA. It complements the Cochrane review (which did not meta analyse results) to include for analysis, level four evidence (retrospective clinical audits). These lack the rigour of the randomised controlled trial (RCT) design but given the complex nature of this invasive surgery, performing RCTs is complex and ethically questionable. Collectively, these level 4 studies provide a wealth of evidence that is often under-utilised in EBM. This meta-analysis demonstrates how various medical specialties differentially define treatment success. I argue this creates uncertainty for observers and non-clinical participants in this debate (eg policy stakeholders and patients), representing a barrier to disinvestment decisions.

In **chapter three** results are presented from a multi-centre retrospective clinical audit of surgical cases conducted as a component of this thesis. Both clinical effectiveness and procedural variability of surgery are reported. The objective of this work is to highlight current practice patterns in Australia given the concerns over efficacy that exist. Cases were sampled from two sleep laboratories in Adelaide; this captured surgery recipients from a pool of over one dozen surgeons within South Australia (both private and public). It is the first time this methodology has been reported to measure procedural variability alongside clinical effectiveness (inclusive of a comparative treatment arm). This demonstration of procedural variability combined with limited effectiveness highlights clinical uncertainty in the application of surgical procedures. Chapters one to three then feed into **chapter four** - a synthesis of the clinical evidence base that supports the need for a degree of disinvestment from surgery as a treatment for OSA.

Chapter five outlines how a qualitative phase of enquiry, directed at exploring the perspectives and experiences of surgery recipients, was approved by three independent research ethics review boards but was not supported by a small group of surgeons, resulting in the project being canceled. Potential consequences of this for impeding HSR and therefore disinvestment are discussed.

In **chapters six and seven** two sets of results are reported from a qualitative phase of enquiry (semi-structured interviews) involving senior Australian health policy stakeholders. The first results (chapter six) are of policy stakeholders' perspectives on the surgical meta-analysis and clinical audit studies detailed above. The second results (chapter seven) are from an extended series of questions relating to challenges and direction for effecting disinvestment mechanisms in Australia. Stakeholder responses highlight that Australia currently has limited formal systems in place to support disinvestment. Themes included how defining and proving inferiority of health care practices is not only conceptually difficult but also is limited by data availability and interpretation. Also, as with any policy endeavour, there is the ever-present need to balance multiple interests. Stakeholders pointed to a need, and a role, for health services and policy research to build methodological capacity and decision support tools to underpin disinvestment.

Chapter eight provides a detailed commentary piece that builds on all previous sections and summarises the specific challenges that exist for disinvestment, including those methodological in nature. The thesis concludes with potential solutions to address these challenges within the Australian and international context. **Chapter nine** revisits the research questions with a summary of findings. As each manuscript details specific methodological limitations of this multifaceted investigation, these will not be re-visited in chapter nine. Instead, some broad level limitations of this project are discussed. The chapter concludes with a summary of the challenges facing disinvestment and some suggested solutions. I conclude that systematic policy approaches to disinvestment represent one measure to further improve equity, efficiency, quality of care, as well as sustainability of resource allocation.

Chapter Two

Publication: Is Surgery Effective? 1: Meta-Analysis

Redefining Success in Airway Surgery for Obstructive Sleep Apnea:

A Meta Analysis and Synthesis of the Evidence

SLEEP 2007; 30(4):461-467.

[2006 Thomson ISI Impact Factor: 5.126]

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Redefining Success in Airway Surgery for Obstructive Sleep Apnea:

A Meta Analysis and Synthesis of the Evidence

SLEEP 2007; 30(4):461-467.

Adam Elshaug (Candidate)

Conceived and conceptualised manuscript orientation and structure, developed protocol, carried out literature review, extracted and interpolated data for meta-analysis, performed meta-analysis (with assistance from statistician-see acknowledgments section of paper), interpreted data, wrote manuscript and acted as corresponding author.

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John Moss

My contribution to this paper involved: Contribution to protocol design, assistance with data interpretation and manuscript evaluation. I give consent for Adam Elshaug to present this paper for examination towards the Doctor of Philosophy

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Elshaug, A.G., Moss, J.R., Southcott, A.M. and Hiller, J.E. (2006): Redefining Success in Airway Surgery for Obstructive Sleep Apnea: A Meta Analysis and Synthesis of the Evidence.
Sleep, v. 30 (4), pp. 461-467, April 2008

NOTE: This publication is included in the print copy of the thesis held in the University of Adelaide Library.

Chapter Three

Publication: Is Surgery Effective? 2: Multi-Centre Retrospective Audit

An Analysis of the Evidence-Practice Continuum:

Is Surgery for Obstructive Sleep Apnoea Contraindicated?

Journal of Evaluation in Clinical Practice 2007; 13(1):3-9.

[2006 Thomson ISI Impact Factor: 1.263]

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An Analysis of the Evidence-Practice Continuum:

Is Surgery for Obstructive Sleep Apnoea Contraindicated?

Journal of Evaluation in Clinical Practice 2007; 13(1):3-9.

Adam Elshaug (Candidate)

Developed research protocol, gained multiple ethics approvals, collected all primary data, designed and carried out data analysis, interpreted data, conceived and conceptualised manuscript orientation and structure, wrote manuscript and acted as corresponding author.

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Elshaug, A.G., Moss, J.R., Southcott, A.M. and Hiller, J.E. (2007) An analysis of the evidence-practice continuum: is surgery for obstructive sleep apnoea contraindicated? *Journal of Evaluation in Clinical Practice, 13 (1) , pp. 3–9, February 2007*

NOTE: This publication is included in the print copy of the thesis held in the University of Adelaide Library.

It is also available online to authorised users at:

http://dx.doi.org/10.1111/j.1365-2753.2006.00793.x

1. Chapter 3 Limitations (further discussion)

Elshaug AG, Moss JR, Southcott A and Hiller JE. An analysis of the evidence-practice continuum: Is surgery for Obstructive Sleep Apnoea contraindicated? *Journal of Evaluation In Clinical Practice 2007;13(1):3-9.*

The retrospective clinical audit (chapter three) contained a sample of 94 individuals. This provided more than adequate statistical power for all reported analyses as per the aims of the study. However, an interesting but unexpected finding from the analyses was the degree of variability present in the application of procedures by the various surgeons; these 94 individuals received 41 varying combinations of surgery. Herein lays the limitation. From this sample size I could not identify predictive factors for use of a particular type of surgery, combination of surgical techniques or 'successful' surgical outcome. This would have been enlightening given the current dearth of such data in the literature. In effect the unexpected and hitherto unreported variability limited the ability to examine factors associated with success.

To isolate predictive factors (via sub-group analyses) would require a much higher sample size. In the planning stages of this project a higher sample size was expected to be feasible as physicians from two additional hospitals showed considerable interest in the project – to the point ethics clearances were obtained at both (additional) hospitals. Ultimately these hospitals were not included. The reason provided by one was,

> *"Unfortunately and shamefully I think this is not something that we can do without a painstaking pulling of files or sleep studies. We lack a data base (although we are working on it). Dr.…… from our dept has developed text searching macros that might be able to search for key words such as "surgery" "UPPP" etc on sleep study reports. But there would be considerable work then to pull the studies and see whether they were relevant and had the necessary information. We don't have a spare pair of hands to put to this task, but if you were willing to do the work we could accommodate you in the lab. My guess is that we would have seen a similar number of surgical cases as other labs. Regards…"*

For this particular project I was willing to undertake this "painstaking" retrieval task given that a similar process was applied at the two participating hospitals as a quality assurance measure. Specifically, the two participant hospitals have long established computerised databases that

allow for multiple and detailed search criteria and cross-checking/matching (see methods section of chapter three). In addition, at these hospitals I manually searched all medical records that scored even a vague 'hit' via the computerised search strategy. This ensured rigorous case selection. However, the uncertainty in the search method from the additional hospital would, I felt, potentially introduce systematic error in the selection of cases. This is particularly so as there was insufficient time or resources to complete a manual search of all medical records to identify cases for inclusion (requiring an manual search of an estimated 4,000 hard copy medical records). For this reason it was decided not to progress. Full support from the medical staff of the fourth hospital was provided (in writing) however senior management withdrew permission due a perception that the project (by allowing access to patient records, albeit de-identified) would potentially lead to, "*the hospital being in breach of the Privacy Act*".

The limitations introduced by the exclusion of these hospitals were two-fold. Firstly, the sample size was compromised (as discussed above). Secondly, these hospitals (one in particular) treated a higher proportion of privately insured individuals. Although the eventual sample did contain a mix of public and private patients, the inclusion of this hospital would have increased the representation of this sub-sample which would have allowed for sub-group analyses.

Chapter Four

Publication: Clinical Evidence for Disinvestment
BMJ Change Page

Upper Airway Surgery should not be First-Line Therapy for Adult Obstructive Sleep Apnoea (OSA)

British Medical Journal (Accepted July 19, 2007 – see appendix four)

[2006 Thomson ISI Impact Factor: 9.245]
Article Provenance: Commissioned based on an idea from the author, externally peer reviewed

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Upper Airway Surgery should not be First-Line Therapy for Adult Obstructive Sleep Apnoea (OSA)

British Medical Journal (Accepted July 19, 2007)

Adam Elshaug (Candidate)

Contacted BMJ editors with detailed concept for BMJ Change Page following idea from John Moss. Conceptualised manuscript orientation and structure by synthesising elements of this thesis in accordance with BMJ guidelines. Wrote manuscript and acted as corresponding author.

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John Moss

My contribution to this paper involved: Original suggestion to submit concept for BMJ Change Page. Contribution to manuscript drafting and revision (design and evaluation). I give consent for Adam Elshaug to present this paper for examination towards the Doctor of Philosophy

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Janet Hiller

My contribution to this paper involved: Contribution to manuscript drafting and revision (design and evaluation). I give consent for Adam Elshaug to present this paper for examination towards the Doctor of Philosophy

Signed ……Date……

Elshaug, A.G., Moss, J.R., Hiller, J.E. and Maddern, G.J. (2008) Upper airway surgery should not be first line treatment for obstructive sleep apnoea in adults. *British Medical Journal, v. 336, pp.44-45, January 2008*

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It is also available online to authorised users at:

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Chapter Five

Stakeholder Engagement 1:
Patient Consultation Phase - Canceled

Becoming immersed in a study requires passion: passion for people, passion for communication, and passion for understanding people. This is the contribution of qualitative research… In the qualitative arena the individual is not inserted into the study, the individual is the backbone of the study.

Janesick. *The Choreography of Qualitative Research Design*…2003: 71

The 2004 Cochrane review of upper airway surgery for the treatment of OSA concludes:

> *In light of the current lack of good trial-based evidence, clinicians should consider restricting surgery for obstructive sleep apnoea to that carried out as part of clinical trials. Where practice is continued,* ***patients should be informed*** *of the experimental nature of the operations.* ***Patients should be told*** *that there is a lack of good trial based evidence of the efficacy of surgery for obstructive sleep apnoea, a course of action that may restrict the use of these operations.* [37] (emphasis added)

It is increasingly considered not only reasonable but invaluable that health service researchers incorporate, where appropriate, the views of prominent stakeholders into the prism of research. Within the scope of this PhD this undoubtedly includes the patient group - those who have undergone the surgical procedures in question. Currently there is a degree research highlighting patient experiences and satisfaction with the gold-standard treatment modality, CPAP [75 76]. There have, however, been relatively few accounts of the self-reported experiences of those individuals who have undergone surgical procedures as a treatment for this condition. In particular, there is no research to confirm whether or not patients have been informed, in accordance with the Cochrane review recommendations, of *the experimental nature of the operations* and of the *lack of good trial based evidence of the efficacy of surgery for OSA.*

Hearing about the subjective experiences of an Australian cohort may greatly assist in building the research base in this area. One arm of this PhD project sought to explore the personal experiences and satisfaction of individuals who had undergone surgery as a treatment for OSA. Eligible participants were to be drawn from the same pool as those who made up the previously reported audit (chapter three). This study sought to approach patients with a questionnaire exploring their personal experiences and level of satisfaction with the surgery process, and to explore levels of pre-surgical information provision (by physicians and surgeons) as to their treatment options, evidence for effectiveness and likely prognoses.

A preliminary questionnaire was devised and piloted in consultation with several respiratory and sleep medicine physicians and one surgeon. Preliminary drafts were presented to three student members of the Discipline of Public Health of the University of Adelaide, as well as to three technical staff members of clinical sleep laboratories in Adelaide. These individuals assisted in

clarifying the structure and content of questions to be asked. Following pre-piloting and subsequent questionnaire refinement, submissions were made to three independent Human Research Ethics Committees (HREC) seeking approvals for three research phases:

(1) Formal questionnaire piloting with a sub-group of individuals (patients) who had undergone surgery as a treatment for OSA. Formal piloting with a sub-group of the target audience would have allowed for further, minor amendments to be made if required and for reliability and validity testing of the questionnaire. It was anticipated that this pilot sample would comprise between 4 and 10 individuals.

(2) A full scale mail out of the final questionnaire to all individuals selected as eligible participants (surgery cases from Jan 2001 to Nov 2005 from two large teaching hospitals in Adelaide; those utilised in the audit study). All personal contact details for this group exist in the medical record systems of the relevant hospitals. With ethics approval these details would have been utilised for the mail out using the contact system reported by Dillman[115]. This would have involved the questionnaire being accompanied by a detailed covering letter. A replacement questionnaire would have been sent to non-respondents 2-3 weeks after the initial mailing. A final contact attempt would have been made by telephone 1-2 weeks after the replacement questionnaire.

The questionnaire would have allowed for insight into the experiences and satisfaction of individuals who had undertaken surgery as a treatment for OSA, as described above.

(3) The third phase of the ethics application sought to ask participants' permission to link the questionnaire information with their objective sleep study data in order to explore correlations between objective sleep study findings (post surgery) and subjective questionnaire responses. The final item in the questionnaire explained the concept of data linkage and sought the participants' consent for data linkage to occur in the future, by this research team, for the purposes of comparing objective sleep study data with questionnaire responses.

