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Abstract
Background: This study evaluates the efficacy of lingual tonsil resection by transoral robotic surgery (TORS) in a large group of patients with recurrent lingual tonsillitis (RLT).
Methods: Eighty-four patients with RLT treated with a lingual tonsil surgical resection using TORS were analyzed in terms of their postoperative results, disease recurrence, postoperative dysphagia, and quality of life.
Results: A reduction of the mean number/year of acute lingual tonsillitis (LT) episodes emerged after surgery (5.17 vs 0.54 events), comparing the mean number of preoperative and postoperative LT episodes, a statistical significance emerged ($P = .0001$). The postoperative endoscopic evaluation showed 94.1% of patients with absent or poor lymphatic tissue on the tongue base. Evaluation of postoperative dysphagia showed good results with an average score of 85.9 ± 7.5.
Conclusion: This article explains how in patients with chronic LT with medical therapy failure and impaired quality of life, TORS could represent a valid therapeutic option.
K E Y W O R D S
chronic lingual tonsillitis, postoperative swallowing, quality of life, transoral robotic surgery

1 | I N T RO DU CT I O N

Lingual tonsillitis (LT) is an acute inflammation of the lymphatic tissue of the base of tongue (BOT; lingual tonsil), possibly appearing with pharyngeal symptoms as foreign body sensations, pharyngodynia or dysphagia, and fever and/or signs of upper airways infection.1-4
Due to non-specific symptomatology, LT has often been underestimated by physicians and rarely investigated in the scientific literature. LT etiopathogenesis is somewhat unknown. The published studies about the topic reported that lingual tonsils inflammation could be correlated to lymphatic hypertrophy of the lingual tonsil tissue (LTH), pharyngolaryngeal reflux due to

repeated mucosal insult, upper airways infections, and use of drugs such as phenytoin. 5-11

LT diagnosis may not be easy: clinicians can use a medical history (pharyngeal symptoms as foreign body sensations, pharyngodynia, dysphagia, and fever and/or signs of upper airways infection) and fiber optic examination (acute inflammation of the lymphatic tissue of the BOT and purulent exudate at the base of the tongue) as tools to suspect acute episode of LT (Figure 1). When multiple events per year of acute LT are present, the diagnosis of recurrent lingual tonsillitis (RLT) is made.2-5

LT it is treated with the same therapy administered in pharyngotonsillitis cases (antibiotics and corticosteroids). However, as in the case of pharyngeal tonsillitis, in case of RLT inflammation due to the need to take medical therapies several times a year, this condition could negatively impact on the quality of life of patients.4,5

In the last few years, the base of the tongue has always been defined as an area of difficult surgical approach in non-oncological pathologies due to exposure problems, access, and manipulation of this region. Nowadays, with the introduction of the transoral robotic surgery (TORS) with the Da Vinci surgical system (Intuitive Surgical, Inc., Sunnyvale, California), tongue base surgery has considerably increased, especially in non-oncologic pathologies. Today, the main conditions to perform lingual tonsillectomy using TORS are squamous cell carcinoma and obstructive sleep apnea syndrome (OSAS) related to LTH.12,13

In a preliminary report in 2017, Montevecchi et al14 first proposed the feasibility and efficacy of the trans-oral robotic surgical approach to remove the lingual tonsil in cases of chronic inflammation.

The purpose of this article is to describe the use of TORS to perform a lingual tonsillectomy, in a large group of patients suffering from RLT. The robotic surgical technique for lingual tonsil resection is described. TORS efficacy in terms of resolution of infective episodes/year, possible complications, and functional outcomes are reported. Finally, the suitability of TORS in the case of LT is discussed.

2 | MATERIALS AND METHODS

2.1 | Study protocol

This retrospective single-center study was performed at the Otolaryngology, Head and Neck, and Oral Surgery Department of the Morgagni Pierantoni Hospital in Forlì, Italy.

Five-hundred TORS procedures performed in our tertiary referral center between May 2008 and October 2019 were initially reviewed. One-hundred and four patients treated with TORS surgery to remove the lingual tonsil lymphatic tissue following recurrent inflammation of lingual tonsil (RLT) were initially selected for the study protocol. Data were first extracted and assessed by the principal investigator (M. D.), and thereafter independently (studied) by two of senior co-authors (C. V. and G. I.) using standardized data forms. Definite inclusion and exclusion criteria were applied for the study patient's enrollment.

