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REVIEW ARTICLE

Role of subtotal tonsillectomy ('tonsillotomy') in children with sleep disordered breathing

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Abstract

Introduction: Sleep disordered breathing in children causes disturbance in behaviour and also in cardiorespiratory and neurocognitive function. Subtotal tonsillectomy ('tonsillotomy') has been performed to treat sleep disordered breathing, with outcomes comparable to established therapies such as total tonsillectomy or adenoidectomy. This review critically assesses the role of subtotal tonsillectomy in a paediatric setting.

Method: The Medline database (1966 to October 2012) was electronically searched using key terms including subtotal or intracapsular tonsillectomy, tonsillotomy, tonsillectomy, paediatrics, and sleep disordered breathing.

Results: Eighteen papers were identified and reviewed. Subtotal tonsillectomy would appear to have an efficacy equal to that of total tonsillectomy for the treatment of sleep disordered breathing, and has significant benefits in reducing post-operative pain and analgesia use. Subtotal tonsillectomy patients appear to have less frequent post-operative haemorrhage compared with total tonsillectomy patients.

Conclusion: In children, subtotal tonsillectomy is associated with fewer post-operative complications whilst having a comparable effect in improving sleep disordered breathing, compared with total tonsillectomy.

Key words: Tonsillectomy; Sleep Disordered Breathing; Sleep Apnea; Pediatrics

Background

Sleep disordered breathing

Sleep disordered breathing can vary greatly in severity, from partial obstruction of the upper airway tract, leading to snoring, through to 'significant upper airway obstruction resulting in oxygen desaturation and/or sleep fragmentation',[1] otherwise known as obstructive sleep apnoea (OSA). The prevalence of sleep disordered breathing is variable, with primary snoring affecting 8 to 12 per cent of children, whilst 1 to 3 per cent of pre-school and school-aged children have OSA syndrome.[2] The consequences of OSA are well documented and include physiological sequelae such as cardiorespiratory impairment and growth retardation.

Furthermore, sleep disordered breathing is associated with significant behavioural and neurocognitive problems.[3,4] Kohler *et al.*[4] reported that the magnitude of neurocognitive deficits did not relate to the severity of upper airway obstruction. They also found no neurocognitive improvements six months post-adenotonsillectomy, suggesting that early diagnosis and subsequent intervention may be required prior to the onset of neurocognitive problems. Recent research has found sleep disturbance even amongst primary snorers, which may be caused by relatively mild changes in oxygen saturation or by increases in respiratory arousals. In an observational study comparing 13 snoring versus 13 non-snoring children, test results for several domains of neurocognitive function differed significantly between the two groups, despite all subjects having an obstructive respiratory disturbance index within the normal range (i.e. 0.6 per hour). Results for some Intelligence Quotient (IQ) subgroups, including mean verbal IQ, mean global IQ, selective attention score, mean sustained attention score and mean memory index, were all significantly lower in the snorers than the non-snorers.[5] This implies that even mild sleep disordered breathing may have a significant impact on children, perhaps due to the chronicity of their sleep disruption.

Adenotonsillar hypertrophy is the main cause of sleep disordered breathing in children, and there is increasing evidence that adenotonsillectomy is effective in improving sleep, daytime behaviour and quality of life (QoL).[6,7] Adenotonsillectomy has been established as the definitive management of sleep disordered breathing, with significant improvement in sleep disturbance, physical symptoms, emotional symptoms and daytime functioning following the

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procedure.[7] Unfortunately, paediatric sleep problems are under-recognised: a recent Australian Position Paper[8] indicated that only 1 in 7 to 1 in 10 children who would benefit from an adenotonsillectomy currently receive the procedure.

Tonsillectomy

Tonsillectomy is one of the oldest and most commonly performed surgical procedures; however, there remains no standard technique amongst all surgeons.[9] Current indications for this procedure include recurrent tonsillitis and tonsillar hypertrophy resulting in sleep disordered breathing. An increasing proportion of tonsillectomies is conducted for the treatment of sleep disordered breathing, fuelled by the increasing number of paediatric patients being diagnosed with the condition, as well as realisation of the consequences of leaving the condition untreated.

