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Original article

Surgical ablation of lingual tonsils in the treatment of obstructive sleep apnea

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ABSTRACT

Objectives/Hypothesis: The retrolingual space is one of the potential sites of obstruction identified in patients with obstructive sleep apnea syndrome (OSAS). Hypertrophied lingual tonsils (LT) can obstruct the airway at this level. The goal of this study was to measure the tolerance and efficacy of lingual tonsillectomy in patients with OSAS.
Study design: A retrospective chart review was conducted recruiting all patients with OSAS confirmed on sleep recording, who either had failed or refused medical treatment and who underwent lingual tonsillectomy.
Materials and methods: Diagnosis of LT hypertrophy was made by full ENT clinical examination using a flexible endoscopy, completed by MRI and followed by drug-induced sleep endoscopy. The surgical intervention was carried out endoscopically by diode laser or coblation. The primary endpoint to measure efficacy was drop in apnea-hypopnea index (AHI) on sleep recording at 6 months. Secondary endpoints comprised reduced snoring and Epworth Sleepiness Scale (ESS) and postoperative symptom tolerance.
Results: Eleven patients aged 44.3 ± 12.6 years were included. AHI dropped from 29.5 ± 21.7/h to 11.6 ± 9.6/h: i.e., by 60% ($P = 0.005$). Five patients had AHI < 10/h: i.e., cure rate of 45%. ESS dropped from 13 ± 3.4 to 8.1 ± 4.9 ($P = 0.012$). No complications were observed.
Conclusions: LT ablation seemed effective in OSAS with retrolingual obstruction in failure of medical treatment.
Level of evidence: 4.

1. Introduction

Obstructive sleep apnea syndrome (OSAS) is a frequent and major medical concern [1]. It is responsible for multiple complications related to daytime sleepiness and induces cardiovascular pathologies [2]. Obstructive sleep apnea syndrome is characterized by iterative partial or complete obstruction of the upper airway (pharynx) during sleep. Specific narrowing of the retrolingual space is thought to be due either to tongue malposition or to increased tongue-base volume. In some cases, tongue volume is normal but the lingual tonsils (LT) are hypertrophied.

The LTs consist of lymphoid tissue mainly localized on either side of the tongue base in Waldeyer's ring, present in some cases during adulthood [3]. Different surgical treatments aim to widen the retrolingual space. In case of anatomical lingual malposition due to mandibular retrusion, the most effective treatment is bimaxillary advancement; but this is heavy and selective surgery [4]. In case of functional lingual malposition, electrical stimulation of the hypoglossal nerve [5] enables widening of the retrolingual space during sleep; however, this treatment is expensive. Other surgical techniques include tongue base volume reduction by either a transoral or a transcervical approach [6,7], and repositioning of tongue insertions (hyoid suspension, genioglossus transposition, and lingual suspension) [6].

In terms of surgical techniques, there is often some confusion between base of tongue reduction surgery and lingual tonsillectomy; however, isolated lingual tonsil involvement is rare and rarely described in the literature.

The different surgical techniques that target LT hypertrophy need to be further evaluated.

The main goal of the present study is to evaluate the efficacy and tolerance of surgical ablation of the LTs in OSAS.

2. Materials and methods

All patients with sleep disorder were seen in our ENT head and neck surgery outpatient clinic. Questionnaires were used to assess snoring qualitatively and measure daytime sleepiness on

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the Epworth Sleepiness Scale (ESS) [8]. Patients also underwent full clinical ENT examination in seated position, oriented towards sleep respiratory disorders and identifying obstruction sites on flexible endoscopy to evaluate the nasal fossae, tongue base, epiglottis, and lingual tonsils.

When OSAS was suspected, a sleep study was routinely prescribed, with type I, II or III polysomnography (PSG). OSAS was defined by apnea-hypopnea index (AHI) > 10/h, and classified as mild (10 < AHI < 15/h), moderate (15 < AHI < 30/h) or severe (AHI > 30/h).