2.2 | Inclusion and exclusion criteria

Patients of all ages were included in the study.
Clinical and endoscopic diagnosis of RLT was the first inclusion criteria.
Definite criteria and/or the number/year of LT that indicate the need for a surgical tonsillectomy was not defined from the existing literature. In our clinical practice, we have considered more than four episodes/year of acute inflammation of the lingual tonsil for the surgical indication of robotic tonsillectomy. Besides, all enrolled patients had continuous symptoms of benign LTH, reducing their quality of life, such as sore throat, change in the speech, foreign body sensation, dysphagia, and airway obstruction according to the literature.15
Patients subjected to TORS for cancer or OSAS were excluded from the study. The same patients with an endoscopic follow-up <6 months were considered not suitable for the study because the clinical follow-up was considered too short.
Finally, patients whose preoperative and postoperative data were not collected, and lost patients during clinical follow-up were not included in the study.

2.3 | Investigated features

Clinical features of enrolled patients were collected concerning the preoperative and postoperative period: pharyngeal clinical symptoms, episodes of LT/year, and drug use (antibiotics or corticosteroids)/years. Tonsillectomy performed at a young age was also investigated.
Preoperative and postoperative lingual tonsil evaluation, TORS surgical time, intraoperative removed tissue, and postoperative complications were collected, analyzed, and reported.

2.4 | Lingual tonsils evaluation

In our center, a physical examination with a flexible laryngoscopy and a video recording were performed in all patients as part of routine airway examination preTORS and annually during clinical follow-up.
Endoscopic records of the enrolled patients were observed to collect preoperative and postoperative Friedman classification. In this study, the data regarding the preoperative endoscopic evaluation (before surgery), and the final follow-up (last endoscopic evaluation recorded) were reported.
The video was reviewed by the same observer (M. D.) to have a consistent valuation of the LT grade assigned. According to the Friedman grading system (Table 1), lingual tonsil hypertrophy was graded on a scale ranging from 0 to 4.9,16,17

2.5 | Notes of surgical technique

TORS procedures have been performed with the Da Vinci Surgical System (Si model, Intuitive Surgical, Inc.) according to the robotic setting established by O'Malley et al18 (Figure 2).
Visualization of the BOT region was obtained with a 30º up, high magnification, and a three-dimensional endoscope (Figure 2A). A 5 mm Maryland forceps and a 5 mm monopolar cautery with a spatula tip were used.14
The procedure usually requires a piecemeal resection in two surgical steps because being lingual tonsil generally composed of two sub-units divided by the glosso-epiglottic ligament (Figure 2B-D). In this way, it is possible to identify and preserve the main anatomic structures of this region.14,18,19

The volume (cm3
) of the removed LT was measured in all the procedures using a graduated syringe partially filled with water into which the surgical pieces were placed, thus causing a rise in the level of the water.20
All samples were sent for pathologic evaluation after surgical resection to confirm the presence of LTH. Immunohistochemical staining of the samples (CD 20, CD79a, CD5, CD30, CD10, bcl2, and bcl6 expression) to rule out lymphoproliferative disorders was performed.

2.6 | Quality of life and postoperative dysphagia

A cohort of enrolled patients were contacted (by telephone and email) to assess their quality of life (Glasgow Benefit Inventory; GBI) and post-operative dysphagia with the use of GBI and MD Anderson Dysphagia Inventory (MDADI) questionnaires.
The GBI was designed to evaluate the effect of ear, nose, and throat (ENT) surgical procedures on the patient's quality of life, with a brief interview of 18 questions. The answers were summarized to give a total score ranged from −100 (poorest outcome) to +100 (best result). GBI was also divided into three distinct subscales that investigate the general, social, and physical impact of a specific surgical procedure. 21
The MDADI questionnaire is a self-administered, psychometrically validated questionnaire published by Chen et al in 2001. 22 It was developed to evaluate the impact of dysphagia on quality of life after head and neck surgical procedures. We used this questionnaire to assess postoperative dysphagia in patients who underwent TORS for chronic LT. A composite score from 20 (extremely low functioning) to 100 (high functioning) was reported.