During the last decade of the nineteenth century, in an era prior to anaesthesia, tonsillotomy (subtotal tonsillectomy) was regularly performed with a tonsillotome (tonsillar guillotine), which ensured that the procedure was as quick and painless as possible. This led to retention of remnant lymphoid tissue post-procedure, which in the pre-antibiotic era was thought to result in rheumatic fever and valvular heart disease. Full dissection tonsillectomy was first described in the first decade of the twentieth century, involving removal of the entire tonsil and its underlying capsule. Although techniques of total tonsillectomy were refined over the last century, tonsillotomy was completely abandoned for many decades prior to the 1990s.

Technique of subtotal tonsillectomy

Subtotal tonsillectomy techniques preserve the faucial arches and the mucosal plicae, whilst removing the proliferative lymphoid substance of the tonsil and crypt epithelium. This contrasts with the rather more destructive nature of total tonsillectomy: a recent histological review of 1220 tonsillectomy specimens found skeletal muscle in 89 per cent, seromucinous glands in 35 per cent and fat in 21 per cent, suggesting that normal surrounding tissues are regularly violated in traditional tonsillectomy.[10]

As a consequence, otolaryngologists have 'rediscovered' the more primitive subtotal reduction adenotonsillectomy (or 'tonsillotomy') procedure, in an effort to reduce the post-operative morbidity that is well reported in the literature. The use of this 'partial tonsillectomy' procedure in the context of treating patients with sleep disordered breathing was first described by Lantsov and Kovaleva[11] in the Russian medical literature. There have been many subsequent reports and analyses of subtotal tonsillectomy procedures as treatment for tonsillar hypertrophy related to sleep disordered breathing.[12,13] Two recent systematic reviews[14,15] reported that subtotal tonsillectomy has comparable efficacy in treating sleep disordered breathing whilst decreasing post-surgical complications (e.g. haemorrhage and pain), compared with total tonsillectomy.

Post-operative surgical outcomes

Resolution of obstructive sleep apnoea

Mitchell *et al.*[7] suggested that total tonsillectomy cured OSA in most cases, based on subjective findings. Tal *et al.*[16] confirmed this objectively by studying polysomnographic results in children with OSA, both before and after total tonsillectomy. In another study, the majority of children were 'cured' of sleep disordered breathing, based on polysomnographic measurements, six months after tonsillectomy.[17]

In a comparative study utilising polysomnography following subtotal and total tonsillectomy, de la Chaux *et al.*[18] found similar efficacy for both procedures. In their study, 20 children were followed up with repeated polysomnography within 12 months of the procedure. In the subtotal tonsillectomy group, the mean apnoea hypopnoea index ± standard deviation fell from 14.9 ± 8.7 events to 1.1 ± 1.6 events, suggesting that all children were effectively 'cured' of OSA by their procedure. This compares with reported cure rates of 28–100 per cent for total tonsillectomy.[19] Such a wide range is probably due to variation in how cure is defined. More than one episode of apnoea per hour is recognised as abnormal in healthy children; however, some research has utilised a range of between one and five episodes per hour.[1] In addition, the threshold beyond which the apnoea hypopnoea index becomes clinically significant is still unclear in the paediatric population.

The findings of de la Chaux *et al.*[18] are further supported by outcomes reported by Cantarella *et al.*[20] These latter authors randomly allocated 29 children with sleep disordered breathing to undergo either subtotal or total tonsillectomy, and compared pre- and post-operative polysomnographic results. A significant improvement in apnoea hypopnoea index was observed in both groups, indicating comparable efficacy of both procedures. Undoubtedly, more polysomnographic comparison data are needed, although the collection of such data is fraught with difficulty. Therefore, evidence thus far would suggest that subtotal and total tonsillectomy have equal efficacy in treating sleep disordered breathing in the paediatric population.[21]

Pain and analgesia consumption

Modern surgical techniques have made total adenotonsillectomy safer for children; despite this, the prevalence of post-operative complications (e.g. haemodynamic instability, prolonged pain and poor nutrition) in the first few post-operative days has remained relatively stagnant. Therefore, an appealing justification for subtotal tonsillectomy is its less invasive character, with probable smoother recovery during the post-operative period. This view is supported by the literature,[14,15,20,22–24]

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including two recent systematic reviews[14,15] which compared post-operative recovery for both procedures. The latter review,[15] of 15 randomised, controlled trials (RCTs) in paediatric populations with sleep disordered breathing, found significant reduction in post-operative pain and secondary haemorrhage prevalence following subtotal tonsillectomy in 9 trials. Twelve of the reviewed studies assessed post-operative oral intake; nine reported significantly earlier return to normal diet in the subtotal tonsillectomy group.

