Lingual tonsils hypertrophy; a cause of obstructive sleep apnea in children after adenotonsillectomy: Operative problems and management

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ABSTRACT

Objective: Although adenotonsillar hypertrophy has been reported to be the commonest cause of pediatric obstructive sleep apnea (OSA), enlargement of the lingual tonsils is increasingly being recognized as a cause, even after adenotonsillectomy. The aim of our study was to elucidate the lingual tonsils hypertrophy as a cause of pediatric OSA and also to evaluate the efficacy of lingual tonsillectomy in relieving symptoms of the disease considering the peri-operative problems and management.
Methods: Sixteen children with lingual tonsils hypertrophy after adenotonsillectomy were included in the study. Computerized tomography (CT) and/or magnetic resonance imaging (MRI) were used for detection of the lesions. They underwent lingual tonsillectomy with special anesthetic care, flexible laryngoscopy and polysomnography were done pre- and post-operatively. Follow up of the patients was carried out for at least 1 year.
Results: Three cases developed post-operative airway obstruction that is caused by tongue base edema. Complete improvement of snoring and apnea was achieved in 10 cases. Despite complete ablation of lingual tonsils, persistent snoring was detected in six cases, while apnea was detected in two cases. Down's syndrome, mucopolysaccharidoses, and obesity may be underlying factors for persistent symptoms.
Conclusions: Lingual tonsils hypertrophy could be the cause of obstructive sleep apnea in children after adenotonsillectomy, lingual tonsillectomy is an effective treatment for these cases, however perioperative airway problems should be expected and can be managed safely. Persistent symptoms after lingual tonsillectomy may be due to the presence of co-morbidities such as cranio-facial deformities, obesity, and/or mucopolysaccharidoses.

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1. INTRODUCTION

Obstructive sleep apnea (OSA) is a common problem in pediatric patients; it affects 1–10% of all children, while snoring is estimated to be present in 3–12% [1]. The pathologies responsible for these disorders in children are more diverse than those in adults. Although most affected adults and older children experience obstruction at the pharyngeal level, younger individuals may be affected at a variety of sites in the upper respiratory tract. In small children, the distance between these sites may be quite small; resulting in stertor and/or stridor, and the source of the noisy breathing may be difficult to localize [2]. Although adenotonsillar hypertrophy has been reported to be the commonest cause of pediatric OSA, enlargement of the lingual tonsils is increasingly being recognized as a cause, particularly in children with craniofacial anomalies even after adenotonsillectomy [3–5].

Lingual tonsils hypertrophy (LTH) was first described by Vesalius in 1543; however its true incidence is still underestimated. This fact may be explained by the position of the lingual tonsils, since they are difficult to visualize on routine physical examination. Improved visualization facilities of the upper airway made possible detection of the disease in this region [6–8]. Lingual tonsils are components of the Waldeyer's ring of lymphoid tissues, they are situated at the base of the tongue, and their hypertrophy may cause OSA. It has been reported in previous studies as a cause of unexpected difficult mask ventilation and/or tracheal intubation, this makes the most important concern in the anesthetic management of the children with OSA due to LTH is the preservation of the upper airway patency specially when there is associated cranio-facial anomalies [5–11]. Also, trauma to this area caused by surgery may results into tongue base edema leading to post-operative airway obstruction that needs special care duringrecovery [3,9,10].

The aim of our study was to elucidate the lingual tonsils hypertrophy as a cause of pediatric OSA and also to evaluate the efficacy of lingual tonsillectomy in relieving symptoms of the disease considering the peri-operative problems and management.

2. METHODS

This case series study enrolled 16 children who presented with OSA after adenotonsillectomy, the cases were admitted and operated upon in the Department of Otolaryngology of Cairo University through the period from February 2006 to January 2010. Eleven males and 5 females, their ages ranged between 5 and 16 years with a mean of 8 years and 3 months. The study was approved by the institutional review board and the principles outlined in the Declaration of Helsinki were followed.

The following inclusion criteria were adopted:

- Diagnosis of lingual tonsils hypertrophy as the only cause of obstructive breathing seen by radiological (LTH appears as a soft tissue mass at the base of the tongue impeding the epiglottis and obstructing the hypopharynx) and/or endoscopic examination (the lesion appears as a corrugated soft tissue mass anterosuperior to the epiglottis).
- No age group was excluded.
- No past history of lingual tonsillectomy.