When OSAS was diagnosed with retrolingual space identified as the site of obstruction, conservative treatment was introduced, by continuous positive airway pressure (CPAP) or an oral dental appliance (OA). In case of failure of conservative treatment, usually due to poor tolerance or partial efficacy, patients often ask for definitive surgical treatment. In such cases, drug-induced sleep endoscopy (DISE) was performed to identify the sites and mechanism of obstruction (DISE was not routinely performed in our department before 2010). In case of doubtful diagnosis of hypertrophied lingual tonsils, T2-sequence MRI was performed. Hypertrophy was assessed by measurement of LT height and maximum thickness on sagittal slice.

DISE was carried out in a surgery room under anesthesia maintained by propofol using an electrical syringe to reach a target concentration. The depth of sleep was monitored by the standard vital signs and the bispectral index (BIS) [9].

During DISE, obstruction sites were identified and classified qualitatively according to the VOTE classification [10].

In case of retrolingual obstruction with LT hypertrophy, lingual tonsillectomy was considered.

Surgical ablation was performed under general anesthesia with nasotracheal intubation, or sometimes orotracheal intubation without tracheotomy. Then a laryngoscope was introduced, with a multi-valve retractor at the level of the lingual V to enable good exposure. Surgery was performed using a microscope or more rarely a rigid endoscope.

Lingual tonsillectomy classically uses a diode laser with continuous firing by a fiber vehicle (Dormier Medtech), or else coblation (Coblator, Arthrocare, USA). Hemostasis was carried out throughout the intervention, using monopolar forceps. Lingual tonsillectomy was performed one side at a time, dissecting down to the vallecula. Muscle fibers of the tongue were conserved. Access to the inferior part of the vallecular was achieved by repositioning the tongue depressor deeper in the middle for proper visualization of the free border of the epiglottis.

Postoperative hospital stay was extended until recovery of oral feeding and pain relief without intravenous analgesics. Antacids were prescribed. Tolerance was assessed by duration and type of analgesia (WHO steps 1, 2 or 3) and pain intensity on a 0–10 scale.

Patients were seen after discharge in the outpatient clinic one week after surgery to complete evaluation of tolerance of the procedure, then again between 3 to 6 months to evaluate efficacy: patient interview with Epworth score and assessment of residual snoring. Subsequently, a sleep study was performed within 6 months.

We studied retrospectively the cases of all patients who underwent isolated LT ablation from 2007 to 2016.

The primary endpoint was AHI after surgery. Patients were considered cured when AHI was less than 10/h. Surgery was considered effective for AHI < 20/h with 50% reduction. Reduction in ESS and snoring were secondary endpoints. Tolerance of the surgical procedure was evaluated based on postoperative complications (respiratory, hemorrhage, and infection), hospital stay, analgesic level, and time to resumption of oral feeding.

2.1. Statistics

Means and standard deviations were calculated. The Mann-Whitney test was used to compare pre- versus postoperative date. Results were considered significant when P values were < 0.05.

3. Results

Eleven patients (5 males and 6 females) fulfilled our inclusion criteria. Mean age and body mass index (BMI) were 44.3 ± 12.6 years and 28.8 ± 5.6 kg/m^2 respectively. Obesity (BMI > 30 kg/m^2) was found in 63.6% of the patients. Ten patients had snoring disorder, 8 had gastroesophageal reflux disease (GERD), 6 were smokers, and 8 had history of palatine tonsillectomy. 8 patients were considered CPAP or OA failures, and 3 refused initial conservative treatment (CPAP or OA) and requested first-line LT ablation.

Mean preoperative AHI was 29.5 ± 21.7/h. Four patients had mild OSAS (AHI = 12.5 ± 5.7/h), 3 moderate OSAS (IAH = 24.7 ± 1 .6/h), and 4 severe OSAS (AHI = 50.0 ± 11.3/h). Mean AHI in obese and non-obese patients was 32.3 ± 25.0/h and 24.5 ± 16.2/h, respectively.

MRI was performed in 9 patients to confirm diagnosis of OSAS. LTs were 32.3 ± 15.6 mm long and 19.7 ± 9 mm thick, measured on sagittal slices.

Nine patients underwent DISE. Four showed complete isolated retrolingual obstruction, 4 complete retrolingual obstruction plus oropharyngeal or soft-palate obstruction, and 1 showed partial retrolingual plus soft-palate obstruction.