Evidence on subtotal tonsillectomy and post-operative pain reduction is conflicting. For example, Hultcrantz *et al.*[22] compared 41 children with tonsillar hypertrophy, of whom 21 received total tonsillectomy and 20 subtotal tonsillectomy; the subtotal tonsillectomy group was free from pain earlier (in 5 *vs* 8 days) and used half the analgesia of the total tonsillectomy group. In contrast, Chang *et al.*[23] conducted a prospective, randomised trial comparing subtotal versus total tonsillectomy using the coblation technique in 69 children; the mean pain score of the two groups did not differ on post-operative days 1 or 2, but on days 5 and 6 there was significant benefit ($p < 0.05$) in the subtotal group. Cantarella *et al.*[20] reported that significantly less analgesia was consumed by children undergoing subtotal versus total tonsillectomy ($p = 0.006$).

Post-operative haemorrhage

Reducing the prevalence of significant post-operative bleeding is of paramount importance. This is particularly so in infants, who have a much lower circulating blood volume and consequently a decreased ability to overcome haemodynamic compromise.

In a large, retrospective, observational study of more than 6000 patients who had undergone total tonsillectomy, Windfuhr and Chen[25] found a bleeding rate of 2.94 per cent. The vast majority of cases (78 per cent) comprised primary haemorrhage within 24 hours of the procedure. In a large, multi-centre case series of 870 children, Solares *et al.*[13] found that only 0.7 per cent of patients undergoing subtotal tonsillectomy suffered post-operative bleeding.

However, the greatest post-procedural morbidity is caused by secondary haemorrhage (i.e. haemorrhage occurring more than 24 hours post-procedure). In a large review of 6794 patients, Windfuhr and Chen[25] found 0.07 per cent of patients who had undergone total tonsillectomy required blood transfusion, with a 0.007 per cent mortality rate. A large meta-analysis reported a post-tonsillectomy haemorrhage rate of 4.5 per cent;[26] however, rates in individual studies vary considerably.[27] A recent, large, Austrian study[28] reported a post-tonsillectomy haemorrhage rate of 15 per cent; this is even higher than the 3.5 per cent bleeding rate reported by the 2007 National Prospective Tonsillectomy Audit[29] of English and Northern Irish hospitals, studying 34 000 patients. A number of reasons exist for the high bleeding rate reported by the Austrian authors. Firstly, this study utilised a particularly strict definition of tonsillectomy haemorrhage, having five different grades of bleeding. Grade A included 'anamnestic recorded blood tinged sputum', and was classed as a minor bleeding episode. In addition, Austrian patients remain in hospital for approximately three nights post-tonsillectomy, considerably longer than the typical Australian patient. This demonstrates that post-operative bleeding may be more common than reported in the literature, and perhaps even more common than reported by patients – one Scottish study[30] reported that, even when questioned in detail, up to 40 per cent of patients failed to report a minor degree of bleeding to their treating doctor.

Overall, it appears that the risk of both primary and secondary haemorrhage is reduced in subtotal tonsillectomy, compared with total tonsillectomy.[31] This is possibly due to lack of exposure of the tonsillar vasculature when the capsule remains intact.[21] In a retrospective series of 312 patients reported by Koltai *et al.*,[12] 6 patients receiving total tonsillectomy required hospital readmission, compared with only 1 subtotal tonsillectomy patient. Walton and colleagues' review[15] found no significant difference between the two groups regarding primary haemorrhage incidence; however, there were significantly fewer secondary haemorrhage episodes in the subtotal tonsillectomy group ($p = 0.04$). Acevedo and colleagues' systematic review[14] included 33 articles representing various levels of evidence (ranging from prospective, randomised studies to retrospective, non-randomised, non-blinded studies), and reported a 71 per cent reduction in post-operative haemorrhage in the subtotal tonsillectomy group; however, when they performed a sub-group analysis including only prospective, randomised, blinded studies, they found no significant difference. Thus, based on current evidence, subtotal tonsillectomy appears to be associated with less post-operative bleeding when compared with total tonsillectomy.

Quality of life

A recent study by Wood *et al.*[21] compared total tonsillectomy with subtotal tonsillectomy as regards Glasgow Children's Benefit Inventory scores. This QoL assessment tool measures improvements in many QoL domains, including confidence, self-esteem, ease of distractibility, concentration, liveliness, irritability, sleep patterns and behaviour, together with physical health factors such as days absent from school and medical presentations post-tonsillectomy. There was no difference between the total and subtotal tonsillectomy groups. Similar reports of equal QoL improvements in both groups have been published.[13,22,32,33] Stelter and colleagues' RCT[34] compared subtotal and total tonsillectomy, and found significant benefit for both procedures, for all Glasgow Children's Benefit Inventory domains.

