The impact of new evidence on regional variation in paediatric tonsillectomy and adenoidectomy: a historical review

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Abstract

Background. Tonsillectomy and adenoidectomy have been among the most commonly performed procedures in children for approximately 100 years. These procedures were the first for which unwarranted regional variation was discovered, in 1938. Indications for these procedures have become stricter over time, which might have reduced regional practice variation.

Methods. This paper presents a historical review on practice variation in paediatric tonsillectomy and adenoidectomy rates. Data on publication year, region, level of variation, methodology and outcomes were collected.

Results. Twenty-one articles on practice variation in paediatric tonsil surgery were included, with data from 12 different countries. Significant variation was found throughout the years, although a greater than 10-fold variation was observed only in the earliest publications.

Conclusion. No evidence has yet been found that better indications for tonsillectomy and adenoidectomy have reduced practice variation. International efforts are needed to reconsider why we are still unable to tackle this variation.

Introduction

Tonsillectomy and adenoidectomy have been among the most commonly performed procedures in children for approximately 100 years. These procedures were the first for which Glover discovered large practice variation, in 1938, that could not be explained by differences in patients’ needs. Hence, this variation was possibly driven by differences in physicians’ beliefs about the effectiveness of surgery and the lack of high-quality evidence on indications for surgery. Glover’s report is considered the foundation of research on practice variation. Therefore, the study of practice variation in small areas is still referred to as the Glover phenomenon.

Four decades later, John E Wennberg resumed the study of geographical practice variation, using the first population-based hospital discharge database in the USA. Wennberg is also the founder of the Dartmouth Atlas Project, which aims to provide data on practice variation for the general public. The results on medical practice variation raised inescapable questions about the effectiveness and quality of healthcare, and about treatment indications, as regional intervention rates did not necessarily correspond with disease incidences. One of the central concerns is over-utilisation of procedures, as it is thought that this results in higher costs without a clinical benefit per se for patients.

Evidence on the appropriate indications and effectiveness of these procedures improved over time, resulting in new international guidelines. It is presumed that these efforts have reduced the magnitude of regional practice variation for these procedures.

This study aimed to investigate whether better evidence and new guidelines have indeed reduced practice variation. We provide a historical perspective on practice variation in tonsillectomy and adenoidectomy among children. We also provide the history of professional consensus on indications for these procedures and on their effectiveness. Our hypothesis was that stricter indications for tonsil surgery over time resulted in reduced practice variation and thereby improved quality of care, as Wennberg predicted.

Materials and methods

We performed a narrative review of the literature to provide a historical account of practice variation in tonsil surgery. The search was performed by a trained librarian (JWS) on a broad scope of databases (Appendix 1). Additional information was obtained from books in the Walaeus Library (Library of Leiden University Medical Center). Data collection and the screening of eligible books and articles was performed by one author...
Changes in indications for tonsil surgery in children

The very first report of partial tonsil surgery dates from 700 BC, and was found in the Hindu Sanskrit document Atharva-Veda.\textsuperscript{13} Subsequently, the first adenoidectomy was described in 1867, after the invention of the laryngoscopy mirror.\textsuperscript{14} Over time, indications changed, but, until the twentieth century, adenotonsillectomy (tonsillectomy with or without adenoidectomy) was relatively uncommon. Furthermore, it had already been reported that an operation is rarely necessary as children generally outgrow their complaints.\textsuperscript{1}

Despite this knowledge, the numbers of adenotonsillectomies increased tremendously after the invention of anaesthesia and antisepctic techniques, and after the development of focal infection theory, as it was believed that local infections could cause systemic diseases in any part of the body.\textsuperscript{15–17} Consequently, an adenotonsillectomy was considered a cure for a variety of disorders early in the twentieth century, such as anorexia, rheumatism, nephritis, mental retardation and enuresis.\textsuperscript{18}

In response to the consequent increase in adenotonsillectomies, the first studies on the effectiveness of adenotonsillectomies were published.\textsuperscript{19} In 1963, the first randomised, clinical trial comparing adenotonsillectomy to watchful waiting in children was published, which concluded that surgical treatment was effective for recurrent pharyngitis and otitis media.\textsuperscript{20}

The focal infection theory was largely forgotten after the introduction of modern treatments, such as antibiotics.\textsuperscript{21} Consequently, by the late 1960s, tonsillectomy was predominantly performed to prevent acute tonsillitis, rather than to prevent secondary streptococcal infections throughout the body.\textsuperscript{22}