Diode laser was used for 9 patients, and coblation for 2 patients. Mean operative time was 102 ±22 mins, with no significant difference between laser and coblation techniques.

Mean hospital stay was 3.1 ± 1.5 days. The mean postoperative pain score was 5.9 ± 2.5/10. All patients received paracetamol during their stay. Ten, however, needed a step-2 analgesic (tramadol) and 3 a step-3 analgesic (morphine). Only step-1 painkillers were necessary at discharge, for a mean 12.2 ± 3.0 days.

Normal oral feeding was resumed at 1.1 ± 0.8 days after surgery. Mean BMI progressed from 28.6 ± 5.1 kg/m^2 preoperatively to 27.9 ± 5.3 kg/m^2 postoperatively.

No complications were observed.

Postoperative PSG was performed at a mean 6.2 ± 3.2 months. All patients showed a drop in AHI, from a mean 29.5 ± 21.7/h to 11.6 ± 9.6/h (P = 0.005): i.e., 60% reduction in AHI. Five patients (45.0%) were considered cured; surgery was considered effective in 7 (55.0%).

Fig. 1 shows results according to severity of OSAS. In the severe group, AHI decreased from 50 ± 11.3/h to 19 ± 11.3/h; in the moderate group, from 24.7 ± 1.6/h to 9.0 ± 7.9/h; and in the mild group, from 12.5 ± 5.8 to 6.2 ± 3.9/h.

Fig. 2 shows results according to type of obstruction: complete obstruction versus complete obstruction associated with a second site. In isolated complete retrolingual obstruction (n = 4), AHI decreased from 22.2 ± 8.8/h to 5.6 ± 1.8/h (74.5% decrease); in retrolingual obstruction associated with a second site (n = 4), AHI decreased from 22.3 ± 10.9/h to 15.5 ± 6.0/h (30.5% decrease).

Mean ESS normalized, decreasing from 13.0 ± 3.4 to 8.1 ± 4.9 (P = 0.012) (38.4% decrease). The majority of patients reported no further snoring (66%).

4. Discussion

This study showed that patients with OSAS experience a clear reduction in nocturnal respiratory events after isolated lingual tonsil ablation when hypertrophied tonsils were implicated in pharyngeal obstruction during sleep. There is also improvement in

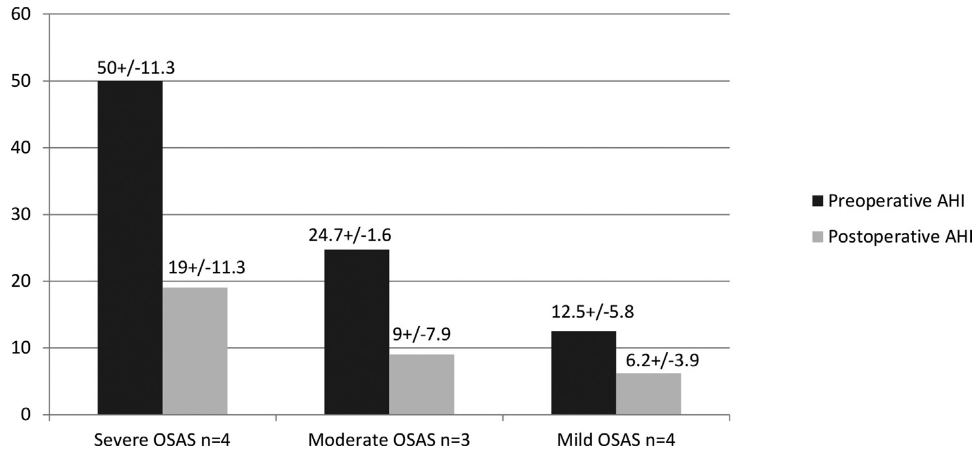

Fig. 1. AHI progression according to severity of OSAS.