- Cases with recurrent adenoidal hypertrophy and/or redundant soft palate were excluded.

All cases were subjected to the following:

-Pre-operative assessment:

History obtained from the parents or the guardians about the symptoms suggestive of OSA including the nocturnal symptoms (snoring with difficult breathing, observed apnea and restless sleep) and daytime symptoms (daytime somnolence) [11].

Physical examination was done for assessment of associated cranio-facial abnormalities or obesity. Radiological evaluation in the form of computerized tomography (CT) and/or magnetic resonance imaging (MRI) on the head and neck region for detection of the site of obstruction. Flexible laryngoscopy was used for detection of the cause and site of obstruction. Cases were assessed by overnight polysomnography (PSG) for at least 6 h in a quiet, dark room. The numbers of apneas,
hypopneas, apnea/hypopnea index and minimum O2 saturation were measured.

The severity of OSA based on polysomnography was measured according to the guideline of Schwengel et al. [12]. Routine laboratory investigations in the form of complete blood picture, coagulation profile and blood grouping were done.

- Operative procedure:

Fifteen minutes before induction of general anesthesia, all patients were pre-medicated with 0.3 mg/kg of oral midazolam, then the nasal cavity was prepared with 0.05% xylometazoline drops and 2% lidocaine gel. General anesthesia was induced by inhalation of 8% sevoflurane in 100% oxygen, as consciousness of the patient was lost; Jew thrust was used to maintain the patency of the airway during spontaneous breathing. At that time, an intravenous (IV) line was established; atropine 0.01 mg/kg, dexamethasone 0.1 mg/kg and ondanseteron 0.1 mg/kg were given. When an adequate depth of anesthesia was attained, the glottic opening was evaluated using Millar straight blade, with visualization of the glottic opening; the trachea was intubated with age-appropriate nasal RAE endotracheal tube. If the glottic opening could not be visualized despite proper sized laryngoscopic blade and application of external laryngeal manipulation, asleep fiberoptic nasal intubation was the choice. If the patient can neither be ventilated nor intubated after inhalational induction of general anesthesia, laryngeal mask airway (LMA) was used to maintain ventilation, if failed, cricothyroidotomy was the choice. After endotracheal intubation, Atracurium (muscle relaxant) is used for controlled mechanical ventilation and all patients received paracetamol (perfalgan) IV infusion 15 ml/kg over 15 min to induce post-operative analgesia. A Jennings mouth gag is introduced to maintain the mouth open. The tongue tip is grasped with a babcock round blunt tip forceps, and then the tongue is pulled anteriorly by the left hand while a unipolar diathermy probe hold by the right hand is used to excise the lingual tonsils. Completion of surgical procedure is considered when all lymphoid tissues at the tongue base are completely removed. Sometimes, retraction of soft palate is needed using two rubber catheters passing through the nostrils for better exposure of tongue base, bleeding was controlled with coagulator diathermy. At the end of surgery, sevoflurane was discontinued and the residual neuromuscular

blockade was reversed with neostigmine (0.05 mg/kg IV) and atropine (0.01 mg/kg IV). When the patient is awake, the trachea is extubated and the patient is placed in the lateral position, and then transferred to the post-anesthesia care unit (PACU) with observation of respiration and oxygen saturation.

- Post-operative care and follow up:

All cases received oral antibiotics and paracetamol for 1 week, and oral steroids for 3 days, the patients were discharged from hospital in the 3rd post-operative day except cases that developed post-operative airway compromise, they stayed on monitored
hospital bed till the risk is eliminated. Children were seen at the end of the 1st and 2nd post-operative weeks, with follow-up appointments at 1, 3, 6, and 12 months. By the end of the follow up period, parent's questionnaire was used for assessment of subjective improvement of obstructive symptoms, flexible laryngoscopy was also performed for all cases for detection of any recurrence, and polysomnography was used with recording of the same pre-operative parameters.

3. RESULTS

Sixteen children with OSA caused by lingual tonsils hypertrophy were enrolled in this study, the parents/guardians of the patients stated that the children have complained of snoring and obstructive sleep breathing 5 months to 3 years (with a mean of 1 year and 2 months) after adenotonsillectomy. Pre-operative examination of the patients showed no abnormalities except in five cases; one case with Down's syndrome, two cases with mucopolysaccharidoses (MPS) and two cases with obesity.