Obstructive sleep apnoea (OSA) was subsequently described as a new indication for adenotonsillectomy, as tonsil and adenoid hypertrophy is thought to be the most common cause of this condition.\textsuperscript{15,23,24} Although it is one of the most common indications for surgery now, the latest Cochrane Review found that the evidence on the effectiveness of adenotonsillectomy in children with OSA was only of moderate quality.\textsuperscript{25}

Several RCTs reported an overall benefit of adenoidectomy for the resolution of middle-ear effusion in children, with only small benefits to hearing and without any clear advantage in terms of treatment of acute otitis media.\textsuperscript{26} However, no evidence was found regarding the effectiveness of adenoidectomy for recurrent or chronic nasal symptoms in children.\textsuperscript{27} In 2011, a trial on this topic reported similar outcomes, and higher healthcare and societal costs, for adenoidectomy compared to watchful waiting.\textsuperscript{11,28,29} During an international consensus conference in 2017 on the management of otitis media with effusion (OME), it was advised that adenoidectomy only be performed in children aged over four years who suffer from OME with symptomatic adenoid hypertrophy.\textsuperscript{30}

In 1984, specific indications for tonsillectomy in children with severe tonsillitis were defined, known as the Paradise criteria, and international consensus was reached on this topic.\textsuperscript{7} According to these criteria, surgery is indicated for children aged 3–15 years who experienced at least seven episodes of tonsillitis during the previous year, or at least five episodes per year during the previous two years, or more than three episodes per year during the previous three years. Although those criteria are widely used, even for adults, the study had important limitations, such as the small number of subjects, crossovers between treatment arms of subjects and the lack of blinding.

At the beginning of the twenty-first century, two trials that included children who did not fulfil the severe tonsillitis criteria reported no major benefit and higher costs of performing an adenotonsillectomy compared to watchful waiting, which led to stricter indications for surgery.\textsuperscript{10,11,28,31} These recommendations were implemented in international guidelines (Table 1).\textsuperscript{28,32–35}

All current guidelines for tonsillectomy are still based on the Paradise criteria. Three out of five guidelines do not provide recommendations for adenoidectomy alone. In addition to the evidence on the lack of effectiveness for some children, it has been suggested that removal of the tonsils might increase long-term risks of later respiratory, allergic and infectious diseases in the adult.\textsuperscript{36}

Variation outcomes

In total, this review included 21 articles on practice variation in tonsil surgery rates.\textsuperscript{1,37–36} Fifteen articles focused on regional practice variation in tonsillectomy rates, with or without adenoidectomy. Five papers focused on regional practice variation in adenoidectomy rates alone. Seven articles adjusted rates for age and sex, and four articles stratified rates by age, sex or insurance status.

Variation in tonsillectomy rates in Europe

Nine articles studied practice variation in tonsillectomy rates in Europe, covering the years from 1938 to 2014.\textsuperscript{1,37–44}

As mentioned before, Glover was the first to report tremendous variations in age- and sex-specific tonsillectomy rates, between 1932 and 1938, among children living in similar neighbourhoods and having similar illnesses, which he explained in terms of variation in medical opinion (Table 2).\textsuperscript{1,37–44,57,58}

In 1958, even more striking variation was reported in the selection of children.\textsuperscript{39} While a first group of physicians selected 61 per cent of 1000 children for tonsillectomy, another group of physicians selected 94 per cent of these children after three re-examinations. ‘Snoring due to enlarged tonsils’ was mentioned in that publication, although interrupted breathing was not yet described.

After these publications, many studies examined European regional variation in tonsillectomy rates among children, and found a decrease in regional variation compared to the variation observed by Glover. For example, two-fold differences
between operation rates and referral patterns were found between two Scottish regions and the physicians within the same region, over the years 1961 to 1970, and again for the year 1990.39,40 This small difference between regions might be explained by the increasing attention to indications for tonsillectomy during that period.3,7

Unfortunately, in later years, large regional practice variation in surgical rates was again observed in European countries.41–44

Table 1. Indications for tonsillectomy and adenoidectomy by evidence-based guidelines