The final version of the questionnaire (see appendix two) gained approval from all three HRECs (see appendix two). However, late in the planned roll out phase, a final version of the questionnaire was presented to a group of clinical stakeholders (physicians and surgeons) for appraisal and if necessary further refinement. At this meeting three clinical stakeholders indicated their withdrawal of support for the project. This withdrawal of support specifically affected access to the patient database at one of the two large hospitals, representing a substantial proportion of the sample. Despite the second hospital maintaining its support for the project (and representing the majority of the potential sample) it was decided that this reduction in sample size would substantially under-power potential cross analyses between the qualitative data and the objective sleep study data (i.e. accessible via record linkage). Specifically, based on the now revised sample size (pooled from one hospital only), a questionnaire response rate of 50% would capture 30 returned questionnaires. Obviously this number would vary depending on different response rate scenarios. Following deliberations with my supervisors it was decided on these grounds, reluctantly, to cancel this investigative arm and move on to the next phase of the project.

The clinicians justified their sudden withdrawal of support for the questionnaire by referring to the 'legal climate' present in Adelaide at the time. It is understood that they were referring to legal action between patients and clinicians. Since this legal action did not involve the research described in this thesis, it will not be explored further here. What remains noteworthy, and of considerable interest for the research and policy making community, is the ability of a relatively small, albeit key group of stakeholders to withdraw support and therefore affect the viability (in this case resulting in the cancellation) of health services research that might otherwise be considered in the public interest because of its relevance for the quality and safety of health care. It is also a salutary reminder of how, once evidence is in the public domain, it may be used for purposes that were not originally intended.

My inability to pursue this element of my research further was disappointing because it continues to be argued that surgery for the treatment of OSA is a viable and efficacious procedure in certain circumstances[116] and the results presented in the preceding chapters support this notion that a small percentage of recipients may benefit from surgery. Furthermore,

efficacious CPAP treatment is dependent upon patient compliance, resulting in many patients' expressed desire for surgical 'cure'. This potential for a surgical 'cure' makes the *availability* of surgery attractive for some. To this extent the practice continues to be presented to patients as a treatment option throughout Australian hospitals and private surgical clinics (as will be discussed further in forthcoming chapters).

This specific project sought to overcome a lack of knowledge that currently exists about the personal experiences and satisfaction of people who have undergone upper airway surgery, particularly in Australia. It might also have uncovered the degree to which patients felt fully informed prior to their surgical experience, illuminating not only the effectiveness of, but the satisfaction with surgeons' communication strategies with them as a potential and soon-to-be surgery recipient. Such results might have elucidated whether prior knowledge, both of treatment options and likely treatment outcomes, correlates with actual experiences. All of this would have allowed for an exploration of the potential strengths and weaknesses in the communication strategies of clinical stakeholders and of the potential existence of supplier-induced demand within this context. Although participation would not necessarily have resulted in any direct benefit for those involved, it would have contributed to patient involvement in evidence generation and in doing so may have assisted future patients, the health care community and health policy. It may also have contributed to hypothesis generation for health services researchers in bridging the gaps in translation of evidence to policy and practice. What remains of interest is how this surgical case study is perceived by policy stakeholders and how it might prime an exploration of policy stakeholder perspectives to further build the evidence base for the larger issue of disinvestment in health policy. This will unfold in the following chapters.

Practical men who believe themselves to be quite exempt from any intellectual influences are usually the slaves of some defunct economist.... It is ideas not vested interests which are dangerous for good or evil

John Maynard Keynes 1936

Chapter Six

Publication: Stakeholder Engagement 2: Policy Stakeholders' Perspectives (surgery)

Exploring Policymakers' Perspectives on a Clinical Controversy:

Airway Surgery for Adult OSA

Journal of Evaluation in Clinical Practice (To be submitted)

[2006 Thomson ISI Impact Factor: 1.263]

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Exploring Policymakers' Perspectives on a Clinical Controversy:

Airway Surgery for Adult OSA

Journal of Evaluation in Clinical Practice (To be submitted)

Adam Elshaug (Candidate)

AE developed research protocol, letter of approach to participants, preparatory reading material and interview schedule (piloting and refinement questions), selected and contacted potential participants, carried out interviews for collection of all primary data, carried out thematic analysis, interpreted data, conceived and conceptualised manuscript orientation and structure, wrote manuscript and acted as corresponding author.

Signed ……Date……

Janet Hiller

My contribution to this paper involved:
Contribution to research protocol design, selection of potential interview participants as well as data interpretation and manuscript evaluation. I give consent for Adam Elshaug to present this paper for examination towards the Doctor of Philosophy

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John Moss

My contribution to this paper involved:
Contribution to research protocol design, piloting and refinement of interview schedule, selection of potential interview participants as well as data interpretation and manuscript evaluation. I give consent for Adam Elshaug to present this paper for examination towards the Doctor of Philosophy

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Elshaug, A.G., Hiller, J.E. and Moss, J.R. : Exploring Policymakers' Perspectives on a Clinical Controversy: Airway Surgery for Adult OSA.
Journal of Evaluation in Clinical Practice (To be submitted)

NOTE: This publication is included in the print copy of the thesis held in the University of Adelaide Library.

Chapter Seven

Publication: Stakeholder Engagement 3: Policy Stakeholders' Perspectives (Disinvestment)

Exploring Policymakers' Perspectives on Disinvestment from Ineffective Health Care Practices

International Journal of Technology Assessment in Health Care

(Accepted September 21, 2007 – see appendix four)

[2006 Thomson ISI Impact Factor: 1.151]

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Exploring Policymakers' Perspectives on Disinvestment from

Ineffective Health Care Practices

International Journal of Technology Assessment in Health Care (Accepted September 21, 2007)

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AE developed research protocol, letter of approach to participants, preparatory reading material and interview schedule (piloting and refinement of questions), selected and contacted potential participants, carried out interviews for collection of all primary data, carried out thematic analysis, interpreted data, conceived and conceptualised manuscript orientation and structure, wrote manuscript and acted as corresponding author.

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Contribution to research protocol design, selection of potential interview participants as well as data interpretation and manuscript evaluation. I give consent for Adam Elshaug to present this paper for examination towards the Doctor of Philosophy

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Contribution to research protocol design, piloting and refinement of interview schedule, selection of potential interview participants as well as data interpretation and manuscript evaluation. I give consent for Adam Elshaug to present this paper for examination towards the Doctor of Philosophy

Signed ……Date……

Elshaug, A.G., Hiller, J.E. and Moss, J.R. (2008) Exploring policy-makers' perspectives on disinvestment from ineffective healthcare practices. *International Journal of Technology Assessment in Health Care, v. 24 (1), pp. 1-9, January 2008*

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Chapter Eight

Publication: Building the Evidence Base for Disinvestment

Challenges in Australian Policy Processes for Disinvestment from Existing, Ineffective Health Care Practices

Australia and New Zealand Health Policy (Resubmitted with revisions Sept 22, 2007)

[BioMed Central open access; http://www.anzhealthpolicy.com/ - no impact factor]

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Challenges in Australian Policy Processes for Disinvestment from Existing, Ineffective Health Care Practices

Australia and New Zealand Health Policy (Resubmitted with revisions September 22, 2007)

Adam Elshaug (Candidate)

Conceived, conceptualised and designed the first substantive draft of this manuscript (orientation and structure), and subsequent draft revisions. Wrote manuscript and acted as corresponding author.

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Janet Hiller

My contribution to this paper involved:
Manuscript evaluation and development. I give consent for Adam Elshaug to present this paper for examination towards the Doctor of Philosophy

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Sean Tunis

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An international perspective (USA) in addition to evaluation and development of two drafts.
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John Moss

My contribution to this paper involved:
Manuscript evaluation and development. I give consent for Adam Elshaug to present this paper for examination towards the Doctor of Philosophy

Signed ……Date……

Elshaug, A.G., Hiller, J.E., Tunis, S.R. and Moss, J.R. (2007) Challenges in Australian policy processes for disinvestment from existing, ineffective health care practices.
Australia and New Zealand Health Policy, v. 4, pp. 23-

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It is also available online to authorised users at:

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Chapter Nine

Conclusions

It must be considered that there is nothing more difficult to carry out, nor more dangerous to handle, than to initiate a new order of things. For the reformed has enemies in all those who profit by the old order, and only lukewarm defenders in all those who profit by new order, this lukewarmness arising partly from fear of their adversaries, who have the laws in their favour; and partly from the incredibility of mankind, who do not truly believe in anything new until they actually have had actual experience of it.

Machiavelli "*The Prince*" 1513

This thesis has consisted of multiple phases of enquiry into a burgeoning but also politically sensitive area of health services and policy research. During the 3.5 years of gestation, I faced certain challenges but overall I feel confident in how I have addressed the original research questions. There are specific methodological limitations to this thesis and these were noted in all relevant chapters. In this, the final chapter, I will not re-visit these limitations individually but instead present what I consider to be a broad discussion of the answers to the research questions posed.

Research question one: *Does upper airway surgery for obstructive sleep apnoea syndrome represent clinically effective treatment?*

I have concluded that surgery represents clinically effective treatment, for only a minority of recipients. The 2005 Cochrane review included seven randomised controlled trials (n=412), and concluded that the results of surgery were inconsistent: significant improvement in polysomnography occurred in only three trials, and that in health-related quality of life occurred in four trials [89]. For both measures, the trials made only limited comment on clinical significance, and the review concluded that there was a lack of an impact on symptoms (except in two trials) and that overall a significant benefit was not demonstrated. The Cochrane review represents an important contribution to this particular case study. However, relying on the Cochrane review alone has limitations, given its sole inclusion of RCT designs and the exclusion of methodologies deemed less rigorous. For highly invasive surgical techniques, RCT designs are difficult and often impracticable. Also, given the existing evidence for limited effectiveness, RCTs are difficult to justify on ethical grounds. In this case study any singular reliance on the Cochrane review excludes otherwise valuable elements of evidence that are relevant and of meaning in decision making processes.

The meta analysis reported in my chapter two sought to overcome this limitation by including eighteen surgical studies that sit lower on the traditional hierarchy of evidence (n=385; seventeen level four audits, one randomised controlled trial). The pooled success rate (using an AHI of ≤ 5) for Phase I procedures was 13%, and for Phase II procedures was 43%. In addition to this report, the multi-centre audit reported in chapter three revealed substantial procedural variability, with the observed cohort (n=94) receiving 41 varying combinations of surgery and an overall surgical success rate of 13%. In chapter four I presented this clinical evidence together with that from a recent systematic review of 48 studies that found up to 62% of 21,346 surgery recipients reported persistent adverse effects, such as persistent dry throat, globus

sensation, difficulty in swallowing (including spontaneous nasal regurgitation), voice changes, and smell and taste disturbances. Up to 22% regretted surgery [86]. Collectively this evidence points to controversy in the continued endorsement (and government subsidised funding) of these procedures. Given the apparent indictment of surgery, the remaining research questions sought to unpick the complexities of this particular case study in terms of the ways in which surgery for OSA represents a gap in evidence-policy-practice, and, what policy mechanisms exist to further analyse and address this case study, and others like it.

Research question two: *What are the possible explanations for any gap between evidence, policy and practice, and what are the potential resolutions?*

Throughout the thesis I have demonstrated that debate and controversy exists within and between the specialty fields of sleep medicine and ear, nose and throat (ENT) surgery regarding the appropriateness of upper airway surgical procedures for treatment of OSA. I have elucidated some of the elements that are contributing to that controversy, including the presentation and interpretation of evidence regarding the effectiveness of surgical procedures, independent of, but also in comparison to non-surgical treatment alternatives such as the gold standard OSA treatment modality available, CPAP [90].

Rose [117] (Chapter 2, p.10) discusses how decision making in medicine underlies the process we choose to call 'diagnosis'. He suggests what we really mean is that we are diagnosing a 'case for treatment' and not a disease entity. Although the surgeon may choose certain individuals who are labelled as 'cases of OSA', Rose would argue that for the surgeon these are actually 'cases for OSA surgery', for OSA itself occurs on a spectrum and most will never come to a surgeon's attention. So how is it that a surgeon might justify his or her selection of 'a case for treatment'? I have discussed how surgeons, through their collective published works, have an alternate perception of the efficacy of these procedures based on, among other things, an alternative definition of success. The surgical community, in reporting outcomes in peer-review (surgical) literature has, and largely continues, to perpetuate claims of treatment 'response', 'effectiveness', 'success' and even 'cure' of OSA as a reduction in the AHI of equal to, or greater than 50%. Some go on to specify this criterion plus a reduction in AHI to 20 or less [105-107]. I have presented the problems associated with interpreting these conservative and variable criteria. That is, such reporting of surgical 'success' does not correspond to the clinical endpoints as determined by the professional sleep medicine bodies. The reporting of surgical audits is performed overwhelmingly by surgeons, who adopt the less stringent criteria of

treatment 'success'. The implications of this for population health and health policy are considerable, with patients potentially foregoing alternative treatments due to the (mis)perception of a surgical 'cure', and policy stakeholders being shielded from any critique of the finer detail that makes this a clinical controversy.

The evidence upon which these surgical practices are carried out appears out of step with that which is the basis of the parallel specialty field of sleep medicine. There is evidence that surgeons have high confidence in their own judgement and low confidence in clinical practice guidelines [118]. How their judgement is formed is, in itself, a complex phenomenon not well understood (and beyond the scope of this thesis). It is known that the subcultures within medicine, each with their own models of knowledge acquisition and understanding, make knowledge transfer a fractured, lengthy and imprecise process [14 25 119]. The surgical practice patterns observed in this thesis fit well with what Kuhn has defined as a scientific paradigm [6]. One prominent example of this is the clinical consensus that is evident in mass publication bias by surgeons reinforcing the less-stringent surgical definition of success. We might expect surgeons to be guided at least in part by clinical practice guidelines [118]; the problem is that the guidelines here (in the form of surgical publications) appear to exclude important epidemiological evidence that has surfaced since 2002. The inconsistency uncovered in this project thus supports the notion of a lag that exists in the development and reporting of evidence and the subsequent development of practice guidelines (or similar mechanisms for clinically driven change patterns) that reflect contemporary evidence.

