

Engaging Stakeholders in Research: The Case of Pediatric Obstructive Sleep-Disordered Breathing and Adenotonsillectomy

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Introduction

Patient-centered outcomes research (PCOR) incorporates patient characteristics, preferences, and values to produce research outcomes that “people notice and care about.”¹ PCOR prioritizes stakeholder engagement, inclusive of patients and families, to inform the research question as well as elaboration of healthcare options and shared decision-making to create a partnership between patients/families and healthcare providers. As such PCOR has the potential for superior outcomes by helping “people communicate and make informed decisions, allowing their voices to be heard in assessing the value of healthcare options.”¹

The value of stakeholder-engaged research can be seen through PCOR². It alternately asks what outcomes are most important to all individuals it affects and engages the perspectives of these stakeholders in achieving the desired outcomes in research. In the case of pediatric obstructive Sleep-Disordered Breathing (oSDB), stakeholders include not only snoring children, but also parents, family members, pediatricians, surgeons, policy-makers, and health services and outcomes researchers. We rely heavily on stakeholder engagement in our research on decision-making for adenotonsillectomy surgery, which serves as a prime example of the value of PCOR³⁻⁶.

Pediatric oSDB affects up to 12% of children, and has various consequences on child health, including physical, cognitive, and emotional sequelae⁷⁻¹⁰. Tonsillectomy with adenoidectomy is the most common treatment for oSDB, and the second most commonly performed pediatric ambulatory surgical procedure in the United States, with over 500,000 cases performed per year^{9, 11}. The incidence has increased in past decades, and there is a risk of true morbidity with each procedure¹². There has further been much speculation in both research and the media regarding overuse/inappropriate use or unexplained variation in utilization for this common procedure¹³⁻¹⁶. In qualitative interviews of surgeons and pediatricians who treat pediatric oSDB, clinicians stated that while tonsillectomy is a good treatment option in many cases, there are surgeons who will perform the surgery indiscriminately (“I could care less if I perform an extra tonsillectomy, but I know some doctors who any kid who snores, they take their tonsils out”⁵).

Considering the significant potential benefits but also risks and wide variation of use in tonsillectomy, research into parent and clinician decision-making processes that precede this procedure are important to ensure higher quality, ev-

idence-based, informed decisions for surgery^{17, 18}. The inclusion of stakeholders in this research emphasizes the validity, relevance, and comprehensiveness of the questions asked and the investigations conducted. In this chapter, we present our initial experiences engaging stakeholders to inform research on pediatric oSDB and adenotonsillectomy.

Stakeholder-engaged Research Projects

Qualitative key informant interviews. We conducted interviews with 11 parents of children who had been evaluated or treated for oSDB⁶. We also conducted interviews with 10 physicians/ scientists who treat children with oSDB.⁵ The central themes of perspectives learned from these interviews have informed our current research and continue to assist in the development of future outcome measures and decision support tools. Interviews were conducted to learn parents' experiences regarding a) their children's experience of sleep-disordered breathing, b) their healthcare experience when seeking treatment, and c) factors which influenced their decision-making regarding adenotonsillectomy as a treatment option^{3, 6}. Parents were asked about the defining reasons they brought their children to otolaryngologists for evaluation, their knowledge and concerns about the risks and benefits of surgical care, their preferences for treatment, and their experiences with clinicians including interpersonal styles and communication.

We found that parents stressed the importance of clinician information-sharing about risks and benefits of treatment, and clinical factors such as symptom severity when making treatment decisions. Parents described their reasons for seeking treatment through an emotional lens, such as fear about their children's apnea ("that she wouldn't wake up, that scared me"). Parents elaborated on fear of anesthesia as often a more important consideration than actual surgical risks ("your little baby is being put to sleep. That was my biggest concern. The actual tonsillectomy I didn't care about"). They emphasized the importance of interpersonal factors and communication from clinicians on impacting a positive care experience as opposed to influencing their treatment decisions ("Everyone was very, very nice. It eased our fears and made us so comfortable").

10 physicians across disciplines (pediatrics, otolaryngology, pulmonary/sleep medicine, anesthesiology) and practice settings (academic, community, private) were interviewed. They had varying views of tonsillectomy use ("surgery to me is a last resort;" "I never hear anyone complain about tonsillectomy"). Physicians provided a variety of perspectives on treatment of SDB, which fell into three major themes: *approach to treatment and alternatives* ("I get a feel for the parents"); *communication and decision-making between physicians and families* ("I'm about explaining to parents and letting them decide"); and *sociocultural factors and barriers to care* ("some ask questions like 'is my child going to become sterile as a result of this?'"). Physicians had widely variable practice patterns and views regarding appropriateness of subspecialty referral and treatment.