<table>
<thead>
<tr>
<th>Country</th>
<th>Tonsillectomy ± adenoidectomy</th>
<th>Adenoidectomy alone</th>
</tr>
</thead>
<tbody>
<tr>
<td>Denmark32</td>
<td>Tonsillectomy should only be offered to children under 15 years of age with recurrent tonsillitis, who, as a minimum, meet Paradise criteria. Tonsillectomy should be considered in children aged &lt;12 years with sleep-disordered breathing &amp; tonsillar hypertrophy</td>
<td>There is no guideline available on adenoidectomy alone</td>
</tr>
<tr>
<td>The Netherlands20</td>
<td>Tonsillectomy is indicated in children who meet Paradise criteria. Consider a tonsillectomy in children who have 4–6 episodes of recurrent tonsillitis per year. Tonsillectomy is indicated in children with OSA who do not suffer from co-morbidities</td>
<td>Do not perform an adenoidectomy in children with upper respiratory tract infections. Consider an adenoidectomy in children suffering from chronic nasal obstruction</td>
</tr>
<tr>
<td>Scotland33</td>
<td>Tonsillectomy should be considered for recurrent, disabling sore throat due to acute tonsillitis when episodes are well documented, adequately treated &amp; meet Paradise criteria</td>
<td>2019 guidelines do not provide specific guidance on adenoidectomy alone</td>
</tr>
<tr>
<td>UK34</td>
<td>Use shared decision-making strategies when children meet SIGN* criteria. If there is clear sleep apnoea, surgery should be discussed</td>
<td></td>
</tr>
<tr>
<td>USA35</td>
<td>Tonsillectomy is an option for children with recurrent throat infections that meet Paradise criteria for frequency, severity, treatment &amp; documentation of illness. Recommendation to counsel caregivers about tonsillectomy as a means to improve health in children with sleep-disordered breathing, co-morbid conditions or abnormal polysomnography</td>
<td>Adenoidectomy should not be performed unless a distinct indication exists (e.g. nasal obstruction, chronic adenoiditis). 2019 guidelines do not provide specific guidance on adenoidectomy alone. An earlier version stated that adenoidectomy alone is indicated in cases of recurrent suppurative or chronic OME, or in cases of chronic sinusitis, in children not responding to maximal medical therapy (e.g. appropriately chosen antibiotic, topical nasal steroid sprays, saline irrigations)</td>
</tr>
</tbody>
</table>

*According to the Scottish Intercollegiate Guidelines Network (‘SIGN’) criteria, sore throats should be due to tonsillitis, with five or more tonsillitis episodes per year, with the patient having had symptoms for at least a year, and the episodes should be disabling and prevent normal function. OSA = obstructive sleep apnoea; OME = otitis media with effusion

Table 2. Papers on practice variation for tonsillectomy in Europe

<table>
<thead>
<tr>
<th>First author</th>
<th>Year</th>
<th>Country</th>
<th>Level of variation</th>
<th>Stratification or adjustment</th>
<th>Results</th>
</tr>
</thead>
<tbody>
<tr>
<td>Glover1</td>
<td>1938</td>
<td>UK</td>
<td>Regions, hospitals</td>
<td>Unadjusted crude rates, &amp; stratified by age &amp; sex, in 4 regions</td>
<td>Highest vs lowest average rate among school children for 1932–1936 was 3.8 vs 0.3 per 1000 inhabitants; 8-fold variation in risk of death with surgery</td>
</tr>
<tr>
<td>Glover37</td>
<td>1948</td>
<td>UK</td>
<td>Regions</td>
<td>Unadjusted percentages</td>
<td>Highest vs lowest average percentages among children for 1936–1938 was 5.0% vs 0.1% of all school children</td>
</tr>
<tr>
<td>Bakwin38</td>
<td>1958</td>
<td>England</td>
<td>Physicians</td>
<td>Not applicable</td>
<td>Highest vs lowest percentage for children who underwent surgery per region in 1953 was 4.2% vs 0.4%. After 1st group of physicians selected 61.0% out of 1000 children for tonsillectomy, another group of physicians selected 94% after 3 re-examinations</td>
</tr>
<tr>
<td>Bloor39</td>
<td>1978</td>
<td>Scotland</td>
<td>Regions, physicians</td>
<td>Unadjusted crude rates</td>
<td>Differences between two socio-demographically similar Scottish regions for 1961–1970: operation rate was 10.4 per 1000 children for region 1 &amp; 13.0 per 1000 children for region 2. Intra-regional variation ranged from 6.2 to 15.8 operations per 1000 children</td>
</tr>
<tr>
<td>Bisel40</td>
<td>1994</td>
<td>Scotland</td>
<td>Regions</td>
<td>Unadjusted crude rates</td>
<td>Highest vs lowest rate in 1990 for children aged &lt;15 years was 8.0 vs 3.6 per 1000 inhabitants</td>
</tr>
<tr>
<td>Fedeli41</td>
<td>2009</td>
<td>Italy</td>
<td>Health units</td>
<td>Age- &amp; sex-adjusted</td>
<td>Highest vs lowest rate among children aged 2–9 years was 2.9 vs 12.5 per 1000 person-years</td>
</tr>
<tr>
<td>Suleman42</td>
<td>2010</td>
<td>England</td>
<td>Local authorities (NHS)</td>
<td>Unadjusted crude rates</td>
<td>Highest vs lowest rate (95% CI) for 2005–2010 among children aged &lt;15 years was 754 (690–822) vs 102 (83–125) per 100 000 inhabitants</td>
</tr>
<tr>
<td>UK NHS Atlas of Variation43</td>
<td>2012</td>
<td>England</td>
<td>Primary care trust</td>
<td>Age- &amp; sex-standardised</td>
<td>Tonsillectomy in children aged 0–17 years ranged from 83.1 to 500.4 per 100 000 population (6-fold variation). Exclusion of highest &amp; lowest rates gave 2.9-fold variation (range, 145.1–423.7)</td>
</tr>
<tr>
<td>Weeks44</td>
<td>2014</td>
<td>France (vs England, USA)</td>
<td>Countries, regions</td>
<td>Age- &amp; sex-standardised</td>
<td>Highest minus lowest rate for 2008, 2009 &amp; 2010 among children aged &lt;18 years was 3.6, 3.7 &amp; 3.1 respectively. SCV across years in France was 7.6. Lower variation in France compared to USA &amp; England</td>
</tr>
</tbody>
</table>