A systematic review of the most current sleep medicine, health outcome related evidence highlights that the ENT definition(s) bear only a limited relationship to the evidence-based criteria defining 'effective' treatment. Contemporary evidence supports the stricter sleep medicine classifications. The more stringent reduction in AHI has been shown to improve numerous health outcome measures, in cardiovascular disease [62 64-68], heart failure [69 70], endocrinology [71-74], and health-related quality of life [75-77]. Importantly, all of these reports have emerged since 2002 and their existence and synthesis is necessary in order to demonstrate the apparent inconsistency within current surgical practice. Indeed, it is the meta-analysis paper of chapter two that offers the first demonstration of this link and to have presented it to the scientific, clinical and policy communities.

The surgical case study highlights some of the challenges that exist in the appraisal and synthesis of evidence for EBM/HTA applications when varying definitions of treatment success exist across multiple specialties, all treating the same condition. Here there is domination of one

definition in the surgical literature, coupled with the lag that exists in the assimilation of more contemporary evidence. One might suggest this is a conundrum of present day poly- and co-morbidity where multiple specialty groups are increasingly treating similar disease profiles and progressions without fully recognising or assimilating the wealth of relevant health outcomes research that is occurring in parallel fields - for the same condition. As a result, questionable and/or outdated definitions of clinical endpoints (in this case those associated with 'surgical success') dominate contemporary reporting, and therefore, appraisal processes.

Should surgery ever be examined by policy makers for (disinvestment) assessment, the overwhelming evidence to date (prior to this thesis) would appear generally supportive of the procedures. However, I have demonstrated that existing evidence deserves to be questioned. All of this is particularly sobering when one considers the tens of thousands of surgical recipients estimated per year internationally [120] [86], and, if alternative OSA treatment modalities such as CPAP or weight loss are being rejected or under-utilised due to the (mis)perception of a surgical 'cure'.

In chapters two, three and four I have attempted to address the tensions between the relevant medical specialties, and the evidence, in several ways. Firstly, by proposing that in all future surgical trials, in addition to reporting the statistical significance of findings and the AHI reduction by 50% and or ≤ 20, that 'cure' rates based on accepted definitions are also reported. That is, post-surgical 'success' be reported based on AHI of ≤ 5 and or ≤ 10. In this way, while debate might continue, the reporting of surgical outcomes will adhere to current, more stringent standards. Second, I have called for the relevant medical specialties in this area to clarify their position within this debate, with intra and inter-specialty consensus the ultimate objective. Third, I have made recommendations that would see referral pathways tightened so that, a) potential surgery recipients do not progress to surgery before (at very least) a formal diagnosis of OSA has occurred - as was uncovered to be occurring (in chapter three); b) that all future surgery occurs in clinical trials, and; c) that potential surgery recipients are fully informed of the treatment alternatives, the poor outcomes, the side effects and potential for relapse following surgery. In the meantime, I claim that the onus of proof is placed upon the surgical community to develop and/or improve predictive models that would identify who will and who will not benefit from the various surgical procedures.

With this case study as a back drop, **research question three** went on to ask: Does Australia have established policy mechanisms, including health care reimbursement structures, that can

react to and influence existing health care practices with uncertain clinical and cost-effectiveness? The final **research question** followed: Can these policy mechanisms adapt to the ever-changing nature of evidence?

For this thesis a consumer perspective was sought in an attempt to add patient insight and weight to the discussion points and conclusions drawn from this project. However, as outlined in chapter five, this was not to be. This work lacks the voice of surgery recipients in building answers to these research questions. Beyond this, the provision of answers required an analysis of current policy models in Australia and, to a lesser extent internationally, that either exist or may be formed to support and effect disinvestment. As a starting point for this investigative phase, senior policy stakeholders were interviewed to gain their perspectives on these matters.

To establish a context for disinvestment, stakeholders were provided with information about the surgery for OSA case study for comment. Thematic analysis highlighted concern with the diversity of surgical procedures coupled with their limited effectiveness (suggesting potential clinical uncertainty in the appropriate choice and application of procedures). Not surprisingly there were associated concerns about suboptimal resource allocation. Stakeholders noted the methodological complexities, the ethical issues raised and the necessary role of patients in considerations regarding appropriateness for these procedures, and, any similar examples for potential 'disinvestment analyses'. Policy stakeholders acknowledge that the surgical procedures appear appropriate only for a minority, with consensus that policy level restrictions to government funding for these procedures may be warranted. Chapter six thus highlights that this clinical controversy is of interest and relevance from a policy perspective. It further highlights the need for clinical consensus on definitions of surgical 'success' in treating OSA, as this forms an important basis for policy considerations on the matter.

Chapter seven reported the policy stakeholders' perspectives regarding disinvestment more broadly. In partial answer to research questions three and four, three primary themes were identified. 1) The current focus in Australia on assessment of new and emerging health technologies/practices and lack of attention toward existing practices is due to resource limitations (for an established body to adopt the role) and methodological complexity. This reinforces earlier discussions in this section. Participants suggested that a parallel model to Australia's current assessment process for new medical technologies (i.e. the Medical Services Advisory Committee, MSAC) would be best-positioned to facilitate disinvestment. 2) To advance the disinvestment agenda requires an explicit focus on the potential for cost-savings

coupled with improved quality of care. This, according to policy stakeholders, would serve to foster political motivation and support. 3) Support (financial and collaborative) is needed for research advancement in the methodological underpinnings associated with health technology assessment and for disinvestment specifically.

For this phase of enquiry there are methodological considerations. For chapter six specifically (policy stakeholders' perspectives on surgery for OSA), despite being provided with preparatory reading and a comprehensive reference list, policy stakeholders were asked to respond to limited evidence (here, based on a limited number of publications and that from a Cochrane review relating to surgery). Further, although participants could seek clarification on specific issues they could not, in a single telephone or face to face interview, be fully briefed on all of the relevant considerations. Nonetheless, this study (as reported in chapters six and seven) was carried out for a specific purpose – to generate policy-related insights that could inform and answer the research questions, and to generate hypotheses to be tested in subsequent research. Given the systematic investigation, organisational representation, seniority of position of respondents and saturation of responses achieved – this stakeholder engagement succeeded as an informative, exploratory study. It provided rich information to enlighten the subsequent policy analysis reported in chapter eight.

Chapter eight sought to further build on these findings with a disinvestment policy analysis. In answer to these remaining research questions, Australia does have a template *policy mechanism… that can react to and influence existing health care practices with uncertain clinical and cost-effectiveness* in the existing MSAC model (or similar). Indeed, it is within the MSAC mandate to do so. However, certain challenges to the disinvestment process were detailed in chapter eight. The most prominent included, 1) reluctance by key stakeholders to provide resources for a functioning disinvestment body such as MSAC to focus on *existing* technologies in addition to those that are new and emerging. This incorporates a lack of reliable administrative mechanisms (legal frameworks etc) to identify and prioritise *existing* technologies and practices for which there is relative uncertainty as to clinical and cost-effectiveness. 2) Political, clinical and social challenges of removing an established technology (including challenges to limiting coverage to specific patients, institutions, or providers), and, 3) Failure to deploy resources to support a research agenda to advance disinvestment methods. I have discussed how these challenges arise from the many complexities that are associated with the removal of entrenched technologies. These complexities are economic, methodological, ethical, and social.

I have examined disinvestment as incorporating and building on the principles and skill base of 'traditional' EBM and HTA. The contribution of disinvestment is its concentration on *existing* technologies or services, as opposed to *new* and *emerging*; though this line can often be somewhat blurred. Disinvestment may be somewhat easier with pharmaceuticals than with other technologies. This is supported by a recent report by Linden and co-workers who reviewed 159 technologies from 88 NICE appraisals [121]. Of these, 84 (53%) were judged as new and 75 (47%) were existing technologies - a high proportion. However, upon further analysis, a total of 119 (75%) were pharmaceuticals, 22 (14%) were devices, 14 (9%) were procedures, and 4 (3%) were categorised as miscellaneous. Clearly, existing devices and procedures are under-represented. The appraisal process for these existing practices is more complex as often no individual is 'hurt' by existing practices, unlike for pharmaceuticals that may be 'flagged' due to adverse events. Instead, individual patients and patient groups are simply inappropriately or under-treated due to limited clinical effectiveness.

As I have discussed, perhaps the biggest problem facing effective disinvestment is the reversal of the burden of proof. For centuries, *primum non nocere* (first, do no harm) has been a fundamental ethical obligation on all physicians (now, all health care workers). This has served as an ethical justification for funding and regulation bodies to insist on safety and effectiveness. In its turn, the requirement for cost-effectiveness (in one form or another) draws its justification from the overall scarcity of health resources. Thus the burden of proof lies with the sponsor of the new practice, device or pharmaceutical. If the sponsor cannot provide proof of benefit, the application fails. In attempting to retire (disinvest) an apparently obsolete technology, there is a likelihood that the regulator/reviewer will have to prove that the technology is ineffective or non-cost-ineffective. In this thesis I have discussed how defining and proving inferiority is conceptually difficult and that it is now the regulator rather than the sponsor who has to make a compelling argument. Also, the regulator must first identify or be made aware of the practice 'requiring' disinvestment analyses. In Australia there appear to be limited groups with the incentives, resources or structure to do this, and, as the policy stakeholders in this analysis discussed, relying on clinicians alone to do so is inadequate. In chapter eight I outline several recommended initiatives to be implemented in Australia (and internationally where appropriate) to advance disinvestment, including:

- Government partnerships to involve the professional colleges and relevant stakeholder groups (consumer/community) to put disinvestment on the agenda to build awareness,

collaboration and improved health outcome data generation and reporting (ongoing medico-vigilance).

- Dedicated funds and distinct processes (i.e. a transparent legal framework) within the MSAC and PBAC to:
    - Identify technologies and practices about whose outcomes there is relative uncertainty for disinvestment analysis/review
    - Conduct disinvestment assessments/reviews of the selected item(s)

  This should involve a parallel and expanded role of these committees to address existing practices in an analogous manner to their current focus on new and emerging technologies, practices and pharmaceuticals.
    - At this juncture in Australia's health policy landscape, collaborative links to advance disinvestment should be made between the relevant stakeholder bodies, including: MSAC/PBAC, state departments of health, the Australian Commission on Safety and Quality in Health Care, the National Institute of Clinical Studies (and the NHMRC more broadly).

- For existing health care items for which there is relative uncertainty, consideration for the implementation of 'funding with evidence generation'. That is, ongoing reimbursement being agreed for only a limited number of years pending evidence generation/review processes - with the possibility of extensions being considered.

- Dedicated funding and cross-disciplinary collaboration to build health services and policy research capacity with a focus on advancing disinvestment research methodologies and decision support tools for policy stakeholders.

These disinvestment measures require greater attention in Australia and internationally, both for quality of care and sustainable resource allocation.

A Machiavellian approach might suggest it may be better for society to have people using a cheap but ineffective technology or practice rather than an expensive, new, but also ineffective technology. I consider that this is an inadequate approach to an ethical pursuit of efficiency, equity, quality and safety of care, and sustainability of health resource allocation. Disinvestment calls for *a new order of things* and as Machiavelli recognised 500 years ago, this, invariably, is a difficult and sometimes dangerous thing, but often a no less worthy thing.

Appendices

Appendix 1: Retrospective surgical audit: Ethics approvals

Appendix 2: Patient Questionnaire: Ethics approvals, introductory letter, questionnaire

Appendix 3: Policy stakeholder interviews: Introductory letter, preparatory reading, and interview schedule

Appendix 4: Letters of acceptance for 'in-press' journal articles

Appendix 5: Signed statements of authorship from non-supervisory co-authors: AM Southcott, G Maddern and S Tunis

Appendix One

Retrospective Surgical Audit:
Ethics Approvals

Appendix One is included in the print copy of the thesis held in the University of Adelaide Library.

Appendix Two

Patient Questionnaire:
Ethics Approvals
Introductory Letter
Questionnaire

THE UNIVERSITY
OF ADELAIDE
AUSTRALIA

OFFICE OF THE DEPUTY VICE-CHANCELLOR (RESEARCH)

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CRICOS Provider Number 00123M

Applicant: Dr JR Moss

Department: Public Health

Project Title: *Mailout questionnaire to gauge patient experiences and satisfaction with upper airway surgery as a treatment for obstrutive sleep aponea syndrome(OSA)*

THE UNIVERSITY OF ADELAIDE HUMAN RESEARCH ETHICS COMMITTEE

Project No: H-008-2006 RM No: 0000006795

APPROVED for the period until: 31 December 2006

subject to: (i) receipt of the hospital covering letters, and (ii) modification to the information letter to participants. It is noted that this study involves Mr Adam Elshaug, PhD candidate.

Refer also to the accompanying letter setting out requirements applying to approval.