CT was performed for 10 cases and MRI was performed for 3 cases while both were done for 3 cases (Figs. 1 and 2).

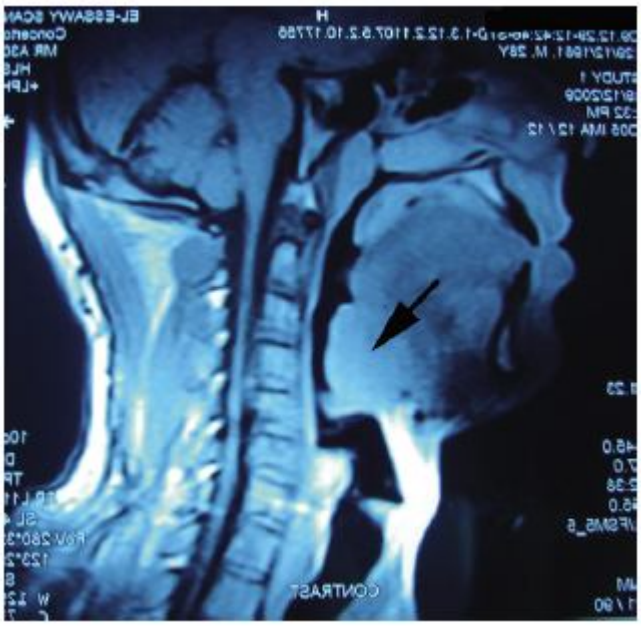

Fig. 1. MRI sagittal section, the arrow points to hypertrophied lingual tonsils that
obstructing the airway.

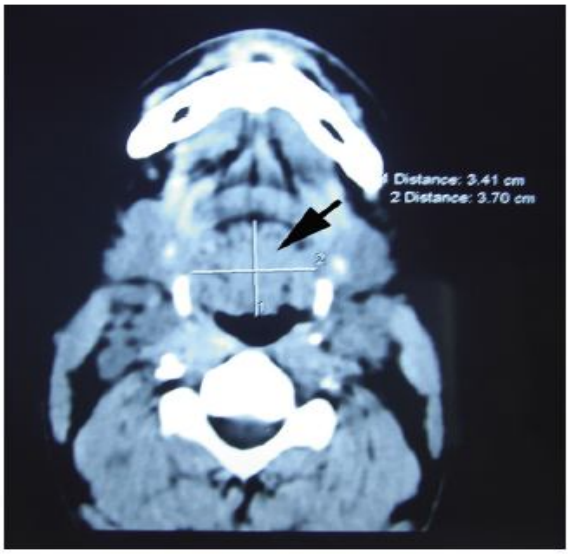

Fig. 2. CT axial section, the arrow points to hypertrophied lingual tonsils.

Lingual tonsil hypertrophy appeared to be the cause of airway obstruction in all cases, in addition the case of Down's syndrome showed narrow and shallow nasopharynx and the two patients of MPS showed narrow nasopharyngeal space with thickened postero-superior and lateral walls.
Flexible laryngoscopy showed LTH in all cases (Fig. 3), with relatively narrow nasopharynx in five cases; one case with Down's syndrome, two cases with MPS and two cases with obesity.

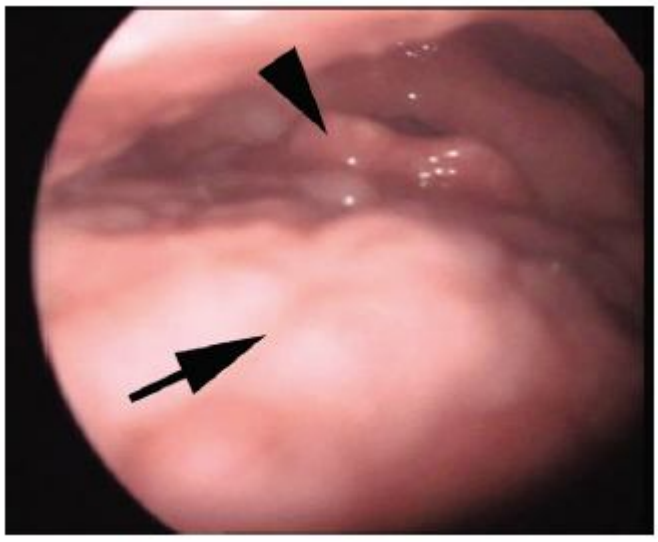

Fig. 3. The airway seen by flexible laryngoscopy, the arrow points to hypertrophied
lingual tonsils and the arrow head points to the epiglottis.