2.7 | Statistical analysis

Mean age, time of follow-up intraoperatively removed tissue, and time of surgical procedure were compared using the Student's t test. A value of $P < .05$ was considered statistically significant.
The Wilcoxon test was used to evaluate differences in the number of acute LT/year and drug use before and after TORS procedures. The same analysis was used to compare the differences in preoperative and postoperative Friedman classification. A value of $P < .05$ was considered statistically significant.
The Chi-square test was used to evaluate differences in preoperative and postoperative symptomatology in all patients examined. A value of $P < .05$ was considered statistically significant.
Linear regression testing was carried out to examine a possible correlation between the number of LT/year and the volume of the LT removed.

2.8 | Ethical standards

This research study was performed following the principles of the Declaration of Helsinki (1964) and approved by the Institutional Review Board of the Morgagni Pierantoni Hospital, Forlì, and all patients expressed their consent to the study enrollment.

3 | RESULTS

3.1 | Patient's features

Eighty-four patients who underwent TORS lingual tonsillectomy for a diagnosis of chronic LT (16 male and 68 females; mean age 48.4 years old; range 14-74 years old), were considered suitable for the study analysis under the inclusion and exclusion criteria. The patient's features have been reported in Table 2.

Patient characteristics with significant statistical significance included female sex ($P < .0001$). 83.3% of enrolled patients had a history of palatal tonsillectomy when a child; 16.7% still had palatine tonsils.

3.2 | Preoperative clinical symptoms and Friedman classification

Preoperative clinical signs associated with LT are reported in Table 3. Persistent foreign body sensation and persistent pharyngodynia were the significant symptoms of chronic LT reported in the study group (71.4% of patients). The preoperative endoscopic evaluation showed 25 cases (29.7%) with LTH grade 3 and 59 (70.3%) with LTH grade 4, according to the Friedman classification (Table 1).

3.3 | Intraoperative and postoperative outcomes

Mean TORS surgical procedure was 31.9 ± 9.3 minutes (range 10-50 minutes). No significant intraoperative complications occurred; a not programmed tracheotomy due to intraoperative intubation problems was necessary in two (2% and 3%) of patients. The mean volume of lymphatic tissue removed was 11.7 ± 6.2 cm^3. All patients had a postoperative anatomopathological diagnosis of lymphoid follicular hyperplasia. Postoperative bleeding occurred only in one patient (1.1%) of the study group (Table 2). Postoperative bleeding occurred as a surgical complication approximately 7-10 days after the procedure; it was observed due to the fall of tonsillectomy scabs from the base of the tongue. No visible nerve damage (branches of the glossopharyngeal nerve) was observed during surgical procedures.

A possible correlation between the number of acute LT/year and the amount of intraoperative tissue removed was tested using regression analysis; no significant association was observed (Figure 3, $P = .9$).

3.4 | Clinical and endoscopic outcomes

The mean time of the clinical follow-up was 49.6 ± 27.1 months (range 6-109 months).

The endoscopic follow-up evidenced a typical BOT scar in 79 patients (94%), while an exuberant scar tissue (excessive proliferation of granulation tissue) was observed in five cases (6%). Postoperative endoscopic evaluation (Friedman classification) showed 94.1% of patients with grade 0 or 1 and 5.9% of patients with grade 2 or 3. Differences between preoperative and postoperative BOT classification are reported in Table 1. Statistical differences emerged in all subclassification groups ($P < .05$ in each case).

A reduction of the mean number/year of acute LT episodes emerged after surgery in the investigation of clinical symptoms (5.15 vs 0.97, Table 4); comparing the average number of LT in the preoperative and postoperative period, a statistical significance emerged ($P = .0001$, Figure 4).

The same statistical difference emerged regarding preoperative and postoperative drug use (P = .0001; Table 4). Differences in preoperative and postoperative symptoms referred to chronic LT are reported in Table 3; postoperative dysgeusia was present in eight patients (9.5%) of the study as a result of a robotic base tongue surgery.

No differences in preoperative and postoperative persistent dysphagia emerged (P = .3).

3.5 | Quality of life after surgery and post-operative dysphagia

A cohort of 60 (71.4%) patients among those enrolled in the study were subjected to questionnaires to assess their quality of life after surgery (GBI) and post-operative dysphagia (MDADI Anderson).