Associate Professor Garrett Cullity
Convenor
Human Research Ethics Committee

Date: 1 MAR 2006

Government of South Australia
Central Northern Adelaide
Health Service

06 March 2006

The Queen Elizabeth Hospital
28 Woodville Road
WOODVILLE SOUTH SA 5011

Lyell McEwin Hospital
Haydown Road
ELIZABETH VALE SA 5112

Mr A Elshaug
Respiratory Medicine
The Queen Elizabeth Hospital

Dear Mr Elshaug Application Number 2006006

The Ethics of Human Research Committee Chairperson has considered your protocol under Expedited Review (Section 2.27, National Statement on Ethical Conduct in Research Involving Humans) entitled:

"Mail out questionnaire to gauge patient experiences and satisfaction with upper airway surgery as a treatment for Obstructive Sleep Apnoea Syndrome (OSA)"

The following documents have been reviewed and approved:

- CNAHS Ethics of Human Research Committee (TQEH & LMH) Application Form
- Participant Information Sheet, Version 3 dated 23 February 2006
- Questionnaire, dated 23 February 2006
- Covering Letter (Dr AM Southcott, TQEH) dated 15 February 2006

Approval Status: **FINAL**

Period of Approval: **06 March 2006 – 06 March 2007**

Please note the terms under which Ethical approval is granted:

1. Researchers are required to immediately report to the Ethics of Human Research Committee anything which might warrant review of ethical approval of the protocol, including:

(a) serious or unexpected adverse effects on participants;
(b) proposed changes in the protocol; and
(c) unforseen events that might affect continued ethical acceptability of the project

2. Protocols are approved for up to twelve months only and a report is required at the end of the study or 12 month period. Extensions will not be granted without a report to the Committee.
3. Confidentiality of the research subjects shall be maintained at all times as required by law
4. All research subjects shall be provided with a Patient Information Sheet and Consent Form, unless otherwise approved by the Committee
5. The Patient Information Sheet and Consent Form shall be printed on the relevant site letterhead stating the contact details for the researchers
6. The Patient Information Sheet must state that the Executive Officer can be contacted for information regarding conduct of the study, policies and procedures, or if the participant wishes to make a confidential complaint
7. A report and a copy of any published material should be forwarded to the Committee at the completion of the project.

Yours sincerely

A/Prof Timothy Mathew
Chairman
Ethics of Human Research Committee (TQEH & LMH)

Ethics of Human Research Committee (TQEH & LMH)
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Building the Evidence Base for Disinvestment

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6 March 2006

Mr Adam Elshaug
Discipline of Public Health
Level 9, Tower Building
10 Pulteney Street
UNIVERSITY OF ADELAIDE
ADELAIDE SA 5005

Dear Mr Elshaug,

Re: "Mail out questionnaire to gauge patient experiences and satisfaction with upper airway surgery as a treatment of Obstructive Sleep Apnoea Syndrome (OSA)."
Patient Information Sheet, Version 3 (23 February 2006).
RAH PROTOCOL NO: 060301.

I am writing to advise that Research Ethics Committee approval has been given to the above project.

Research Ethics Committee deliberations are guided by the NHMRC National Statement on Ethical Conduct in Research Involving Humans.

The general conditions of approval follow:

- Adequate record-keeping is important. If the project involves signed consent, you should retain the completed consent forms which relate to this project and a list of all those participating in the project, to enable contact with them in the future if necessary. The duration of record retention for all research data is 15 years.
- You must notify the Research Ethics Committee of any events which might warrant review of the approval or which warrant new information being presented to research participants, including:
  (a) serious or unexpected adverse events which warrant protocol change or notification to research participants,
  (b) changes to the protocol,
  (c) premature termination of the study.
- The Committee must be notified within 72 hours of any serious adverse event occurring at this site.
- Approval is ongoing, subject to satisfactory annual review. An annual review form will be forwarded to you at the appropriate time.

If University of Adelaide personnel are involved in this project, you, as chief investigator, must submit a Human Research Approval Notification form (available at: http:www.adelaide.edu.au/research/ethics/human/guidelines/) within 14 days of receiving this ethical clearance to ensure compliance with University requirements and appropriate indemnification.

Yours sincerely,

Dr M James
CHAIRMAN
RESEARCH ETHICS COMMITTEE

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Date

Upper Airway Surgery Questionnaire

Dear .....,

You are invited to participate in a research project being conducted by the Discipline of Public Health, the University of Adelaide, Australia. This project is part of an effort to learn more about the personal experiences and satisfaction of those who have undergone upper airway surgery as a treatment for obstructive sleep apnea, snoring or similar condition. Please read this sheet carefully and be confident that you understand its contents before deciding whether to participate.

Why have you been approached?
In recognising the importance that this research might play in the future care of patients, the sleep laboratories at The Queen Elizabeth Hospital and the Royal Adelaide Hospital conducted a search of their sleep study records to find eligible participants. These records indicate that you have had at least one surgical procedure in an attempt to treat a sleep disorder, such as obstructive sleep apnea, snoring or similar condition. For this project we are interested in hearing about your own personal experiences and satisfaction with upper airway surgery.

Participation is Voluntary
Participation in any research project is voluntary. If you do not wish to take part, you are not obliged to. If you decide to take part and later change your mind, you are free to withdraw from the project at any stage without providing a reason. Your decision to take part, not to take part or to withdraw will not affect any routine treatment, your relationship with those treating you, or your relationship with The Queen Elizabeth Hospital or The Royal Adelaide Hospital.

What are the benefits associated with participation?
Results from this project will overcome a lack of knowledge that currently exists about the personal experiences and satisfaction of people who have undergone upper airway surgery. Although your participation will not result in any direct benefit for you, it may assist future patients, the health care community and health policy makers with information to draw upon in future decision-making processes regarding these types of surgery.

If I agree to participate, what will I be required to do?
Your participation requires the completion of the enclosed Upper Airway Surgery Questionnaire, which will take between 15-25 minutes to complete. You are welcome to examine the Questionnaire before you agree to participate. Other than a time commitment, completion of the questionnaire is at no cost to you. A reply paid envelope is provided for you to return the completed questionnaire.

What will happen to the information I provide?

Your answers are completely confidential and will be reported only as summaries in which no individual's answers can be identified. This survey is voluntary, and if you decide not to participate this will not affect the management of your health, now or in the future. The results of the research will be collated and analyzed in a research thesis, and may be submitted to a scientific journal for publication, presented at scientific and health policy conferences, and outlined in a media release. On the final page of the questionnaire there is a tick-box section where you can allow or disallow your questionnaire results to be 'linked' with your sleep study records. Your questionnaire results are extremely informative to us on their own but additional information might allow for further trends to be examined. This is explained fully on the final section. Again, individual responses will not be reported at any point, only collated group results. At this stage we feel it is important to assure you of the following points:

- **This university-based research team is completely independent and has no link with the relevant surgeons or surgical departments that may have carried out your surgery.**
- **Your personal details will remain confidential and your responses to this questionnaire will remain anonymous. These are requirements made by the ethics committees who have permitted this research and are obligations that are taken very seriously by this research team.**

You are welcome to receive a copy of the final, collated results of this research. If you would like to receive these results (once completed) please indicate so by ticking in the box provided at the end of the questionnaire.

If for some reason you prefer not to be involved, please let us know by writing your name on the questionnaire, then return the blank questionnaire in the enclosed reply paid envelope. This will ensure you are removed from the mailing list and not contacted again.

Who should I contact if I have any questions?

If you can have any questions or comments about this study, the project researcher would be happy to talk with you confidentially. Please contact Mr Adam Elshaug (project researcher) on (08) 8303 3577 during business hours, by e-mail at adam.elshaug@adelaide.edu.au or by writing to Adam Elshaug, Discipline of Public Health, Mail Drop 207, The University of Adelaide, South Australia, 5005.

How do I get started?

You can help us very much by taking the time to share your experiences and opinions about upper airway surgery. If you are happy to continue then please move on to the enclosed questionnaire. We would be particularly grateful if you could complete and return the questionnaire at your earliest convenience.

Thank you very much for helping with this important research.

Sincerely,

Adam Elshaug
Project Researcher

FIRST, SOME INFORMATION ABOUT YOU:

- **YOUR NAME (optional): .....**

Providing your name is entirely optional but will inform the research team that you have received this questionnaire. In doing so we can remove you from the mailing list so you do not receive the questionnaire again, or receive follow up phone calls after a period of time. Even if you prefer not to be involved, please let us know by writing your name on the questionnaire, then return the blank questionnaire in the enclosed reply paid envelope. This will ensure you are removed from the mailing list and not contacted again. This is the ONLY reason your name is collected; ALL responses to this questionnaire will remain anonymous and presented only as group results.

- **YOUR GENDER** (please circle): **MALE / FEMALE**

- **YOUR AGE** (in years as at January 1, 2006): **..... years**

- **How many upper airway surgical operations have you undergone as a treatment for Obstructive Sleep Apnea, snoring, or similar condition:** (that is, when you partially or completely stop breathing while asleep due to a closing of the airway)
  Note: By operation we mean 'one anaesthetized surgical event' only (even if multiple procedures were performed in one operation)

(please tick) ❑ One ❑ Two ❑ Three ❑ More than three

- **When was your last operation on the upper airway (as a treatment for Obstructive Sleep Apnea, snoring or similar):**

(please tick) ❑ Less than 1 year ago ❑ 1-2 years ago ❑ 2-3 years ago ❑ Over 3 years ago

SECTION 1: INFORMATION ABOUT SIDE EFFECTS RESULTING FROM YOUR LATEST UPPER AIRWAY SURGERY

IN SECTION 1, ALL QUESTIONS RELATE TO THE LATEST OPERATION YOU HAVE UNDERGONE

Item 1:
Did you experience side effects following surgery of the upper airway (please circle): **YES / NO**
If you are unsure a list of possible side effects has been provided in Item 2 over the page.
If yes, please continue and complete all items in section 1. If no, please move directly to section 2.

PLEASE TURN THE PAGE AND CONTINUE SECTION 1.

Item 2:
Below is a list of side effects that have previously been reported to occur following upper airway surgery. Please tick any that you experienced following your surgery (tick as many as necessary) and indicate the time period over which the side effect lasted (note: if a side effect comes and goes it is considered ongoing and should be grouped for the entire time it has existed):

(tick)	(please tick relevant time period)
❑ Pain on/around the surgical site	❑ Up to 2 weeks ❑ 2-8 weeks ❑ 2-6 months ❑ 6-12 months ❑ Over 12 months ❑ Do not recall
❑ Bleeding on/around the surgical site	❑ Up to 2 weeks ❑ 2-8 weeks ❑ 2-6 months ❑ 6-12 months ❑ Over 12 months ❑ Do not recall
❑ Severe swelling due to fluid (edema)	❑ Up to 2 weeks ❑ 2-8 weeks ❑ 2-6 months ❑ 6-12 months ❑ Over 12 months ❑ Do not recall
❑ Difficulty breathing	❑ Up to 2 weeks ❑ 2-8 weeks ❑ 2-6 months ❑ 6-12 months ❑ Over 12 months ❑ Do not recall
❑ Voice changes	❑ Up to 2 weeks ❑ 2-8 weeks ❑ 2-6 months ❑ 6-12 months ❑ Over 12 months ❑ Do not recall
❑ Difficulty swallowing	❑ Up to 2 weeks ❑ 2-8 weeks ❑ 2-6 months ❑ 6-12 months ❑ Over 12 months ❑ Do not recall
❑ Excessive dryness of throat	❑ Up to 2 weeks ❑ 2-8 weeks ❑ 2-6 months ❑ 6-12 months ❑ Over 12 months ❑ Do not recall
❑ Uncontrolled/excessive coughing	❑ Up to 2 weeks ❑ 2-8 weeks ❑ 2-6 months ❑ 6-12 months ❑ Over 12 months ❑ Do not recall
❑ Sensations of lumps in throat	❑ Up to 2 weeks ❑ 2-8 weeks ❑ 2-6 months ❑ 6-12 months ❑ Over 12 months ❑ Do not recall
❑ Infection at site of surgery	❑ Up to 2 weeks ❑ 2-8 weeks ❑ 2-6 months ❑ 6-12 months ❑ Over 12 months ❑ Do not recall
❑ Loss of taste	❑ Up to 2 weeks ❑ 2-8 weeks ❑ 2-6 months ❑ 6-12 months ❑ Over 12 months ❑ Do not recall
❑ Dizziness	❑ Up to 2 weeks ❑ 2-8 weeks ❑ 2-6 months ❑ 6-12 months ❑ Over 12 months ❑ Do not recall
❑ Numbness on/around surgical site	❑ Up to 2 weeks ❑ 2-8 weeks ❑ 2-6 months ❑ 6-12 months ❑ Over 12 months ❑ Do not recall
❑ Food or fluid discharge via nose	❑ Up to 2 weeks ❑ 2-8 weeks ❑ 2-6 months ❑ 6-12 months ❑ Over 12 months ❑ Do not recall

If other side effects occurred that are not present in this list please write them on the dotted lines below

❑	❑ Up to 2 weeks ❑ 2-8 weeks ❑ 2-6 months ❑ 6-12 months ❑ Over 12 months ❑ Do not recall
❑	❑ Up to 2 weeks ❑ 2-8 weeks ❑ 2-6 months ❑ 6-12 months ❑ Over 12 months ❑ Do not recall

PLEASE TURN PAGE AND CONTINUE SECTION 1

SECTION 1 (continued): INFORMATION ABOUT SIDE EFFECTS RESULTING FROM YOUR LATEST UPPER AIRWAY SURGERY

Item 3:
Taking into consideration your overall experience of pain and inconvenience, how would you rate your side effects:
(Inconvenience may incorporate time away from work, home or family duties; avoidance of socialising; being restricted from normal day-to-day activities etc)

PAIN (please tick one only) ❑ Extreme ❑ Substantial ❑ Moderate ❑ Mild ❑ None ❑ I do not recall

INCONVENIENCE (please tick one only) ❑ Extreme ❑ Substantial ❑ Moderate ❑ Mild ❑ None ❑ I do not recall

Item 4:
Were your side effects in line with what you expected, based on the information that was provided to you prior to surgery:
(tick one only)

- ❑ Definitely not, the side effects I experienced were much more severe than expected based on the information I received
- ❑ No, the side effects I experienced were more severe than expected based on the information I received prior to surgery
- ❑ The information I received was adequate and my experience of side effects was about in line with the information I received
- ❑ Yes, the side effects I experienced were less severe than expected based on the information I received prior to surgery
- ❑ Definitely yes, the side effects I experienced were far less severe than expected based on the information I received

Item 5:
Regarding side effects, are you satisfied with the level of communication and information that was provided to you prior to surgery: (tick one only)

- ❑ Not satisfied, based on my experience I believe more information should be provided for patients considering this surgery
- ❑ Satisfied, based on my experience I believe adequate information is provided for patients considering this surgery

END OF SECTION 1, PLEASE TURN PAGE AND START SECTION 2

SECTION 2: INFORMATION ABOUT SURGICAL EFFECTIVENESS

**Section 2 relates to the information you may have received prior to every upper airway operation you have undergone

Item 1:
A recommendation has been made that "surgeons should inform patients of the experimental nature of these operations". That is, upper airway surgical operations.