Cantarella *et al.*[20] used the Obstructive Sleep Apnea 18 questionnaire to measure QoL following subtotal and total tonsillectomy. This questionnaire has proven

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reliability, and has been validated as a discriminative instrument using polysomnography.[35] Cantarella *et al.* found significant and statistically equal improvement in questionnaire scores in both groups, providing further evidence that subtotal and total tonsillectomy produce equal QoL benefits.

Post-surgical tonsillar hypertrophy

Total tonsillectomy, when performed adequately, includes removal of the entire volume of lymphoid tissue via dissection along the capsular plane; thus, regrowth of tonsillar tissue should not occur. Subtotal tonsillectomy does carry some risk of post-operative tonsillar hypertrophy[36] and there are historical reports of significant tonsillar regrowth.[37] This complication, in part, prompted the previous trend towards total tonsillectomy as the procedure of choice. However, more modern reports have shown variable disparity between post-operative tonsillar regrowth following total and subtotal tonsillectomy.

Solares *et al.*[13] found a tonsillar regrowth rate of 0.46 per cent over a follow-up period of 1.2 years after subtotal tonsillectomy, whereas Çelenk *et al.*[38] reported a much higher re-growth rate of 16.6 per cent within 1.5 years of surgery. This observed difference could be postulated to be due to the younger age of Solares and colleagues' patient population (mean age, 3.9 years), and the fact that these patients' tonsils had not yet reached their maximal volume.[39] In the only study of extended follow-up periods to date, Eviatar *et al.*[40] assessed 39 children 10–14 years after subtotal or total tonsillectomy. They reported a re-operation rate of 5 per cent after subtotal tonsillectomy compared with 0 per cent after total tonsillectomy, although the reasons for these second procedures were unclear. Recently, Zagolski[41] analysed 793 patients and found a relationship between the number of respiratory tract infections and the rate of post-operative tonsillar hypertrophy. This finding supports the suggestion by Kohler *et al.*[4] that tonsillotomy should be performed as soon as possible after diagnosing tonsillar hyperplasia. A diet high in sugar also appeared to be a risk factor. Further studies are required to evaluate long-term tonsillar regrowth following subtotal tonsillectomy; however, it is unlikely that subtotal tonsillectomy will prove to be as efficacious as total tonsillectomy in preventing tonsillar regrowth. The risk of tonsil regrowth should be clearly discussed with patients or their parents prior to proceeding with subtotal tonsillectomy.

Conclusion

Sleep disordered breathing in children has been shown many times to be associated with physical and physiological disturbances if left untreated. Total tonsillectomy, usually with adenoidectomy, has for many years been the treatment of choice for the management of sleep disordered breathing. Subtotal tonsillectomy had fallen out of use because of concerns over its efficacy, and complications reported during the pre-antibiotic era. However, published data now demonstrate subtotal tonsillectomy to be of equal efficacy with total tonsillectomy in the treatment of sleep disordered breathing. The literature also demonstrates improved QoL after subtotal tonsillectomy, compared with traditional total tonsillectomy, together with significant reduction in post-operative pain and analgesia use. Furthermore, the prevalence of post-operative haemorrhage is lower in subtotal tonsillectomy patients than in total tonsillectomy patients.

There is evidence to suggest that subtotal tonsillectomy is associated with a degree of post-operative tonsillar hypertrophy, making some otolaryngologists reluctant to perform a subtotal procedure. However, there is currently extremely limited data on what predisposes patients to tonsillar regrowth following subtotal tonsillectomy. There is a need for more data in this area, which could be used to more accurately identify patients who may be unsuitable for subtotal tonsillectomy and thus may require the more traditional procedure.

There is a paucity of studies using objective data to compare the pre- and post-operative OSA status of children receiving subtotal versus total tonsillectomy. However, some objective data, and an increasingly solid bank of subjective evidence, suggest that the subtotal procedure does not produce inferior outcomes regarding sleep disordered breathing, compared with total tonsillectomy. When this is combined with the clearly documented reduction in both post-operative pain and haemorrhage, it is appropriate to recommend that subtotal tonsillectomy should be offered to parents of children with sleep disordered breathing.

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