The positive impact of surgery in the quality of life emerged: the general subscale showed a mean value of +49.5 ± 21.5, the social subscale had a mean value of +28.8 ± 20.3, and the physical subscale registered a mean value of +65.8 ± 32.1 (Figure 5).

The MDADI Anderson's questionnaire results showed a mean value of the composite score of 85.9 ± 7.5 (range 50-95.7) (Figure 5). None of the patients enrolled presented a grade compatible with functioning dysphagia.

4 | DI SCU SSION

Palatine, nasopharyngeal, and lingual tonsils constitute the famous Waldayer ring at the level of the upper airway.23 Recurrent inflammation and adenotonsillar hypertrophy are widespread, especially in the pediatric population, and surgery is often necessary to reduce acute events of infection/inflammation and possible associated disorders.4,24

Moreover, it has long been established the collapse site changes due to the effects of aging in older patients. 25 As with inflammation of the palantine tonsils, inflammation of the lingual tonsil is also possible. The frequency and duration of this infection define the condition of chronic LT. However, RLT is a misunderstood pathology, often associated with LTH and pharyngeal symptoms. Even though many studies have tested the importance of lingual tonsil hypertrophy in sleep disorder breathing, very few studies have analyzed the frequency of RLT and its etiopathogenetic factors. Persistent inflammation of the lingual tonsil, similar to the palatine and adenoid tonsils, may require repeated drug use and reduces patient's quality of life who suffer from it. 3 Parham and Newman15 showed RLT as a tangible entity with debilitating symptomatology in everyday life treated with the same therapy administered in pharyngotonsillitis cases: antibiotics and corticosteroids. Since 1989 authors have been discussing LTH as a result of hypopharyngeal infection translated in terms of recurrent epiglottitis. Wilson et al 26 noted their tonsillectomies patients had an enlargement of their lingual tonsils on follow-up examinations implicated in the development of these recurrent inflammation episodes.

Montevecchi et al14 first reported in 2017, the usefulness of TORS to treat 10 patients suffering from LT. They removed a mean volume of lymphatic tissue of 16.5 ± 13 cm3 . No major intra-operative and post-operative complications were observed. After a 15.2 ± 12 months follow-up, no relapses were reported, and patients did not complain of any postoperative symptoms related to chronic LT.

TORS offers many advantages in the BOT surgery: it presents an excellent and high-resolution three-dimensional vision of the operative field, which is not easily obtainable through the conventional surgical methods offered for the BOT surgery (ablation, CO2 laser). Besides, it allows access to hidden areas such as the tongue base and reduces the physiological tremor of the surgeon's hand.27,28 For expert surgeons, this technique eliminates the need for tracheostomy and the risk of postoperative bleeding from the BOT region. Our data demonstrated base tongue bleeding only in 1/84 (1.1%) case for which a surgical revision was necessary.

In this article, we report our considerable experience of TORS uses to treat patients with RLT. In our study, LTH was noted in 83.3% tonsillectomies patients: Guimaraes et al showed that children undergoing tonsillectomy showed a higher degree of BOT hypertrophy (78%) compared to a non-tonsillectomies' patients control group (22%).26,29,30 It would appear that hypertrophy of the lingual tonsil occurs as a compensatory response after palatine tonsils removal. LTH can cause two types of symptoms depending on the time of onset: persistent complaints due to known LTH and overlaps symptoms in case of LT inflammation. All patients enrolled presented preoperative disabling complaints due to recurrent lingual tonsil inflammation. The most frequent symptoms were contextual foreign body sensation and pharyngodynia in 26.2% of cases, and persistent pharyngodynia in 45.2% of cases. Patients were subjected to TORS only if two conditions were present: RLT resistance to different pharmacological therapies and symptoms, reducing their quality of life. Postoperative data regarding the 84 patients enrolled were reviewed after a mean long-term follow-up of 49.6 ± 27.1 months. During postoperative fiberoptic endoscopic evaluation emerged that 59.5% had a grade O by Friedman, 40.5% had an LTH grade 1, whereas 5.8% presented a postoperative grade 2 or 3. As recently reported by Iannella et al,30 these data confirmed that the regrowth of BOT lymphatic tissue after TORS is very low (8.8% of cases).

Focusing on the medical history, our data showed interesting results, considering a significant long-term follow-up: a clear reduction of acute inflammation of the LT was obtained, and a substantial-stop in antibiotics or corticosteroids use was shown. Another encouraging observation was the reduction of persistent symptoms in daily life: only 15.4% presented pharyngodynia with a statistical difference with the preoperative prevalence of this symptom ($P = .0001$).