In discussions that you had prior to surgery, was this issue raised and explained to you: (tick one only)

- ❑ No, this issue was not raised or explained
- ❑ Yes, this issue was touched on but not explained clearly
- ❑ Yes, this issue was raised and explained clearly
- ❑ I do not recall

Item 2:
In discussions that you had prior to surgery, what were you told was the likelihood that your surgery would be successful in treating SNORING: (tick one only)

- ❑ Almost certain to completely eliminate snoring
- ❑ A good to high chance of eliminating snoring
- ❑ Perhaps a 50/50 chance of eliminating snoring
- ❑ A good to high chance snoring would NOT be eliminated but snoring would be reduced
- ❑ Almost certain that snoring would NOT be eliminated but a small chance snoring may be reduced
- ❑ This matter was not raised or explained
- ❑ I do not recall

PLEASE TURN PAGE AND CONTINUE SECTION 2

SECTION 2 (continued): INFORMATION ABOUT SURGICAL EFFECTIVENESS

Item 3:
In discussions that you had prior to surgery, what were you told was the likelihood that your surgery would be successful in treating OBSTRUCTIVE SLEEP APNEA, or any associated form of sleep disordered breathing (this is when you partially or completely stop breathing while asleep due to a closing of the airway): (tick one only)

- ❑ Almost certain to completely eliminate obstructive sleep apnea / sleep disordered breathing
- ❑ A good to high chance of eliminating obstructive sleep apnea / sleep disordered breathing
- ❑ Perhaps a 50/50 chance of eliminating obstructive sleep apnea / sleep disordered breathing
- ❑ A good to high chance obstructive sleep apnea/sleep disordered breathing would NOT be eliminated but it would be reduced
- ❑ Almost certain that obstructive sleep apnea/sleep disordered breathing would NOT be eliminated but a small chance it may be reduced
- ❑ This matter was not raised or explained
- ❑ I do not recall

Item 4:
Regarding information provided to you about surgical effectiveness, are you satisfied with the level of communication and information that was provided to you prior to surgery: (tick one only)

- ❑ Not satisfied, I believe more information should be provided for patients considering this surgery
- ❑ Satisfied, I believe adequate information is provided for patients considering this surgery

PLEASE TURN PAGE AND CONTINUE SECTION 2

Item 5:
Since having surgery, has it been recommended that you still need further treatment for obstructive sleep apnea (or similar condition). This may include more surgery, CPAP, dental device (mouth guard/splint) or weight loss: (select as many as necessary)

❑ Yes, further treatment was/has been recommended (select) ❑ More surgery ❑ Dental device ❑ CPAP ❑ Weight loss

❑ No further treatment was/has been recommended

END OF SECTION 2, PLEASE TURN PAGE AND START SECTION 3 (final section)

SECTION 3 (final section): SATISFACTION WITH SURGICAL EXPERIENCE AND OUTCOME

**Important: Section 3 relates to your satisfaction with ALL upper airway operations and procedures you have undergone

Item 1:
In your opinion, how successful was your surgery (or surgeries) at improving your sleep quality:

- ❑ Completely unsuccessful
- ❑ Mostly unsuccessful
- ❑ Somewhat unsuccessful
- ❑ Somewhat successful
- ❑ Mostly successful
- ❑ Completely successful

Item 2:
Overall, how satisfied are you with the surgery or surgeries you had for obstructive sleep apnea or similar condition: (tick one)

- ❑ Completely dissatisfied
- ❑ Mostly dissatisfied
- ❑ Somewhat dissatisfied
- ❑ Neither satisfied nor dissatisfied
- ❑ Somewhat satisfied
- ❑ Mostly satisfied
- ❑ Completely satisfied

PLEASE TURN PAGE AND CONTINUE SECTION 3 (final section)

SECTION 3 (final section, continued): SATISFACTION WITH SURGICAL EXPERIENCE AND OUTCOME

Item 3:
Given your own experiences, do you regret undergoing upper airway surgery: (tick one only)

- ❑ Yes, I regret having surgery
- ❑ Indifferent
- ❑ No, I do not regret having surgery

Item 4:
Based on your own experiences, would you recommend upper airway surgery to a family member or close friend who has obstructive sleep apnea or a similar condition:

- ❑ No, I would strongly recommend against surgery
- ❑ I would advise against surgery
- ❑ I would make no recommendation one way or another
- ❑ I would advise in favour of surgery
- ❑ Yes, I would strongly recommend surgery

ALMOST THERE, PLEASE TURN OVER THE PAGE TO COMPLETE THE QUESTIONNAIRE...

****IMPORTANT: Please complete the items below and place your completed questionnaire in the return envelope provided and post at your earliest convenience.** Remember you can receive a report of the group results (once completed) by indicating below.

❑ Yes, I would like to receive a copy of the final, collated results of this research*

Name and address (please print clearly): .....

.....

* Note: If this box is not ticked then this is taken as a no, and the report will not be sent.

Important: By completing this questionnaire you have provided us with important and useful information. However, at the moment we do not know how your questionnaire responses compare to your sleep study results. In the future, this research team may be interested in linking your questionnaire results with your sleep study data to see if group trends exist. For example, we could see if a certain type of surgery produces higher or lower levels of satisfaction or, if certain levels of satisfaction correspond with certain sleep study outcomes (following surgery). If this were done you would always remain anonymous (that is, your name and other details would NEVER be divulged). This would allow any relationship between certain questionnaire results and certain sleep study results to be explored in more detail. This process is commonly known as data linkage and the same high standards of participant confidentiality and anonymity are observed when data linkage occurs. Knowing that the same high standards of anonymity and confidentiality are observed, do you consent for this research team to link your questionnaire and sleep study data in the future:

❑ Yes, I give consent for my questionnaire results and my sleep study data to be linked

❑ No, I do not give consent for my questionnaire results and my sleep study data to be linked

You have finished, congratulations and thank you for your participation! You may wish to go over your answers again to check that you are happy with your responses. If completing this questionnaire has made you feel anxious or if you have any questions, you are welcome to discuss any matters confidentially with Mr Adam Elshaug (Project Researcher) at adam.elshaug@adelaide.edu.au or (08) 8303 3577 or at the address provided on the introductory letterhead. Alternatively, any enquiries of a medical nature should be directed to your General Medical Practitioner.

Appendix Three

Policy Stakeholder Interviews:
Introductory Letter
Preparatory Reading
Interview Schedule

THE UNIVERSITY OF ADELAIDE
AUSTRALIA

DISCIPLINE OF PUBLIC HEALTH
FACULTY OF HEALTH SCIENCES

LEVEL 9 TOWER BUILDING
10 PULTENEY ST
(MAIL DROP 207)
UNIVERSITY OF ADELAIDE
ADELAIDE SA 5005

Dear .....,

I am currently a PhD student working in the Discipline of Public Health at The University of Adelaide on a project with Mr John Moss (Head of Department) and Professor Janet Hiller as supervisors. I am writing to you today as we have identified you as a significant stakeholder within Australian health policy and as such would like to request your involvement in an interview regarding clinical effectiveness and health policy.

The aim of the interview is to explore your perspectives of a surgical case study for the treatment of Obstructive Sleep Apnoea Syndrome, then on more general issues relating to clinical and cost effectiveness and health policy within Australia. If you agree to participate it will require that you read approximately 3 pages of preparatory material beforehand (attached). The interview process will be semi-structured. That is, there are 12 questions that I will ask and you can respond in as little or as much detail as you wish. I may ask supplementary questions or prompt you for more information as we progress. I anticipate the interview will take between 15 to 30 minutes.

If you agree to be involved your answers will be incorporated in to a report that will be published in the form of a PhD thesis and also a peer-reviewed journal publication. Importantly, your name, location and job title/description will NOT be divulged. However, I will ask if you would be willing to suggest a nonspecific description (of current and/or past roles) that may be reported to broadly illustrate to the reader your involvement in Australian health policy? With your permission the interview will be recorded onto audiotape where only I will listen to it in order to facilitate an accurate transcribing process?

If you agree to take part in the study, by way of an interview, I am committed to working in with a time that is most convenient for you. The interview can take place in person at a location convenient for you, or if you prefer, by telephone. If you would like to be involved, please contact me at any time on (08) 8303 3577 or 0403 789 397, or email at adam.elshaug@adelaide.edu.au to arrange a time for an interview.

I look forward to your reply and for your participation in the study,
Best wishes
Adam Elshaug

Preparatory Reading Material

***Important Note:** The following material is currently either 'in press' or under review in the *Journal of Evaluation in Clinical Practice* and *SLEEP*. As such we request that this material is kept strictly confidential and is not seen by anyone other than you.

Background: Obstructive Sleep Apnoea Syndrome (OSA) is a health condition characterized by recurring upper airway obstructions in sleep that lead to hypoxia (oxygen desaturation), neurological arousals, sympathetic discharges and increased cardiac load. OSA has been well-documented to cause hypertension [1-3], cardiac morbidity and mortality [4], automobile accidents [5], neurocognitive deficits [6], as well as impaired quality of life [7], and increasingly, glucose intolerance [8]. The condition reciprocally influences, and is influenced by co-morbid health states [9,10]. In 2004, Young et al [11] reported that in the USA, approximately 1 in 5 adults has at least mild OSA and 1 in 15 adults has OSA of moderate or worse severity.

Polysomnography (PSG) is the laboratory procedure for best studying sleep and its variable dysfunctions, including OSA. PSG involves an overnight 'wire-up' where neurological and cardio-respiratory variables are monitored. The diagnosis of OSA is primarily based on the number of breathing abnormalities that occur per hour of sleep (known as the Apnoea/ Hypopnoea index; AHI). Apnoea represents the full cessation in breathing, whereas a hypopnoea is a partial cessation. **According to the peak international sleep medicine body[12,13] OSA severity is measured by the AHI cut points of 5+ (mild), 15+ (moderate), and 30+ (severe). An AHI of 5 or less is deemed within normal limits and confers a negative diagnosis (or indicates effective treatment).** Contemporary evidence supports these classifications, demonstrating the importance of reducing the AHI, in many cases to near or below five events per hour of sleep (thereby controlling OSA) in order to improve numerous physiological, health outcome and quality of life measures. This has been demonstrated in cardiovascular disease[14,15], heart failure[16], endocrinology[17-20], health related quality of life[21-23].

Treatment options for OSA include weight loss, Continuous Positive Airway Pressure (CPAP-considered the gold-standard), mandibular advancement splints (mouth guards) and upper airway surgical procedures. Currently there are multiple surgical interventions utilized in the treatment of adult OSA. Surgery types are broadly categorized into Phase I and Phase II procedures, where Phase I refers to procedures of the soft palate and Phase II as procedures of the hard palate. Phase I procedures are generally less severe in that they often take place as day procedures or up to 1-2 days of inpatient recovery whereas Phase II procedures are more invasive, often requiring multiple days in intensive care followed by general ward recovery. The Cochrane Review on surgery for OSA (to the year 2004) concludes:

> *Clinicians should consider restricting surgery for obstructive sleep Apnoea to that carried out as part of clinical trials. Where practice is continued, patients should be informed of the experimental nature of the operations. Patients should be told that there is a lack of good trial based evidence of the efficacy of surgery for obstructive sleep Apnoea, a course of action that may restrict the use of these operations.* [24]

A data search of the Australian Government Health Insurance Commission web site has shown that throughout Australia the use of these procedures is widespread and increasing, with over 3,000 conducted in the calendar year 2005.

Methods: Our research team conducted a multi-centre, retrospective clinical-record audit within two major Australian teaching hospitals. A retrospective analysis period of 4.5 years was undertaken (January 1, 2001 to June 30, 2005). Concurrent, unselected surgical cases who attended the sleep disorder clinics of the two hospitals were identified (actual surgeries were performed by various surgeons at various locations). Inclusion criteria: Cases acted as their own

historical controls. To be included in this cohort required a case to have completed all of the following: (1) a pre-surgical baseline/diagnostic PSG wherein OSA status and severity was established, (2) at least one stage of upper airway surgery in an attempt to treat OSA and, (3) a post-surgical reassessment PSG to gauge surgical effectiveness. Where a pre-surgery CPAP therapy trial (with PSG) occurred, these data were also captured to enable a CPAP treatment comparison arm. Case information and clinical data were de-identified and audited. Data pertinent to surgery effectiveness (i.e. the AHI) were collected from overnight PSG records.

Results: Ninety-four (94) surgical cases met the inclusion criteria and comprise this cohort (79 males and 15 females). These patients ranged in age (as at first operation) from 19 – 75 years, with a median age of 46 years (SD = 12.8). Sixty-four of these patients (55 Phase I and 9 Phase II recipients) also underwent the additional CPAP therapy arm prior to any surgery.