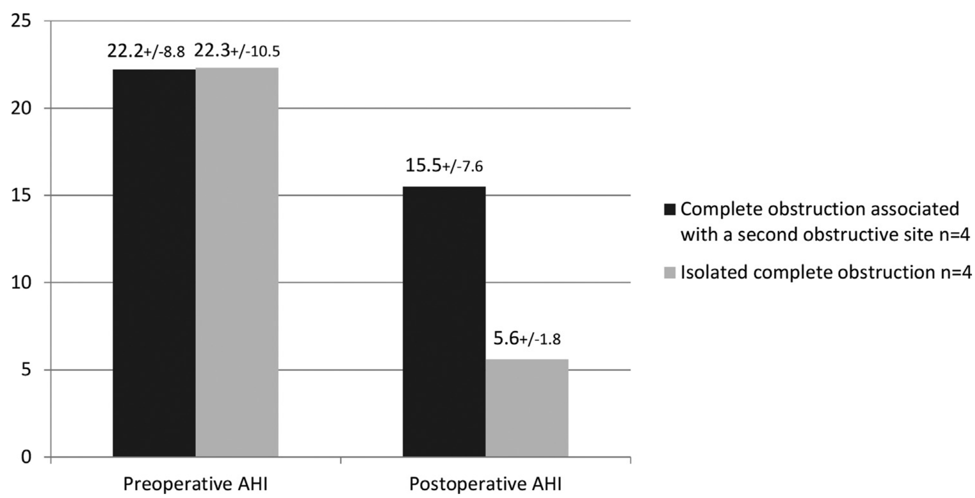

Fig. 2. AHI progression according to type of obstruction.

daytime sleepiness and snoring. The procedure takes less than 2 hours, and is associated with minimal complications.

The first reports of treatment of LT hypertrophy date from the late 19th century. Dr Lennox-Browne, an otorhinolaryngologist in Cambridge, UK, described the symptoms linked to LT hypertrophy in his book “The Throat and Nose and Their Diseases”, published in 1899. Unfortunately, the literature on this pathology today is very sparse.

Multiple factors seem to be associated with LT hypertrophy, such as obesity, smoking, GERD, and previous palatine tonsillectomy [11–14]. Several mechanisms have been suggested: smoking and GERD both induce lymphoid tissue irritation leading to hyperplasia, and palatine tonsil ablation increases exposure to these irritants. In our study, we found a high frequency of the same irritating factors.

These factors are also associated with OSAS; obesity is a clear risk factor, with an odds ratio of 4.17 [1], and smoking has an odds ratio of 4.44 [15]. Wise [16] found GERD in 75% of patients diagnosed with OSAS, though the mechanism is not very well understood. One of the theories is that GERD induces LT hypertrophy which in turn produces obstruction and OSAS.

LT assessment uses awake flexible endoscopy of the retro-basilingual space. According to Friedman, LT can be classified into 5 grades (0–4) depending on tongue-base covering, and LT thickness and level with respect the tip of the epiglottis [17]. Clinical examination plus MRI confirm diagnosis of LT hypertrophy, but DISE is still necessary from a surgical point of view. DISE allows proper visualization of the retro-basilingual obstructions by the LT, and better sheds light on the mechanism of obstruction: primary obstruction related directly to hypertrophied LTs, or secondary obstruction related to the LTs pushing on the epiglottis and leading to a backward tilt [19].

DISE can also correct information collected during awake clinical examination. In a study comparing awake clinical examination and DISE, the retrolingual space appeared as a site of obstruction in 28% of cases during wakefulness, versus 69% during DISE in the same population [18]. DISE can also quantify the obstruction as complete or partial. Finally, it can show other sites of obstruction: soft palate, oropharynx, epiglottis [19].

Regarding surgical LT ablation, the Frenchman Emile Moure proposed conservative treatment by iodinated mouthwash gargles in his textbook “Elementary Treatments and Practice of ENT Diseases”, published in 1904. In case of failure, he recommended lingual tonsillectomy using Lennox-Browne’s tonsillotome.

The surgical ablation of the LT is a delicate intervention: exposure may be restricted by retrognathia, macroglossia or small mouth opening, which are all very common anatomical characteristics seen in patients with OSAS. The LTs can also be an obstacle to intubation and ventilation [22], necessitating temporary tracheotomy in some cases [20]. In our series, no tracheotomies were required.