Foreign body sensation was still present in 9.5% of patients after surgery. However, patients complained of a different perception of this symptom, probably related to BOT tissue scarring after surgery. In some cases (9.5%), according to literature, postoperative dysgeusia, perceived as taste alteration or metallic taste occurred: however, in our experience, this symptom tends to disappear over time; patients of the study who complained dysgeusia had a short follow-up (<1-year). 31 No visible nerve damage (branches of the glossopharyngeal nerve) was observed during surgical procedures, although small nerve branches can be resected during this procedure if the resection is extended to the lingual body. Besides, the prolonged compressive effect of the mouth retractor on the lingual

body during surgery would be responsible for this postoperative condition.

A little increase of dysphagia as food stuck in the throat was noted with a postoperative incidence of 8.3%. Two explanations can justify this last finding: the first is the abnormal scarring at the BOT region, and the second is diminished sensitivity at the BOT due to neural damage. However, a minimal significant impact of TORS on the swallowing function was confirmed in the results of the MDADI test performed to 60 patients of the study. The MDADI is the first validated and reliable self-administered questionnaire that measures patients' swallowing function. The result of the MDADI Anderson questionnaire showed a mean value of the composites score of 85.9 ± 7.5 (range 50-95.7). This is a great result considering that cut off which dysphagia is a disabling clinical symptom is under 20 for a composite score. None of the patients enrolled presented a grade compatible with functioning dysphagia. In 2015, Eesa et al 31 evaluated the outcomes related to swallowing function in patients who underwent TORS for sleep apnea by MDADI questionnaire, which showed a non-significant increase of short-term impact on swallowing function (4.58 ± 7.03 preoperative MDADI score vs 5.18 ± 8.32 post-operative; P = .56). There are no studies reported in the literature related to the quality of life of patients who underwent TORS for RLT. Quality of life of enrolled patients was tested with the use of the GBI questionnaire. The GBI was a sensitive and accurate tool for identifying outcomes between surgical and medical otolaryngology interventions reporting positive and negative scores.21 Excellent results were obtained on the quality of physical life, which showed a clear improvement by setting a result of 65.8 ± 32.1, good outcomes were showed for general grade GBI too, with 49.5 ± 21.5. In the social grade benefit, the GBI showed a value of 28.8 ± 20.3. These data confirm the improved state of health in patients treated by TORS for RLT. Lingual tonsillectomy is not a standard treatment for chronic LT. This study could be an opportunity to progress in the treatment of LT, as it is well defined that the robotic instrumentation provides a valuable tool to ease the surgical resection of the lingual tonsil.

As reported in literature a valid alternative to TORS for the BOT reduction is coblation surgery. 32-35 A systematic review of the literature that compared these two surgical procedures reported that mean rates of failure were 34.4% and 38.5%, respectively, in TORS and coblation groups. Complications occurred in 21.3% of the patients treated with TORS and in 8.4% of the patients treated with coblation surgery. 32 Therefore, TORS seems to give slightly better results, allowing a wider surgical view and a measurable, more consistent removal of lingual tissue. However, the higher rate of minor complication and the significant costs of TORS must also be considered. 33-35 Lingual tonsillectomy by TORS is a quite straight forward technique that originates from the extensive clinical experience for the treatment of sleep apnea patients and in the diagnostic schemes for the unknown primary cancer. 30,31 Therefore, in our opinion, TORS can also be safely applied to patients with chronic inflammatory diseases of the lingual tonsil.

5 | CONCLUSION

In the case of chronic LT when medical therapy fails, and the patient's quality of life is impaired, it is possible to consider a surgical hypothesis with the use of TORS.

This article showed an improvement in the healthy state of patients after TORS for RLT with a significant reduction in the occurrence of acute inflammation and a substantial-stop in drug use. Excellent results in the postoperative quality of life and swallowing function were reported.

CONF LICT OF IN TEREST

The authors declare no potential conflict of interest.

ETHICAL APPROVAL

All procedures performed in studies are under the ethical approval of the Morgagni Pierantoni Hospital institution.

INFORMED CONSENT

Informed consent from all individual participants included in the study was obtained.

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