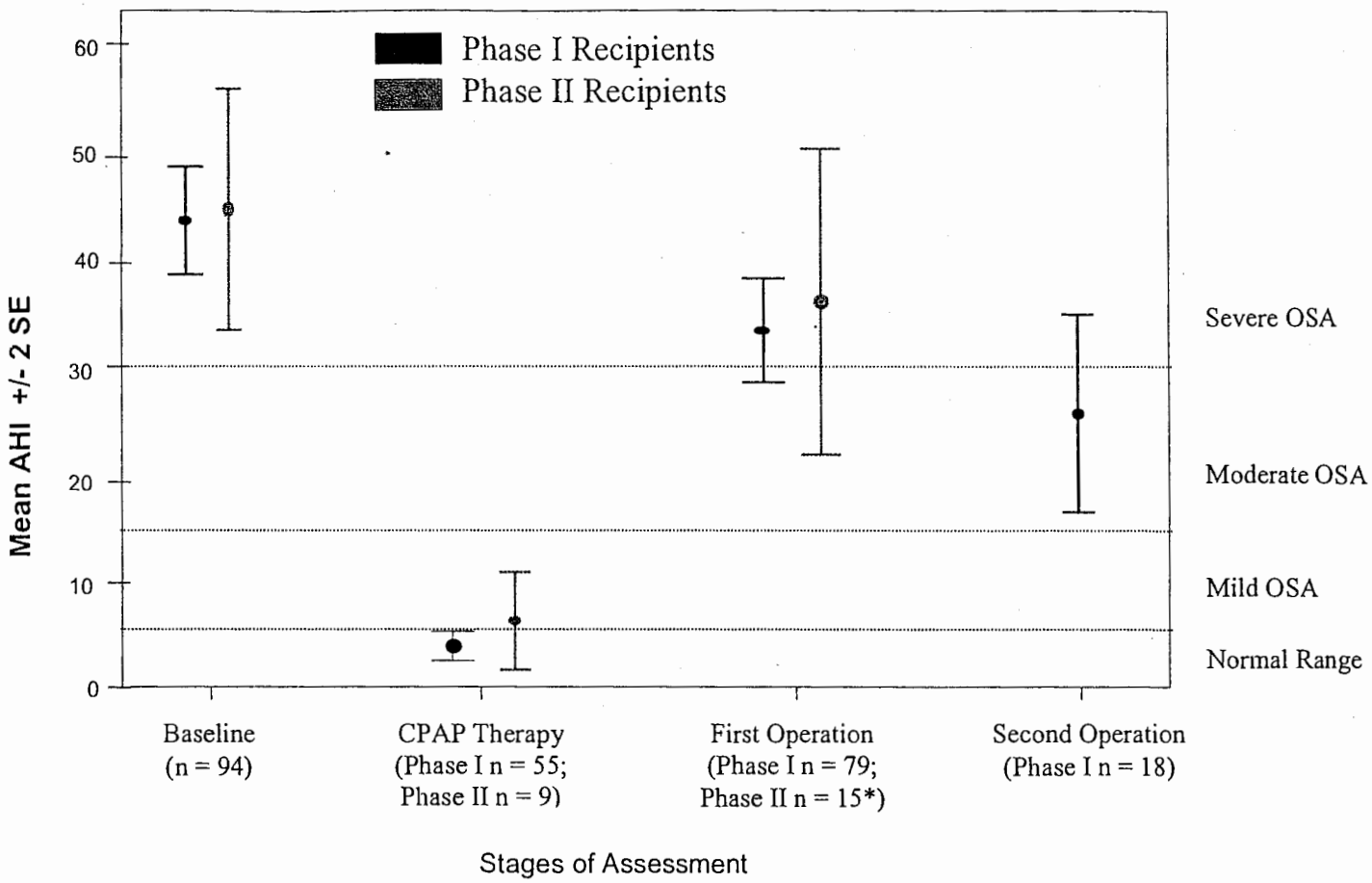
Procedural variability: This cohort was exposed to 12 differing, individually classified surgical types. Results on surgical variability (n=94) demonstrate that this cohort underwent a total of 184 individually classified procedures at their first operation, with a mean of 2.5 (range 1-7) individual upper airway surgical procedures per person. Eighteen Phase I recipients went on to undergo a second operation. Here, this group experienced a further 36 individually classified procedures (mean 1.8 per person; range 1-4) taking the cumulative mean for this sub-group to 4 Phase I (soft palate) procedures per person (range 3-7) over 2 operations. **In total this cohort of 94 individuals received 41 varying combinations of individually classified surgical procedures in an attempt to treat their OSA.**

Surgery effectiveness: In line with best practice, the primary outcome measure utilized to gauge effectiveness is the AHI. For all surgery recipients combined (i.e. Phase I and II), results demonstrate that on average, 1 operation (where a mean of 2.5 individual procedures occurred per person) reduced OSA severity by 20% (patients still had severe OSA) and 2 operations (where a cumulative mean of 4 individual procedures occurred per person) reduced OSA severity by 35% (still moderate OSA). Potential confounders, such as increased weight gain and more/less time spent asleep in the supine position were not significantly different from baseline to surgery assessment and as such can be ruled out. The following tables and graphs further illustrate the results:

Table: Comparative success (fail) rates of the various therapies, as measured by the AHI.

Therapy	% Success (Fail) *Success = AHI ≤ 5*	% Success (Fail) *Success = AHI ≤ 10*
CPAP (n = 64)	72 (28)	97 (3)
All Surgery Combined	13 (87)	27 (73)
Phase II Procedures (n = 15) Note: 7 also had adjunctive Phase I at same time)	20 (80)	20 (80)
Phase I Procedures (n = 79) mean of 2.5 procedures per person in 1 operation	8 (92)	20 (80)
Cumulative Phase I Procedures (n = 18) mean of 4 procedures per person over 2 operations	18 (82)	35 (65)

Figure: Mean (+/- 2 standard errors) apnoea-hypopnoea index (AHI) by group (phase I and phase II recipients) at baseline assessment, at CPAP trial and following surgical operations.



Discussion: This study is the third largest of its kind ever conducted in the world and the largest ever in Australia. More importantly it is the first in the world to investigate procedural variability, in this case from a catchment of some two-dozen surgeons and clinics within one localized health system. These results indicate substantial procedural variability in the application of surgery as a treatment for OSA, with 41 differing surgery combinations performed on 94 individuals. Further, it is evident that the application of these procedures resulted in limited clinical effectiveness with a 13% success rate (87% fail), based on the widely accepted criterion of a reduction in the AHI to 5 or less. At an AHI of 10 or less the success rate was 27% (73% fail).

The second study: Is a meta-analysis of 18 published papers (n=385) examining both Phase I and II procedures for effectiveness. In this review we compare the success rates of procedures using various definitions of success. Specifically, according to the definition applied in many surgery publications (i.e. 50% reduction in Apnoea Hypopnoea Index (AHI) and/or ≤ 20) the pooled success rate for Phase I procedures is 55% (45% fail). However, at AHI ≤ 10 success reduces to 31.5% (68.5% fail) and at AHI ≤ 5, success is reduced to 13% (87% fail). According to these definitions, Phase II success (fail) rates decrease from 86% (14%) to 45% (55%) and 43% (57%) respectively. We contend that consensus is required between the specialties regarding the definition of surgical success as the AASM definition of AHI ≤ 5 may have entirely different policy implications regarding effectiveness, appropriateness etc compared to that reported within surgical papers. More detail of methodology, Meta-analysis forest plots, statistics and a list of articles included in the meta-analysis can be provided upon request.

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References:

1. Grote L, Ploch T, Heitman J, Knacck L, Penzel T, Peter JH. Sleep-related breathing disorder is an independent risk factor for systemic hypertension. *American Journal of Critical Care Medicine* 1999;**160**:1875-1882.
2. Lavie P, Herer P, Hoffstein V. Obstructive sleep apnoea syndrome as a risk factor for hypertension: population study. *BMJ* 2000;**320**:479-482.
3. Peppard PE, Young T, Palta M, Skatrud J. Prospective study of the association between sleep-disordered breathing and hypertension. *N Eng J Med* 2000;**342**:1378-1384.
4. Peker Y, Hedner J, Norum J, Kraiczi H, Carlson J. Increased incidence of cardiovascular disease in middle-aged men with obstructive sleep apnea: a 7-year follow-up. *Am J Respir Crit Care Med* 2002;**166**(2):159-65.
5. Teran-Santos J, Jimenez-Gomez A, Cordero-Guevara J. The association between sleep apnea and the risk of traffic accidents. Cooperative Group Burgos-Santander. *N Eng J Med* 1999;**340**(11):847-851.
6. Kim H, Young T, Mathews G, et al. Sleep-disordered breathing and neuropsychological deficits. *American Journal of Critical Care Medicine* 1997;**156**:1813-1819.
7. Sateia MJ. Neuropsychological impairment and quality of life in obstructive sleep apnea. *Clin Chest Med* 2003;**24**(2):249-59.
8. Punjabi NM, Sorkin JD, Katzel LI, Goldberg AP, Schwartz AR, Smith PL. Sleep-disordered breathing and insulin resistance in middle-aged and overweight men. *Am J Respir Crit Care Med* 2002;**165**(5):677-82.
9. Lavie P, Herer P, Peled R, et al. Mortality in sleep apnea patients: a multivariate analysis of risk factors. *Sleep* 1995;**18**(3):149-157.
10. Lindberg E, Janson C, Svardsudd K, Gislason T, Hetta J, Boman G. Increased mortality among sleepy snorers: a prospective population based study. *Thorax* 1998;**53**(8):631-637.
11. Young T, Skatrud J, Peppard P. Risk Factors for Obstructive Sleep Apnea in Adults. *JAMA* 2004;**291**(16):2013-2016.
12. Chesson A, Ferber R, Fry J, Grigg-Damberger M, Hartse K, et al. Practice parameters for the indications for polysomnography and related procedures. Polysomnography Task Force, American Sleep Disorders Association Standards of Practice Committee. *Sleep* 1997;**20**(2):406-422.
13. The Report of an American Academy of Sleep Medicine Task Force. Sleep-related breathing disorders in adults: recommendations for syndrome definition and measurement techniques in clinical research. *Sleep* 1999;**22**(5):667-689.
14. Becker H, Jerrentrup A, Ploch T, et al. Effect of nasal continuous positive airway pressure treatment on blood pressure in patients with obstructive sleep apnea. *Circulation* 2003;**107**(1):68-73.
15. Phillips B. Sleep-disordered breathing and cardiovascular disease. *Sleep Med Rev* 2005;**9**(2):131-40.
16. Mansfield DR, Gollogly NC, Kaye DM, Richardson M, Bergin P, Naughton MT. Controlled trial of continuous positive airway pressure in obstructive sleep apnea and heart failure. *Am J Respir Crit Care Med* 2004;**169**(3):361-6.
17. Hassaballa HA, Tulaimat A, Herdegen JJ, Mokhlesi B. The effect of continuous positive airway pressure on glucose control in diabetic patients with severe obstructive sleep apnea. *Sleep and Breathing* 2005;**9**(4):176-180.
18. Meston N, Davies R, Mullins R, Jenkinson C, Wass J, Stradling J. Endocrine effects of nasal continuous positive airway pressure in male patients with obstructive sleep apnoea. *J Intern Med* 2003;**254**(5):447-454.
19. Harsch I, Schahin P, Radespiel-Troger M, et al. Continuous positive airway pressure treatment rapidly improved insulin sensitivity in patients with obstructive sleep apnea syndrome. *American Journal of Critical Care Medicine* 2004;**169**:156-162.
20. Babu A, Hedergen J, Fogelfeld L, Shott S, Mazzone T. Type 2 diabetes, glycemic control, and continuous positive airway pressure in obstructive sleep apnea. *Archives of Internal Medicine* 2005;**165**:447-452.
21. Habukawa M, Uchimura N, Nose I, et al. Emotional states and quality of life in patients with obstructive sleep apnea. *Sleep and Biological Rhythms* 2005;**3**(3):99-105.
22. Sin DD, Mayers I, Man GC, Ghahary A, Pawluk L. Can continuous positive airway pressure therapy improve the general health status of patients with obstructive sleep apnea?: a clinical effectiveness study. *Chest* 2002;**122**(5):1679-85.
23. Parish JM, Lyng PJ. Quality of life in bed partners of patients with obstructive sleep apnea or hypopnea after treatment with continuous positive airway pressure. *Chest* 2003;**124**(3):942-7.
24. Bridgman S, Dunn K, Ducharme F. Surgery for obstructive sleep apnoea (Cochrane Review). The Cochrane Library. Chichester, UK: John Wiley & Sons, Ltd, 2004.

Policy Stakeholder Interview Schedule

Key Question	Factors Requiring Exploration
Do you have any questions or points for clarification that came out of the pre reading material?	
What are your impressions after reading the material?	• Rank most prominent
Do you believe these surgeries represent clinical effectiveness?	• Safety? • Achieving intended outcome? • Health outcomes met? • Good value for money?
Do you believe these surgical procedures should continue to be available to the public: **yes** (with or without rules or provisions), **no** or **undecided**?	• If **yes**, with any provisions or rules? • How would you implement and monitor these to ensure they are effective and not open to misuse or neglect? • If **no**, is that under no circumstances at all? • How should this 'ban' be enacted-detail? • If **undecided**, what further information would you want in order to inform your decision?
Are you aware of other specific examples where a clinical service was deemed to be of limited effectiveness and was successfully withdrawn from MBS reimbursement/phased out?	• How did this happen? • What did the process entail? • Which stakeholder's opinions were represented (and were any over or under represented)? • Was there resistance and by whom-was it valid, how long did the process take, • Was it an effective process, can it be improved?
Do you believe that there are other current examples of existing, Medicare funded health care services that may be of questionable clinical and cost-effectiveness?	• Name example • What barriers exist to it being phased out? • Do you consider this to be a problem? • Should this broad issue receive low or high priority in Australian health policy, why?
Building on this, do you believe Australia has a policy mechanism in place that can adequately address this surgery example, and other examples of existing practices that may be of questionable clinical and cost-effectiveness?	• Please provide detail of how the body and/or the policy interact to effect this
Regarding your last answer, do you believe there are strengths as well as limitations or barriers in existing framework or policy processes?	• If so please provide detail of strengths, weaknesses, limitations, areas for improvement, role of vested interests…?

