



Review Article

Tonsillectomy or tonsillotomy? A systematic review for paediatric sleep-disordered breathing

Lai-Ying Zhang^{a,*}, Laurie Zhong^b, Michael David^c, Anders Cervin^d^a The Prince Charles Hospital, Brisbane, QLD, Australia^b The Princess Alexandra Hospital, QLD, Australia^c School of Population Health, University of Queensland, Brisbane, QLD, Australia^d Faculty of Medicine and Biomedical Sciences, University of Queensland, Brisbane, QLD, Australia

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ABSTRACT

Background: Recent evidence has challenged the practice of tonsillectomy in children with sleep-disordered breathing. Tonsillotomy (subtotal/partial/intracapsular tonsillectomy) has been proposed as an alternative with equivalent effectiveness and decreased post-operative morbidity, thus improving cost-effectiveness.

Objective: To systematically review the literature comparing clinical efficacy, post-operative morbidity, and cost-effectiveness of tonsillotomy and tonsillectomy in paediatric (< 16yo) patients with sleep-disordered breathing.

Data sources: A systematic search of MEDLINE, EMBASE, and CENTRAL (1984–July 2014) was conducted. Papers in English directly comparing post-operative outcomes in tonsillectomy and tonsillotomy in children undergoing surgery for sleep-disordered breathing were included.

Review methods: Two authors independently assessed abstracts for relevance, with disagreements resolved by a third author. Selected studies were independently assessed regarding inclusion and exclusion criteria.

Results: Thirty-two studies satisfied inclusion and exclusion criteria (19 randomised, 13 non-randomised). Patient satisfaction, quality-of-life, and polysomnographic improvement post-surgery did not vary between tonsillotomy and tonsillectomy. Tonsillotomy reduced the odds of a secondary haemorrhage by 79% (OR 0.21, 95% CI 0.17–0.27, $p < 0.01$), decreased post-operative pain and reduced return to normal oral intake by 2.8 days (95% CI 1.08–4.52, $p < 0.01$). The odds of readmission were decreased by 62% (OR 0.38, 95% CI 0.23–0.60, $p < 0.01$). Tonsillotomy had a slightly higher rate of symptom recurrence (4.51%) than tonsillectomy (2.55%), the long-term impact of which was unclear.

Conclusion: Current evidence supports tonsillotomy in children with obstructive surgical indications. It is likely to reduce post-operative haemorrhage, pain, and facilitate a faster return to normal diet and activity. Healthcare burden is decreased due to fewer post-operative complications and reduced need for medical re-contact. More research is necessary to assess the risk of recurrence, and further classification of secondary haemorrhage severity is required to fully clarify the clinical benefit of tonsillotomy.

1. Introduction

Tonsillectomy (tonsillectomy) is currently the mainstay of surgical treatment for paediatric sleep-disordered breathing due to tonsillar hypertrophy. It involves the complete removal of the tonsils, including the tonsillar capsule, leaving the underlying pharyngeal muscles exposed to heal by secondary intention. In contrast, tonsillotomy (tonsillotomy, or partial/intracapsular tonsillectomy) avoids capsular disruption by only removing the obstructive tonsillar segment.

While tonsillectomy is the preferred procedure in recurrent tonsillar infection [1,2], studies have suggested that tonsillectomy and

tonsillotomy have similar effectiveness in treating isolated sleep-disordered breathing, both clinically and on polysomnographic testing [3–5]. There is evidence that tonsillotomy has less post-operative morbidity, as larger-calibre blood vessels near the capsule and pharyngeal muscles remain intact [6–8]. This in turn may decrease post-operative pain, minimising analgesia use and allowing patients a faster return to normal diet and activity. It may also decrease the risk of both primary and secondary haemorrhage, the latter of which may sometimes result in life-threatening hypovolemia necessitating a return to theatre. All of these factors raise the possibility that tonsillotomy may be more cost-effective than tonsillectomy, with lower rates of

* Corresponding author. The Prince Charles Hospital, Rode Road, Chermside, Brisbane, QLD 4032, Australia.
E-mail address: lai-ying.zhang@health.qld.gov.au (L.-Y. Zhang).

readmission and medical re-contact for complications; the potentially faster recovery times observed in tonsillotomy may also decrease the time caregivers are required to take off work.

However, there is concern that the tonsillar remnant in tonsillotomy may predispose to post-operative infection [7]. Tonsillar remnants may also re-hypertrophy, with possible recurrence of sleep-disordered breathing which may require a revision tonsillectomy if severe [9], thus exposing the patient to additional operative morbidity and increasing healthcare cost. It is therefore important to determine whether these risks outweigh the potential improved post-operative morbidity and cost-effectiveness offered by tonsillotomy.

Previous systematic reviews comparing the two techniques have either included non-target populations or excluded data from large non-randomised studies. Both Acevedo et al. [10] and Kim et al. [11] incorporated adult patient populations within their analyses, while Walton et al. [12] conducted a meta-analysis of randomised controlled trials only. Our study aims to systematically review the literature comparing clinical efficacy, post-operative morbidity, and cost-effectiveness of tonsillectomy and tonsillotomy, performed in the paediatric population for sleep-disordered breathing. Based on findings from both randomised trials and non-randomised real-world patient registers, this review hopes to make a recommendation on the best procedure for this patient population.