The results of these interviews underscore the importance of communication and information-sharing when working with parents of children with oSDB to make treatment decisions. Further, they depict the real-life concerns parents have when considering treatment for their children which may be undermined in

a brief single surgical consultation. They also confirm a variation in approach to treatment. Stakeholders expanded on aspects of our research topic that we had not considered and are not captured in conventional quantitative analyses, and made us aware of thematic concerns in our consideration of parent and clinician experiences with oSDB treatment. Themes generated from these interviews have provided a base for many future projects that incorporate stakeholder perspectives into scientific investigation of pediatric oSDB and decision-making for adenotonsillectomy. Despite the rather small sample of parents and clinicians, we have been able to use perspectives from these interviews as a “jumping off point” when considering factors of oSDB treatment which are important to both parents of children with oSDB and the clinicians who treat them. They also serve as a foundation to develop practical applications including decision aids and outcome measures to assess parental knowledge after consultation.

The Obstructive Sleep-Disordered Breathing and Adenotonsillectomy Knowledge Scale for Parents. Increased knowledge of oSDB as well as adenotonsillectomy risks and benefits may alleviate the high decision conflict known to occur for parents considering adenotonsillectomy^{18,19}. In addition to engaging stakeholders through qualitative interviews, we have also engaged parents and clinicians in a research project to develop an outcome measure assessing parental knowledge of oSDB and AT surgery. This measure is available for use in both research and clinical practice^{4,20}.

Stakeholder engagement was conducted in multiple stages. In the early stages of parental knowledge scale development, an advisory board of clinicians who treat pediatric oSDB was composed to provide input on development of the initial prototype of the measure. Researchers synthesized a systematic literature review on oSDB and AT surgery with results from the key informant interviews described above to determine the most important elements in the assessment of parental knowledge. These elements were ultimately included in the prototype measure, which yielded 9 preliminary “categories” of knowledge about oSDB and AT, and 89 potential questions. A sample of 32 parents, clinicians, and other laymen were then engaged to test and evaluate the prototype measure in order to evaluate its validity for use in the field²⁰.

Combined parent-clinician stakeholder analysis indicated that the most important areas of parent knowledge about oSDB and AT, when making decisions for treatment of pediatric oSDB, were 1) symptoms of oSDB, 2) treatment options, 3) effects of anesthesia, 4) benefits of tonsillectomy, and 5) risks of tonsillectomy surgery. As a result of these stakeholder-defined themes, the oSDB and AT parent knowledge scale for parents was built around these elements. That is, based on stakeholder responses, the prototype measure was modified to yield a condensed scale containing 5 principle domains (oSDB symptoms, treatment options, experiences with anesthesia, risks of AT, benefits of AT), with 39 individual overall items.

The inclusion of stakeholders in developing the scale was essential. Clinicians provided researchers with insight and an editorial board that integrated important evidence-based facts into the measure. The inclusion of parents allowed researchers to modify and develop the scale in a manner that would be well-received

by their target population. Including stakeholders in developing this measure ensured that it was being appropriately evaluated by the populations who would use it, both those administering it (clinicians), and those taking it (parents).

Option Grid™ decision aid. In addition to developing a measure to assess knowledge, we are also in the process of developing a decision support tool to aid parents as they decide for AT surgery versus watchful waiting in the setting of oSDB. This tool will be based on the Option Grid™ framework, presenting a single-page table containing a series of frequently asked questions (FAQs) about treatment for pediatric oSDB, and providing comparative information about surgery compared to watchful waiting^{21, 22}. Stakeholders will be heavily involved in the development of this decision support tool throughout the process of its creation. Along with the Option Grid™ collaborative at Dartmouth Institute for Health Policy and Clinical Practice, an editorial board consisting of clinicians from multiple disciplines and parents of children previously diagnosed with oSDB will aid in developing this tool. Researchers will synthesize themes of aforementioned key informant interviews with primary outcomes from present literature. This synthesis will be used to develop an extensive list of potential FAQs for the Option Grid™ decision aid, which will be disseminated to a larger random sample of parents of children with oSDB. Parents of children with oSDB will give their perspectives on which FAQs are most relevant and relatable for inclusion in a decision aid about choice for AT surgery in the setting of pediatric oSDB.

A prototype decision aid will then be developed and subsequently administered to a new group of stakeholders for user testing: Parents of children with oSDB who are currently considering treatment will evaluate the decision aid, and provide responses which will aid in its modification. Thus, the three phases of stakeholder engagement for this project include: 1) the theme and FAQ development, 2) the creation of decision aid prototype, and 3) the user-testing of the Option Grid™ decision aid. By integrating stakeholders into not only the evaluation of this tool (stage 3), but also its development, we ensure that through every step of our scientific process, the tool is developed in a manner which will promote its use in modern research and practice.

Conclusions

The methods of the described projects inclusively demonstrate the importance of engaging stakeholders when developing research that has true implications for impacting health delivery systems and clinical practice. Engaging stakeholders in research helps to ensure that research outcomes will not only be of interest to stakeholders, but will contain material which has practical use in the clinical setting. We are optimistic that evolving research in our specialty will incorporate questions, study design, and outcomes which are both clinically relevant and also important to patients and families.

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