1

Key Question	Factors Requiring Exploration
In your opinion, what is the best way forward from a policy perspective (to resolve this surgery case, and others like it), and this may involve working within the existing framework or amending it, establishing investigative bodies, seeking out specific information etc?	
In this process, what roles do and/or should advocacy and interest groups play?	• How important is the role of patient perspectives, interests of clinicians (clinical autonomy) in facilitating or presenting barriers to phasing out of ineffective practices? • Perhaps rank these
What is the role of research and evaluation in resolving this sort of issue?	• Should there be more engagement between all parties – policy advisors, academic research groups, clinicians, patients? • How?
How do think these issues should best be brought to the attention of policy makers?	• What would be the best way to ensure implementation
Is there anything that we should have talked about but we did not, or that you would like to raise or comment further on?	
Are you willing to suggest a nonspecific job description (of current and/or past roles) that may be reported to broadly illustrate to the reader your involvement in Australian health policy?	

Appendix Four

Letters of Acceptance for 'in-press' Journal Articles

Date: Thu, 19 Jul 2007 21:59:29 -0700 (PDT)
From: Mabel Chew <mchew@bmj.com>
Subject: Change page "Upper airway surgery should not be first-line therapy for adult obstructive sleep apnoea" 500629
To: Adam G Elshaug <adam.elshaug@adelaide.edu.au>
Cc: John R Moss <john.moss@adelaide.edu.au>, Guy J Maddern <guy.maddern@adelaide.edu.au>, Janet E Hiller <janet.hiller@adelaide.edu.au>

Subject: BMJ- Manuscript BMJ/2007/500629

Dear Mr. Elshaug

I am delighted to tell you that your article has been accepted for publication in the BMJ. Thank you very much for submitting it to us—we are very pleased to be able to publish it.

We will aim to publish it as soon as we can—usually within about eight weeks, and often much quicker. We do try to publish papers on the same subject together, so there may be some variation in that timing. We will aim to keep you informed of progress.

_X__ A Cross (X) here means that there is still some outstanding information we need from you before we can go ahead and edit your article. Please see below for details (also marked with an X).

If your article is of a type where we send you a proof before publication (see below), the next thing that will happen is that you will receive an edited version from one of our technical editors. Please do return it as soon as possible, so as not to delay publication. In the meantime, if you have any questions please do contact me.

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Date: Fri, 21 Sep 2007 00:49:30 -0600
From: "Dr. Egon Jonsson" <ejonsson@ihe.ca>
Subject: Manuscript submission International Journal of Technology Assessment in Health Care
To: 'Adam Elshaug' <adam.elshaug@adelaide.edu.au>

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The review process of your manuscript: "Exploring Policymakers' Perspectives on Disinvestment.." now is finished.

I am very pleased to inform you of the positive response of the reviewers. They recommend your manuscript for publication on the basis that it addresses an extremely important and challenging area of health technology assessment practice, i.e. how HTA can promote the disinvestment in redundant and superseded treatments and practice. Your findings are clearly presented and placed in the context of well balanced discussion. The paper outlines some useful future directions of the greater implementation of disinvestment practice in technology assessment procedures and also some areas for potential future methodological underpinning of HTA for disinvestment.

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Appendix Five

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NOTE: Statement of authorship appears in the print copy of the thesis held in the University of Adelaide Library.

References (chapters 1, 5 and 9 only – all others within manuscripts)

1. White KL. Evidence-Based Medicine. *Lancet* 1995;346(8978):837-838.
2. Smith R. Where is the wisdom...? The Poverty of Medical Evidence. *BMJ* 1991;303(6806):798-799.
3. White K. Evidence-Based Medicine. *Lancet* 1995;346:837-838.
4. Smith R. Where is the wisdom...? The Poverty of Medical Evidence. *BMJ* 1991;303(6806):798-799.
5. Wennberg JE. Unwarranted Variations in Health Care: The Dartmouth Atlas of Health Care, 2000. Electronic resource: http://www.dartmouthatlas.org/lectures/overview_0.php.
6. Kuhn TS. *The Structure of Scientific Revolution*. 2 ed. Chicago: University of Chicago Press, 1970.
7. Buchan H. Gaps between best evidence and practice: causes for concern. *Medical Journal of Australia* 2004;180((6 Suppl)):S48-S49.
8. Ferlie E, Shortell S. Improving the quality of health care in the United Kingdom and the United States: a framework for change. *Milbank Q* 2001;79(2):281-315.
9. Chassin MR, Galvin RW. The urgent need to improve health care quality. Institute of Medicine National Roundtable on Health Care Quality. *Jama* 1998;280(11):1000-5.
10. Williams A. Health economics: the end of clinical freedom? *BMJ* 1988;297:1183-1186.
11. Medical Services Advisory Committee Australian Government Department of Health and Ageing. Strengthening evidence-based health care in Australia, 2003.
12. Pharmaceutical Benefits Advisory Committee Australian Government Department of Health and Ageing. PBAC Outcomes, 2003.
13. Maynard A, McDaid D. Evaluating health interventions: exploiting the potential. *Health Policy* 2003;63(2):215-226.
14. Grol R, Grimshaw J. From best evidence to best practice: effective implementation of change in patients' care. *Lancet* 2003;362(9391):1225-30.
15. Cabana M, Rand C, Powe N, Wu AW, Wilson MH, Abboud PA, et al. Why don't physicians follow clinical practice guidelines? A framework for improvement. *JAMA* 1999;282(15):1458-1465.
16. Prochaska JO, Velicer WF. The transtheoretical model of health behavior change. *Am J Health Promot* 1997;12(1):38-48.
17. Davis D, Evans M, Jadad A, Perrier L, Rath D, Ryan D, et al. The case for knowledge translation: shortening the journey from evidence to effect. *Bmj* 2003;327(7405):33-5.
18. Riemsma RP, Pattenden J, Bridle C, Sowden AJ, Mather L, Watt IS, et al. A systematic review of the effectiveness of interventions based on a stages-of-change approach to promote individual behaviour change. *Health Technol Assess* 2002;6(24):1-231.
19. Green LW, Eriksen MP, Schor EL. Preventive practices by physicians: behavioral determinants and potential interventions. *Am J Prev Med* 1988;4(4 Suppl):101-107.
20. Green LW, Kreuter MW. *Health promotion planning: an educational and environmental approach*. 2nd ed. Mountain View, Calif: Mayfield, 1991.
21. Davis DA, Thomson MA, Oxman AD, Haynes RB. Changing physician performance. A systematic review of the effect of continuing medical education strategies. *Jama* 1995;274(9):700-5.
22. Solomon DH, Hashimoto H, Daltroy L, Liang MH. Techniques to improve physicians' use of diagnostic tests: a new conceptual framework. *Jama* 1998;280(23):2020-7.
23. Solberg LI. Guideline implementation: what the literature doesn't tell us. *Jt Comm J Qual Improv* 2000;26(9):525-37.
24. Oxman A, Flottorp S. An overview of strategies to promote implementation of evidence based health care. In: Silagy C, Haines A, editors. *Evidence based practice in primary care*. London: BMJ Books, 2001.
25. Grol R, Wending M. What drives change? Barriers to and incentives for achieving evidence-

based practice. *Medical Journal of Australia* 2004;180(6 suppl):S57-S60.
26. Dijkstra RF, Braspenning JC, Uiters E, van Ballegooie E, Grol RT. Perceived barriers to the implementation of diabetes guidelines in hospitals in The Netherlands. *Neth J Med* 2000;56(3):80-5.
27. Krieger J, McNicholas WT, Levy P, De Backer W, Douglas N, Marrone O, et al. Public health and medicolegal implications of sleep apnoea. *Eur Respir J* 2002;20(6):1594-609.
28. Shamsuzzaman A, Gersh B, Somers V. Obstructive sleep apnea: implications for cardiac and vascular disease. *JAMA* 2003;290(14):1906-1914.
29. Newman A, Nieto J, Guidry U, Lind B, Redline S, Shahar E, et al. Relation of Sleep-disordered Breathing to Cardiovascular Disease Risk Factors: The Sleep Heart Health Study. *American Journal of Epidemiology* 2001;154(1):50-59.
30. Young T, Palta M, Dempsey J, Skatrud J, Weber S, Badr S. The occurrence of sleep-disordered breathing among middle-aged adults. *N Eng J Med* 1993;328:1230-1235.
31. Olson L, King M, Hensley M, Saunders N. A community study of snoring and sleep-disordered breathing. Prevalence. *Am J Respir Crit Care Med* 1995;152:711-716.
32. Young T, Peppard P, Gottlieb D. Epidemiology of obstructive sleep apnea: A population health perspective. *American Journal of Respiratory and Critical Care Medicine* 2002;165(9):1217-1239.
33. Gami A, Caples S, Somers V. Obesity and obstructive sleep apnea. *Endocrinol Metab Clin North Am* 2003;32(4):869-894.
34. Peppard PE, Young T, Palta M, Dempsey J, Skatrud J. Longitudinal study of moderate weight change and sleep-disordered breathing. *Jama* 2000;284(23):3015-21.
35. Li K, Kushida C, Powell N, Riley R, Guilleminault C. Obstructive sleep apnea syndrome: a comparison between Far-East Asian and white men. *Laryngoscope* 2000;110(10):1689-1693.
36. Chen W, Kushida C. Nasal obstruction in sleep disordered breathing. *Otolaryngol Clin North Am* 2003;36:437-460.
37. Bridgman S, Dunn K, Ducharme F. Surgery for obstructive sleep apnoea (Cochrane Review). *The Cochrane Library*. Chichester, UK: John Wiley & Sons, Ltd, 2004.
38. Young T, Finn L, Austin D, Peterson A. Menopausal status and sleep-disordered breathing in the Wisconsin Sleep Cohort Study. *Am J Respir Crit Care Med* 2003;167(9):1181-5.
39. Bixler EO, Vgontzas AN, Lin HM, Ten Have T, Rein J, Vela-Bueno A, et al. Prevalence of sleep-disordered breathing in women: effects of gender. *Am J Respir Crit Care Med* 2001;163(3 Pt 1):608-13.
40. Shahar E, Redline S, Young T, Boland L, Baldwin C, Nieto F, et al. Hormone replacement therapy and sleep-disordered breathing. *Am J Respir Crit Care Med* 2003;167(9):1186-1192.
41. Polo-Kantola P, Rauhala E, Helenius H, Erkkola R, Irjala K, Polo O. Breathing during sleep in menopause: a randomized, controlled, crossover trial with estrogen therapy. *Obstet Gynecol* 2003;102(1):68-75.
42. Young T, Skatrud J, Peppard P. Risk Factors for Obstructive Sleep Apnea in Adults. *JAMA* 2004;291(16):2013-2016.
43. He J, Kryger M, Zorick F, Conway W, Roth T. Mortality and apnea index in obstructive sleep apnea. Experience in 385 male patients. *Chest* 1988;94(1):9-14.
44. Lindberg E, Janson C, Svardsudd K, Gislason T, Hetta J, Boman G. Increased mortality among sleepy snorers: a prospective population based study. *Thorax* 1998;53(8):631-637.
45. Lavie P, Herer P, Peled R, Berger I, Yoffe N, Zomer J, et al. Mortality in sleep apnea patients: a multivariate analysis of risk factors. *Sleep* 1995;18(3):149-157.
46. Zaninelli A, et al. Snoring and risk of cardiovascular disease. *International Journal of Cardiology* 1991;32(3):347-351.
47. Hla K, Young T, Bidwell T, Palta M, Skatrud J, Dempsey J. Sleep apnea and hypertension.

A population-based study. *Ann Intern Med* 1994;120(5):382-388.
48. Young T, Finn L, Hla K, Morgan B, Palta M. Snoring as part of a dose-response relationship between sleep-disordered breathing and blood pressure. *Sleep* 1996;19(10 Suppl):S202-205.
49. Nakano H, Hayashi M, Ohshima E, Onizuka A. Relationship between sleep-disordered breathing and hypertension. *Sleep and Biological Rhythms* 2003;1(2):115-116.
50. Palomaki H, Partinen M, Erkinjuntti T, Kaste M. Snoring, sleep apnea syndrome, and stroke. *Neurology* 1992;42(7 Supp 6):75-82.
51. Lavie P. Hypertension in sleep apnea syndrome: lessons from nearly 8,000 male patients. *Sleep* 1999;22:S104.
52. Sateia MJ. Neuropsychological impairment and quality of life in obstructive sleep apnea. *Clin Chest Med* 2003;24(2):249-59.
53. Punjabi NM, Sorkin JD, Katzel LI, Goldberg AP, Schwartz AR, Smith PL. Sleep-disordered breathing and insulin resistance in middle-aged and overweight men. *Am J Respir Crit Care Med* 2002;165(5):677-82.
54. Raldsson P. Clinical symptoms of sleep apnea and automobile accidents. *ORL Journal of Otorhinllaryngology* 1990;52:57-62.
55. Yee B, Campbell A, Beasley R, Neill A. Sleep disorders: a potential role in New Zealand motor vehicle accidents. *Internal Medicine Journal* 2002;32(7):297-304.
56. Teran-Santos J, Jimenez-Gomez A, Cordero-Guevara J. The association between sleep apnea and the risk of traffic accidents. Cooperative Group Burgos-Santander. *N Eng J Med* 1999;340(11):847-851.
57. Kapur K, Blough D, Sandbloom R, Hert R, de Maine J, Sullivan S, et al. The medical cost of undiagnosed sleep apnea. *Sleep* 1999;22(6):749-755.
58. Roland J, Delaive K, Roos L, Manfreda J, Bahammam A, Kryger M. Health-care utilization in the 10 years prior to diagnosis in obstructive sleep apnea syndrome patients. *Sleep* 1999;22(2):225-229.
59. Bahammam A, Delaive K, Ronald J, Manfreda J, Roos L, Kryger M. Health-care utilization in males with obstructive sleep apnea syndrome two years after diagnosis and treatment. *Sleep* 1999;22(6):740-747.
60. Chesson A, Ferber R, Fry J, Grigg-Damberger M, Hartse K, et al. Practice parameters for the indications for polysomnography and related procedures. Polysomnography Task Force, American Sleep Disorders Association Standards of Practice Committee. *Sleep* 1997;20(2):406-422.
61. The Report of an American Academy of Sleep Medicine Task Force. Sleep-related breathing disorders in adults: recommendations for syndrome definition and measurement techniques in clinical research. *Sleep* 1999;22(5):667-689.
62. Becker H, Jerrentrup A, Ploch T, Grote L, Penzel T, Sullivan C, et al. Effect of nasal continuous positive airway pressure treatment on blood pressure in patients with obstructive sleep apnea. *Circulation* 2003;107(1):68-73.
63. Peker Y, Hedner J, Norum J, Kraiczi H, Carlson J. Increased incidence of cardiovascular disease in middle-aged men with obstructive sleep apnea: a 7-year follow-up. *Am J Respir Crit Care Med* 2002;166(2):159-65.
64. Pepperell JC, Ramdassingh-Dow S, Crosthwaite N, Mullins R, Jenkinson C, Stradling JR, et al. Ambulatory blood pressure after therapeutic and subtherapeutic nasal continuous positive airway pressure for obstructive sleep apnoea: a randomised parallel trial. *The Lancet* 2002;359(9302):204-210.
65. Hui DS, To KW, Ko FW, Fok JP, Chan MC, Ngai JC, et al. Nasal CPAP reduces systemic blood pressure in patients with obstructive sleep apnoea and mild sleepiness. *Thorax* 2006;61(12):1083-1090 doi: 10.1136/thx.2006.064063.
66. Milleron O, Pilliere R, Foucher A, de Roquefeuil F, Aegerter P, Jondeau G, et al. Benefits of obstructive sleep apnoea treatment in coronary artery disease: a long-term follow-up