Pre-operative polysomnographic data showed that the mean apnea attacks was 13.8, while it was 42.2 for hypopnea, 10.5 for apnea/hypopnea index, with minimum O2 saturation of 84%. Despite laryngeal inlet was partially seen, 11 children (68.8%) were intubated successfully after 1 or 2 trials. In five children, the glottic opening could not be visualized despite proper sized laryngoscopic blade and application of external laryngeal manipulation, as lingual tonsils displaced the epiglottis posteriorly
against the laryngeal inlet so that obscuring the glottis, so asleep fiberoptic nasal intubation were used, it was successful in four cases. Only one child could not be intubated as the upper airway was obstructed immediately after loss of consciousness and ventilation via face mask was very difficult even with insertion of nasopharyngeal airway, so LMA was used and the child was subjected to awake fiberoptic naso-tracheal intubation later. No cases needed cricothyroidotomy.
Intra-operative bleeding was not a problem, and all cases were controlled successfully using diathermy cauterization. Three cases developed post-operative airway obstruction that is caused by edema of the tongue base; they showed oxygen desaturation to less than 90%. Nasopharyngeal airway was inserted with oxygen supplementation via face mask and hydrocortisone was given (3 mg/kg), oxygen saturation was improved and reintubation was not required. Difficult breathing was eliminated after 24 h; however the three cases were discharged from hospital
in the 4th post-operative day. No cases developed post-operative hemorrhage or infection.
By the end of the follow up period, parent's questionnaire revealed persistence of snoring in six cases (37.5%), while flexible laryngoscopy showed complete ablation of lingual tonsils. Polysomnography showed persistence of apnea in only two cases
(12.5%); however comparison between the pre-operative and postoperative polysomnographic data of the six children who demonstrated persistent snoring showed relative improvement of apnea. Associated co-morbidity in the form of Down's syndrome (1 case), mucopolysaccharidoses (2 cases), and obesity (2 cases)
could be the cause of persistent symptoms (Table 1).

Table 1: Pre-operative and post-operative apnea severity in children who demonstrated persistent postoperative snoring.

Postoperative snorer patients	Age (years)	PSG pre-operative	PSG post-operative	Body weight	Associated craniofacial anomalies

1st patient	7	Mod OSA	Mild OSA	Over-weight	Down's
2nd patient	5	Mod OSA	Mild OSA	Normal	syndrome
3rd patient	5	Mod OSA	No apnea	Normal	MPS
4th patient	8	Mod OSA	No apnea	Obese	MPS
5th patient	13	Mod OSA	No apnea	Obese	No
6th patient	16	MildOSA	No apnea	Normal	No
					No

4. DISCUSSION

Tonsillectomy and adenoidectomy are considered the first line of surgical treatment of pediatric patients with OSA. Persistence of the condition after adenotonsillectomy may be attributed to preexisting narrow pharyngeal airspace, cranio-facial deformities, or primary or secondary LTH, the later has been reported in several
studies as a common cause [6,7,13,14]. Unfortunately, LTH may not be detected clinically on routine preoperative physical examination [15], so that the extent of the obstruction of the glottic opening caused by the LTH might not be evaluated unless a fiberoptic laryngoscopic examination is performed or directly visualized during endotracheal intubation. In our study, we presented 16 children suffering from persistent OSA after adenotonsillectomy proved by preoperative PSG, the cause of the obstructive breathing was LTH as diagnosed by preoperative fiberoptic laryngoscopy. They underwent lingual tonsillectomy under general anesthesia that was induced using sevoflurane as its effect can be easily reversed if difficulties in the airway occurred; also the use of sevoflurane has the advantage of fast induction and rapid emergence [16]. Neuromuscular blocking agents were avoided during induction of general anesthesia to preserve the skeletal muscle support of the upper airway, as it has been reported that the loss of upper airway skeletal muscle support in some cases of LTH may be fatal. In these situations, the lingual tonsils may act as a ball-valve effect obstructing the glottic opening and interfere with oxygenation [17].