Systematic component of variation (‘SCV’): less than 3 indicates low variation, more than 5 indicates differences in practice styles or medical discretion; more than 10 indicates very high variation.37-44 NHS = National Health Service; CI = confidence interval.
**Variation in tonsillectomy rates in other countries**

This section concerns practice variation in tonsillectomy rates, with or without adenotonsillectomy, in other countries. In 1973, Wennberg was the first to resume the study on small-area variation in tonsillectomy rates outside of Europe, after the publication of Glover’s findings in 1938. These publications actually opened the era of practice variation research. Although large variation was observed, he did not specifically focus on tonsillectomy in children.

Six articles included in this review investigated paediatric tonsillectomy rates in the USA, Canada and Australia, published between 1977 and 2016. Practice variation in paediatric tonsillectomy in the USA was not researched until 2012 (Table 3). Despite new evidence and guidelines for children with milder disease by that time, high practice variation between regions in the USA was observed between 2012 and 2016, suggesting that variation in paediatric tonsillectomy care remains problematic. The latest paper from the USA focused on the variation in complication rates as well as regional variation in rates. Tonsillectomy rates were purported to relate to many factors, including social economic status, insurance status, levels of urbanisation and access to healthcare.

In contrast to the relatively low variation in Europe in the 1960s and 1970s, high variation was observed in all investigated regions in Canada at the time when the Paradise criteria were first published (Table 3), and when updated in 1996 and 2002. The 1996 publication was the first to mention OSA as an indication for surgery.

**Variation in adenoidectomy rates**

Less attention has been paid to regional variation in the rates of adenoidectomy without tonsillectomy (Table 4). In total, we included five papers from five different countries, published between 1977 and 2009.

The first paper on adenoidectomy rates was based on data from the USA. It was reported that adenoidectomy rates varied even more widely than tonsillectomy and adenotonsillectomy rates. In Australia, larger differences were detected between inner metropolitan, outer metropolitan and rural areas, from 1986 to 1990, for tonsillectomy combined with adenoidectomy compared to tonsillectomy alone. Between 2002 and 2006, more than three-fold differences were found in Italy and Norway as well, whereas in Finland the highest and lowest rates differed less than two-fold.

However, data on practice variation after the last RCT on the effectiveness of adenoidectomy in children with upper respiratory airway infections, published in 2011, are lacking. The limited data on practice variation in adenoidectomy rates hamper the formulation of definitive conclusions regarding the trial results and guidelines on practice variation.

**International variation in tonsil surgery rates**

Although Glover referred to other articles, he was the first to report on different international incidence rates, describing tonsillectomy rates as similar in the USA and the UK. Three articles included in this review investigated international variation for tonsillectomy and adenotonsillectomy. An international comparison of paediatric adenotonsillectomy rates reported contrasting results in 1998 (Table 5); rates ranged from 19 cases per 10 000 children in Canada to 118 cases...
per 10 000 children in Northern Ireland.54 International differences were also reported for paediatric adenoidectomy rates in 1998, which varied from 17 per 10 000 children in Canada to 129 per 10 000 children in Finland.55 The authors reported high rates in the Netherlands and Belgium as well, but found low rates of antibiotics for acute otitis media in those countries.