study. *Eur Heart J* 2004;25(9):728-34.
67. Phillips B. Sleep-disordered breathing and cardiovascular disease. *Sleep Med Rev* 2005;9(2):131-40.
68. Marin JM, Carrizo SJ, Vicente E, Agusti AG. Long-term cardiovascular outcomes in men with obstructive sleep apnoea-hypopnoea with or without treatment with continuous positive airway pressure: an observational study. *Lancet* 2005;365(9464):1046-53.
69. Kaneko Y, Floras JS, Usui K, Plante J, Tkacova R, Kubo T, et al. Cardiovascular effects of continuous positive airway pressure in patients with heart failure and obstructive sleep apnea. *N Engl J Med* 2003;348(13):1233-41.
70. Mansfield DR, Gollogly NC, Kaye DM, Richardson M, Bergin P, Naughton MT. Controlled trial of continuous positive airway pressure in obstructive sleep apnea and heart failure. *Am J Respir Crit Care Med* 2004;169(3):361-6.
71. Hassaballa HA, Tulaimat A, Herdegen JJ, Mokhlesi B. The effect of continuous positive airway pressure on glucose control in diabetic patients with severe obstructive sleep apnea. *Sleep and Breathing* 2005;9(4):176-180.
72. Meston N, Davies R, Mullins R, Jenkinson C, Wass J, Stradling J. Endocrine effects of nasal continuous positive airway pressure in male patients with obstructive sleep apnoea. *J Intern Med* 2003;254(5):447-454.
73. Harsch I, Schahin P, Radespiel-Troger M, et al. Continuous positive airway pressure treatment rapidly improved insulin sensitivity in patients with obstructive sleep apnea syndrome. *American Journal of Critical Care Medicine* 2004;169:156-162.
74. Babu A, Hedergen J, Fogelfeld L, Shott S, Mazzone T. Type 2 diabetes, glycemic control, and continuous positive airway pressure in obstructive sleep apnea. *Archives of Internal Medicine* 2005;165:447-452.
75. Habukawa M, Uchimura N, Nose I, Kotorii N, Yamamoto K, Matsuyama S, et al. Emotional states and quality of life in patients with obstructive sleep apnea. *Sleep and Biological Rhythms* 2005;3(3):99-105.
76. Sin DD, Mayers I, Man GC, Ghahary A, Pawluk L. Can continuous positive airway pressure therapy improve the general health status of patients with obstructive sleep apnea?: a clinical effectiveness study. *Chest* 2002;122(5):1679-85.
77. Parish JM, Lyng PJ. Quality of life in bed partners of patients with obstructive sleep apnea or hypopnea after treatment with continuous positive airway pressure. *Chest* 2003;124(3):942-7.
78. Verse T, Pirsig W. New developments in the therapy of obstructive sleep apnea. *Eur Arch Otorhinolaryngol* 2001;258(1):31-37.
79. Sullivan CE, Issa FG, Berthon-Jones M, Eves L. Reversal of obstructive sleep apnoea by continuous positive airway pressure applied through the nares. *Lancet* 1981;1(8225):862-5.
80. The Boston Consulting Group (Australia and New Zealand). Proposal for a national sleep health agenda. *Sydney BCG* June 2003.
81. Hudgel D. The role of upper airway anatomy and physiology in obstructive sleep apnea. *Clin Chest Med* 1992;13(3):383-398.
82. Ono T, Lowe A, Ferguson K, Pae E, Fleetham J. The effect of the tongue retaining device on awake genioglossus muscle activity in patients with obstructive sleep apnea. *Am J Orthod Dentofac Orthop* 1996;110(1):28-35.
83. Clark G, Sohn J, Hong C. Treating obstructive sleep apnea and snoring : assessment of an anterior mandibular positioning device. *J Am Dent Assoc* 2000;131(6):765-771.
84. American Sleep Disorders Association. Practice parameters for the treatment of obstructive sleep apnea in adults: The efficacy of surgical modifications of the upper airway. *Sleep* 1996;19(2):152-155.
85. Li KK. Surgical therapy for adult obstructive sleep apnea. *Sleep Medicine Reviews* 2005;9(3):201-209.

86. SBU The Swedish Council on Technology Assessment in Health Care. Obstructive Sleep Apnoea Syndrome: A Systematic Literature Review: Report of a Joint Nordic Project. Stockholm: SBU, 2007.
87. Hicklin LA, Tostevin P, Dasan S. Retrospective survey of long-term results and patient satisfaction with uvulopalatopharyngoplasty for snoring. *J Laryngol Otol* 2000;114(9):675-81.
88. Kezirian EJ, Goldberg AN. Hypopharyngeal surgery in obstructive sleep apnea: an evidence-based medicine review. *Arch Otolaryngol Head Neck Surg* 2006;132(2):206-13.
89. Sundaram S, Bridgman SA, Lim J, Lasserson TJ. Surgery for obstructive sleep apnoea. *Cochrane Database Syst Rev* 2005(4):CD001004.
90. White J, Cates C, Wright J. Continuous positive airways pressure for obstructive sleep apnoea (Cochrane Review). *The Cochrane Library*. Chichester, UK: John Wiley & Sons, Ltd, 2004.
91. Giles TL, Lasserson TJ, Smith BH, White J, Wright J, Cates CJ. Continuous positive airways pressure for obstructive sleep apnoea in adults. *Cochrane Database Syst Rev* 2006;3:CD001106.
92. Loredo JS, Ancoli-Israel S, Dimsdale JE. Effect of continuous positive airway pressure vs placebo continuous positive airway pressure on sleep quality in obstructive sleep apnea. *Chest* 1999;116(6):1545-9.
93. Malhotra A, Ayas NT, Epstein LJ. The art and science of continuous positive airway pressure therapy in obstructive sleep apnea. *Curr Opin Pulm Med* 2000;6(6):490-5.
94. Grote L, Hedner J, Grunstein R, Kraiczi H. Therapy with nCPAP: incomplete elimination of Sleep Related Breathing Disorder. *Eur Respir J* 2000;16(5):921-7.
95. Montserrat JM, Ferrer M, Hernandez L, Farre R, Vilagut G, Navajas D, et al. Effectiveness of CPAP treatment in daytime function in sleep apnea syndrome: a randomized controlled study with an optimized placebo. *Am J Respir Crit Care Med* 2001;164(4):608-13.
96. Stepnowsky CJ, Jr., Marler MR, Ancoli-Israel S. Determinants of nasal CPAP compliance. *Sleep Med* 2002;3(3):239-47.
97. Hsu AA, Lo C. Continuous positive airway pressure therapy in sleep apnoea. *Respirology* 2003;8(4):447-54.
98. Richards D, Bartlett D, Wong K, Malouff J, Grunstein R. Increased adherence to CPAP with a group cognitive behavioral treatment intervention: a randomized trial. *Sleep* 2007;30(5):635-640.
99. Milleron O, Pilliere R, Foucher A, De Roquefeuil F, Aegerter P, Jondeau G, et al. Benefits of obstructive sleep apnoea treatment in coronary artery disease: a long-term follow-up study. *Eur Heart J* 2004;25(9):728-734.
100. Mar J, Rueda J, Duran-Cantolla J, Schechter C, Chilcott J. The cost-effectiveness of nCPAP treatment in patients with moderate-to-severe obstructive sleep apnoea. *Eur Respir J* 2003;21(3):515-522.
101. Kushida CA, Littner MR, Hirshkowitz M, Morgenthaler TI, Alessi CA, Bailey D, et al. Practice parameters for the use of continuous and bilevel positive airway pressure devices to treat adult patients with sleep-related breathing disorders. *Sleep* 2006;29(3):375-80.
102. Scottish Intercollegiate Guidelines Network (SIGN). Management of obstructive sleep apnoea/hypopnoea syndrome in adults: a national clinical guideline. Edinburgh: Royal College of Physicians, 2003.
103. Lim J, Lasserson TJ, Fleetham J, Wright J. Oral appliances for obstructive sleep apnoea. *Cochrane Database Syst Rev* 2006(1):CD004435.
104. Banerjee A, Prichard A, Thomas R. Surgery of the upper airway. *Bailliere's Clinical Anaesthesiology* 1995;9(9):317-335.
105. Kao YH, Shnayder Y, Lee KC. The efficacy of anatomically based multilevel surgery for

obstructive sleep apnea. *Otolaryngol Head Neck Surg* 2003;129(4):327-35.
106. Miller FR, Watson D, Boseley M. The role of the Genial Bone Advancement Trephine system in conjunction with uvulopalatopharyngoplasty in the multilevel management of obstructive sleep apnea. *Otolaryngol Head Neck Surg* 2004;130(1):73-9.
107. Dattilo DJ, Drooger SA. Outcome assessment of patients undergoing maxillofacial procedures for the treatment of sleep apnea: comparison of subjective and objective results. *J Oral Maxillofac Surg* 2004;62(2):164-8.
108. Kim ST, Choi JH, Jeon HG, Cha HE, Kim DY, Chung YS. Polysomnographic effects of nasal surgery for snoring and obstructive sleep apnea. *Acta Otolaryngol* 2004;124(3):297-300.
109. Souter MA, Stevenson S, Sparks B, Drennan C. Upper airway surgery benefits patients with obstructive sleep apnoea who cannot tolerate nasal continuous positive airway pressure. *J Laryngol Otol* 2004;118(4):270-4.
110. Alajmi M, Mulgrew AT, Fox J, Davidson W, Schulzer M, Mak E, et al. Impact of Continuous Positive Airway Pressure Therapy on Blood Pressure in Patients with Obstructive Sleep Apnea Hypopnea: A Meta-analysis of Randomized Controlled Trials. *Lung* 2007;185(2):67-72.
111. Haentjens P, Van Meerhaeghe A, Moscariello A, De Weerdt S, Poppe K, Dupont A, et al. The impact of continuous positive airway pressure on blood pressure in patients with obstructive sleep apnea syndrome: evidence from a meta-analysis of placebo-controlled randomized trials. *Arch Intern Med* 2007;167(8):757-64.
112. Morgan SG, McMahon M, Mitton C, Roughead E, Kirk R, Kanavos P, et al. Centralized drug review processes in Australia, Canada, New Zealand, and the United kingdom. *Health Aff (Millwood)* 2006;25(2):337-47.
113. O'Malley SP. The Australian experiment: the use of Evidence Based Medicine for the reimbursement of surgical and diagnostic procedures (1998-2004). *Aust New Zealand Health Policy* 2006;3(3).
114. Medical Services Advisory Committee. Report of a review of the Medical Advisory Services Committee. Canberra: Australian Government Department of Health and Ageing, 2005.
115. Dillman D. *Mail and internet surveys: the tailored design method*. New York: John Wliey & Sons, Inc, 2000.
116. Powell N. Upper airway surgery does have a major role in the treatment of obstructive sleep apnea: "the tail end of the dog". *Journal of Clinical Sleep Medicine* 2005;1(3):236-240.
117. Rose G. *The Strategy of Preventive Medicine*. Oxford: Oxford University Press, 1994.
118. Kitto S, Villanueva EV, Chesters J, Petrovic A, Waxman BP, Smith JA. Surgeons' attitudes towards and usage of evidence-based medicine in surgical practice: a pilot study. *ANZ J Surg* 2007;77(4):231-6.
119. Miles A, Polychronis A, Grey JE. The evidence-based health care debate - 2006. Where are we now? *Journal of Evaluation in Clinical Practice* 2006;12(3):239-247 doi:10.1111/j.1365-2753.2006.00709.x.
120. Elshaug AG, Moss JR, Southcott A, Hiller JE. An analysis of the evidence-practice continuum: Is surgery for Obstructive Sleep Apnoea contraindicated? *Journal of Evaluation in Clinical Practice* 2007;13(1):3-9.
121. Linden L, Vondeling H, Packer C, Cook A. Does the National Institute for Health and Clinical Excellence only appraise new pharmaceuticals? *Int J Technol Assess Health Care* 2007;23(3):349-53.