Exposure is usually endoscopic. The operative field is visualized by microscope or 70° rigid endoscope [23]. The implantation of the lingual tonsils in the back of the tongue base, which is even more vertical [12,21] in patients with OSAS, makes access even more difficult even when exposure is good. Therefore, other methods have been developed: the transcervical approach [24] and transoral robotic surgery [25]. The present series exclusively used an endoscopic. Hoff [27] studied the robotic transoral surgical system in 285 patients undergoing tongue base or LT surgery, and reported that the procedure was feasible, with crossover needed, and with an average surgery time of 86 mins, compared to 102 mins in our series. The percutaneous transcervical approach [24] seems to be a good method to overcome exposure difficulties. On the other hand, this approach needs an ultrasound examination performed by a radiologist and also needs specific instruments.

For lingual tonsillectomy per se, CO_2 [23] or diode laser, electroresection, coblation or radiofrequency coblation are used [27]. All these techniques are usually performed under general anesthesia and seldom under sedation with spontaneous ventilation.

The laser [23] is a good cutting tool which, coupled to a microscope, separates the dissection planes and reduces the risk of excessive resection and thus of intra- and postoperative bleeding and pain. In our series, we used the diode laser in most cases. This laser is preferable to the CO_2 laser: its wavelength makes it more hemostatic, consequently reducing surgery time; this was observed in a study on the palatine tonsils, where the diode laser was compared to the CO_2 laser [26]. The coblation technique, which uses low temperature [27], allows gradual reduction of the LT and can better respect resection limits; however, resection is more limited, with greater risk of recurrence.

In our study, almost half of the patients with OSAS were cured, with an AHI drop of 60%. Very few articles studied isolated LT ablation in OSAS. Dundar [20] reported a single patient with OSAS, cured by lingual tonsillectomy. Muderris published preliminary results of a retrospective study of 6 patients [21] that evaluated transoral robotic surgery in hypertrophic lingual tonsils. He showed a somewhat similar drop in AHI after lingual tonsillectomy, from 27.5/h preoperatively to 6.3/h postoperatively. The results of tonsillectomy in OSAS need to be compared to other techniques involving widening of the retro-basilingual space. Maxillomandibular advancement surgery seems to be the most effective treatment, as shown in a meta-analysis [30], with 87% efficacy in first or second line. However, it is a long and difficult procedure. Moreover, it entails a high risk of dental malocclusion and sensorineural deficit in the lower face. Hypoglossal stimulation [5] was also shown to be an effective technique, with a drop in AHI from 29.3/h to 9.0/h after one year; it was well tolerated, with a 4.5% rate of complications such as pain, infection or secondary defective device removal; most importantly, it is a very expensive treatment. A meta-analysis [6] compared the various surgical procedures involving tongue-base volume reduction and tongue insertion repositioning, with efficacy ranging between 35% and 68% [29], compared to 55.0% in the present study.

The different surgical techniques used for widening the retro-basilingual space have been shown to be effective in the treatment of OSAS. It can also be interesting to associate different techniques to increase efficacy: e.g., to associate LT ablation and hypoglossal nerve stimulation.

Our study also highlighted the importance of DISE, allowing some prognosis of surgical results. AHI drop seemed to be greater isolated complete retro-basilingual obstruction. The only real failure in our series was an incomplete LT resection, linked to the surgical technique rather than topographic diagnosis.

There were no major intra- or postoperative complications. Hoff [25] likewise did not report any serious postoperative complications, in contrast to Murrderis [28] who reported 2 cases of oropharyngeal stenosis in robotic lingual tonsillectomy associated with epiglottoplasty.

Our study had certain limitations. Its lack of power is due to the small population and the retrospective design. Patients were selected according to awake clinical data rather than DISE findings.

The study nevertheless provides an initial approach to treating OSAS. It could be a support for future prospective studies with larger populations, to better define the selection criteria for patients likely to benefit from this procedure. A longer efficacy evaluation period could also identify risks of residual or recurrent pathology. It would also be interesting to compare the different surgical techniques, especially between diode laser and radiofrequency coblation and/or robotic surgery.

5. Conclusion

In OSAS with narrowing of the retro-basilingual space related to LT hypertrophy, after failure of conservative medical treatment, LT ablation seems to be effective. It is a relatively low-cost and simple procedure, with few complications. Preoperative clinical diagnosis of LT hypertrophy is based on DISE, and is a key element for successful surgery.

Disclosure of interest

The authors declare that they have no competing interest.

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