2. Material and methods

2.1. Study selection

MEDLINE, EMBASE, and CENTRAL were systematically searched (1948–July 2014) using the key words “tonsillectomy”, “adenotonsillectomy”, “tonsillotomy”, “intracapsular”, “subtotal”, “partial”, “subcapsular”, “supracapsular”, and “reduction” in combinations. Both randomised and non-randomised articles in English were included if they studied paediatric patients (< 16yo) without comorbidities (obesity, craniofacial abnormalities, etc.) undergoing tonsillar surgery of any technique for obstructive sleep-disordered breathing, analysing the desired outcome variables of post-operative haemorrhage, pain, infection rate, efficacy, and cost. Studies were excluded if more than 30% of the patient population underwent surgery for a non-obstructive indication. Only studies directly comparing tonsillectomy with tonsillotomy were included. Certain methods of tonsil reduction, in which holes were ablated in the tonsil, were not considered true tonsillotomy and these articles were excluded. Articles including concurrent adenoid surgery were not excluded. Article selection was performed independently by two authors, with differences resolved through a third author. After selection, the reference lists of included articles were manually checked for additional studies. If studies included non-paediatric patients, or patients undergoing surgery for non-obstructive indications, the authors were contacted for additional refinement of results/data. If authors were not contactable, the study was excluded.

2.2. Data extraction

Data was extracted independently by two authors. Primary outcomes included procedural effectiveness as determined by patient satisfaction, quality-of-life, and polysomnographic testing; primary and secondary post-operative haemorrhage rates; duration and severity of post-operative pain; symptom recurrence; and need for completion tonsillectomy. Secondary outcomes included post-operative infection rates and cost-related variables such as readmission rate, rate of medical re-contact, operative time, and time taken off work by caregivers.

2.3. Data analysis

All analyses were conducted using Cochrane Review Manager (RevMan 5.3) and Stata Version 15.0 (StataCorp, College Station, TX,

USA) statistical software. Studies were classified and combined in the analysis according to post-operative outcome and study design. We used the mean difference (MD) with a 95% confidence interval (95% CI) for continuous data, and for dichotomous data, the results were presented as an odds ratio (OR) with a 95% CI. To assess heterogeneity across the studies, we visually inspected forest plots and calculated both the Q (significance level of $P < 0.05$) and I^2 statistics [13]; with values of 40% or more for the latter indicating a substantial level of heterogeneity. Odds ratios and MDs were pooled using a Mantel-Haenszel fixed-effects model unless significant heterogeneity was evident; being the case, an inverse variance random effects model was used. An empirical continuity correction was conducted if dichotomous data had zero-events. Meta-influence analyses were conducted to assess the effect on pooled results by removing one study at a time. The type 1 error rate was set at 0.05 and all tests were two-sided. If a meta-analysis was unable to be performed due to data or procedural heterogeneity across trials, narrative comparison was conducted.

3. Results

In total, 453 articles were identified via search strategy from the three databases (Fig. 1). Thirty studies complied with criteria and were included in the final review. Sixty-six additional studies were identified in the reference lists of included studies, with one study meeting eligibility criteria. Another study, in-press, was identified by the leading author via the journal's mailing list. Three studies originally considered for inclusion were excluded as authors were not contactable. The characteristics of the 32 final included studies are detailed in Tables 1 and 2.

3.1. Nature of included studies

Of the 32 included studies, 19 were randomised controlled trials. Of these, two (Ericsson and Wadsby [14]; Ericsson and Graf [15]) were publications on data collected in a previous trial (Hultcrantz and Ericsson [16]). Hultcrantz et al. [17] was a long-term follow-up of patients already presented in Hultcrantz et al. [18]. In total, within randomised controlled trials, 752 patients received a tonsillotomy and 677 received a tonsillectomy. Microdebrider was the most common technique employed in tonsillotomy, while electrocautery and cold knife were the most popular for tonsillectomy. Approximately half of the studies had follow-up of 1 month or less post-operatively (10 studies), with the remaining nine studies having a follow-up period of at least 6 months. Only one study (Hultcrantz and Ericsson [16]) had a follow-up of more than two years.

The remaining 13 studies were non-randomised. Three of these were prospective studies. 10,204 patients underwent tonsillotomy and 7548 underwent tonsillectomy. Similar to the randomised trials, non-randomised studies predominantly employed microdebrider for tonsillotomy and electrocautery for tonsillectomy. Three studies did not specify tonsillectomy technique. Follow-up was predominantly longer than a year.

Mangiardi et al. [19], a non-randomised study, was included despite > 50% overweight or obese patients. This was decided upon discussion between the three study selection authors, since this was the only study utilising polysomnography.

3.2. Effectiveness

14 studies reported on effectiveness. Eight studies reported on patient or parent satisfaction, and five utilised quality-of-life surveys. Two studies compared the percentage of patients symptom-free on discharge. One study analysed objective symptomatic improvement via polysomnography.

The 10 studies addressing long-term effectiveness (> 1 month) did not report any significant difference in satisfaction or quality-of-life

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