In 11 cases, the glottic opening could be visualized using the Millar straight blade laryngoscope after applying optimal external laryngeal manipulation. The Millar straight blade laryngoscope was used as it is designed so that its tip passes posterior to the epiglottis to elevate it directly, providing better visualization of the glottic opening; this is unlike the curved (Macintosh) blade in which the blade is just reaching the vallecula and this may carry the risk of traumatic injury to the lingual tonsils, with bleeding in the upper airway [18]. In five cases, the glottic opening could not be visualized due to excessive tonsillar tissues, asleep nasal fiberoptic intubation was used; the view was gained in four of them, helped by mandibular elevation with manual protrusion of the tongue by an assistant, and also avoidance of muscle relaxant was helpful as muscle relaxation may cause more posterior displacement of the tongue and epiglottis rendering the intubation more difficult [19]. It should be noted that the difficulty of tracheal intubation by conventional laryngoscope may also be attributed to the presence of co-morbidities such as Down's syndrome (one case), MPS (one case), obesity (two cases) and overweight

(one case), despite the latter has been considered as weak predictor for difficult intubation [20–23].

In some circumstances, LTH may be the cause of inability of intubation and ventilation. It was the situation in one of the two obese patients, even with application of nasopharyngeal airway. LMA was successfully inserted and proper ventilation was
resumed, then sevoflurane discontinued and the patient was awakened and the child was subjected to awake fiberoptic nasotracheal intubation.

However, the use of LMA in patients with LTH is controversial, some published case reports revealed that its use was successful in managing the upper airway obstruction in patients with LTH [19,24], while some authors reported that LMA may cause edema and bleeding of the airway specially with repeated insertion attempts between several intubation trials as the blind insertion of LMA may be traumatic to the lingual tissues [19,21,25]. The upper airway edema and bleeding render the fiberoptic intubation more difficult, so in our study, we limited the use of LMA and considering it as a last option in cases of cannot intubate cannot ventilate via the face mask.

Different techniques for lingual tonsillectomy have been performed, sharp dissection was used by Dundar et al. [7], while Conacher et al. [17] used laser; they reported that hemorrhage and post-operative edema were less frequent.

Recently, Lin and Koltai [26] used endoscopic-assisted coblation lingual tonsillectomy for treatment of cases with LTH; although the method was effective in
relieving symptoms with no reported post-operative edema, 2 out of 26 cases developed adhesions between tongue base and epiglottis, however this complication did not affect feeding or breathing. In our study, we performed lingual tonsillectomy using unipolar diathermy, 3 out of 16 cases developed post-operative edema. However, some precautions were taken intra-operatively to decrease the occurrence of this problem; releasing the tongue traction every 15 min, shortening the operative duration time, regular inspection of tongue base and systemic steroids.

Despite complete ablation of the lingual tonsils as seen by flexible laryngoscopy, parent's questionnaire revealed persistence of snoring in six cases. The cause of persistent snoring may be explained by the associated co-morbidities of those patients,
Down's syndrome, mucopolysaccharidosis, and obesity. Polysomnography (PSG) showed marked improvement of apnea, hypopnea, apnea/hypopnea index and minimum oxygen saturation from 16.7, 42.2, 10.5, and 84% to 1.9, 25.8, 3.2, and 91%, respectively. However, post-operative PSG showed persistent apnea in two
cases that demonstrated mild apnea after they had moderate apnea pre-operatively; one of them has Down's syndrome and the other has MPS. Many authors commented on factors that may contribute to treatment failure of OSA, such factors may include
craniofacial disproportion, hypotonia, and oropharyngeal soft tissue redundancy typically associated with obesity and mucopolysaccharidoses [3,12,17,25–27].

In conclusion, lingual tonsils hypertrophy could be the cause of obstructive sleep apnea in children after adenotonsillectomy, lingual tonsillectomy is an effective treatment for these cases, however peri-operative airway problems should be expected and can be managed safely. Persistent symptoms after lingual tonsillectomy may be due to the presence of co-morbidities such as craniofacial deformities, obesity, and/or mucopolysaccharidoses.

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