Finally, large variations in the numbers of adenoidectomies were also reported between and within Norway and Finland, from 1987 to 2005.56 However, the ‘correct rate’ remained unknown, as differences depend on other factors such as culture, refuge to alternative treatments like antibiotics, the type of healthcare system (easy access to care, high or low costs per person) and socio-economic status, which were not studied in most included articles.59–61

### Conclusion and future perspectives

This review aimed to provide a historical perspective on practice variation in paediatric tonsil surgery, alongside improved evidence on effectiveness and stricter indications for surgery. Glover, who was the first to publish on practice variation in paediatric adenotonsillectomy rates, would be staggered to know that large geographic variation for these procedures still exists.

The quality of evidence on the effectiveness of tonsil surgery in children has increased considerably during the last century. Guidelines have been developed with indications for surgery that are more evidence-based, and the Paradise criteria still apply after almost forty years.7,8,10,28,32–35 However, guidelines on adenoidectomy alone are scarce. This might explain the variation in surgical rates, warranting more research on this topic. Furthermore, data for the most recent years are lacking.

Despite new methods to gain insight into the causes of practice variation,38 determination of the optimal procedure rate remains problematic.59 This is because treatment decisions can be influenced by individual, social and healthcare factors.60,61 These reasons for variation, such as age and socio-economic differences between regions, were not always considered: the described studies evaluated and reported practice variation, but hardly any study was able to correlate explanatory factors with regional variation. Investigation of the explanatory factors might help physicians and decision-makers to better understand methods of tackling unwarranted variation. Future practice variation research should at least adjust for age differences between areas.

It is hard to differentiate between bacterial and viral tonsilitis, as both show similar symptoms. Furthermore, the current
guidelines lack objective standards to differentiate between bacterial and viral tonsillitis in primary care. Although more objective tests exist, such as the rapid streptococcal antigen test, cost-effectiveness studies are needed for routine implementation in primary care.\[62,63\] In addition, the Centor criteria, which differentiate between group A β-haemolytic streptococcus and non-group A β-haemolytic streptococcus as an underlying cause for tonsillitis, are subjective and are not applicable to the paediatric population.\[64\] Therefore, future research should also focus on diagnostics, such as biomarkers for diagnosis and disease severity, to better inform decision-making.

In conclusion, no evidence has yet been found to demonstrate that the new guidelines and better indications for surgery have reduced practice variation in tonsil surgery. International efforts are needed to reconsider why we are still unable to tackle practice variation in tonsil surgery.

**Competing interests**
None declared

**References**

Small area analysis

("Tonsillectomy/"mesh) OR "tonsillectomy/[tw] OR "tonsillectomies/[tw]
OR "tonsillectom.[tw] OR "tonsillectomy/[tw] OR "adenoidectomy/[tw] OR "adenoidectomy.
mes[ti] OR "adenoidectomy/[tw] OR "adenoidectomy/"mesh OR "adenoidectomy/[tw] OR "adeno.
tonsillectomy/[tw] OR "adenotonsillectomy/[tw] OR "adenotonsillectomy/[tw] OR "tongus.

Historical articles

("Tonsillectomy/mesh") OR "tonsillectomy/[tw] OR "tonsillectomies/[tw]
OR "tonsillectom.[tw] OR "tonsillectomy/[tw] OR "adenoidectomy/[tw] OR "adenoidectomy.
(suppl 2):S57

Appendix 1. Literature search
OR "17 years old"[ti] OR "18 years old"[ti]) AND ("0001/01/01"[PDAT] : "1950/12/31"[PDAT])

History of tonsil surgery

PubMed Central

IndexCAT
(("Tonsillectomy") OR Title:"Adenoidectomy") AND (Child OR children OR Infant OR infants OR Adolescent OR adolescents OR pediatric OR paediatric))

Web of Science (http://isiknowledge.com/wos)
(ts=("Tonsillectomy" OR "tonsillectomy" OR "tonsillectomies" OR "tonsillectomy" OR "Adenoidectomy" OR "adenoidectomy" OR "adenotonsillectomy" OR "adenotonsillectomy" OR "tonsil surgery" OR "tonsil surgery" OR tonsil*) AND ti=("History" OR historical*))