Exstirpation of symptomatic lingual thyroid with transoral robotic surgery (TORS): A promising novel treatment option

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ABSTRACT
Ectopic thyroid is a rare condition most often found at the base of the tongue, lingual thyroid (LT). The majority of patients are asymptomatic. Recently, transoral robotic surgery (TORS) has emerged as an option for definitive treatment. Here, we present a 20-year-old patient with symptomatic LT, treated with TORS-assisted surgery without adverse events and with depletion of symptoms. We advocate TORS as a very promising means to be considered when encountering the rare condition of symptomatic LT.

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Introduction
The thyroid gland originates from the pharyngeal pouches that later form the base of the tongue and migrates caudally during the fifth to seventh week of gestation. Migration failure during the embryological descent leads to an ectopic thyroid. It may arise anywhere along the midline of the neck but is most frequently found in the base of the tongue, i.e. a lingual thyroid (LT) [1]. Ectopic thyroid is a rare entity with a prevalence suggested to be around 1/100,000, but numbers are uncertain [2] and presumably due to definition. This is emphasised in that Santangello et al. [3] recently reported ectopic thyroid tissue in as many as 0.9% of cases in a large thyroidectomy material, and also in that microscopic congregations of thyroid tissue is encountered in 10% of normal tongue base tissue [4]. Parathyroid tissue, with its separate embryologic origin, may also be ectopic, but is not encountered in the base of the tongue [5].

Hypothyroidism is overrepresented among patients with ectopic thyroid, including LT. Furthermore, LT can be associated with obstructive symptoms such as dysphagia, dysphonia, cough, snoring, sleep apnoea and foreign body sensation [6]. However, LT may also be fully asymptomatic.

It is debatable whether asymptomatic LT require treatment [7]. Careful follow-up for asymptomatic and euthyroid patients has been recommended [6]. Treatment options for symptomatic patients includes levothyroxine (LT4) treatment, radioactive iodine therapy and surgery.

The non-invasive treatment options for LT are LT4 substitution/suppression and radioactive iodine therapy. However, especially with LT4 treatment, only partial response, if any, can be expected, and side effects in the long term can be deleterious. In the case of radioactive iodine therapy even enlargement of thyroid tissue may arise after treatment, although total responses have been reported [8], and the treatment is associated with harmful effects on gonads and other organs [6], and thus not neither suitable in younger patients. On the other hand, surgical treatment in the historic context has been associated with pronounced morbidity due to the access-related problems with open surgery. Recently, transoral robotic surgery (TORS) have emerged as a promising alternative to open surgical treatment, with, to our knowledge, the first procedure described in 2011 [9]. A PubMed database (www.ncbi.nlm.nih.gov) search on the 29th of July 2022 using either ‘ectopic thyroid’ AND
transoral robotic', 'lingual thyroid' AND 'transoral robotic', or 'lingual goiter' AND 'transoral robotic' as entries, retrieved only 14 publications comprising 18 cases [9–22]. Further literature research for this study, generated by these publications, provided 6 more cases [7,23], thus in total 24 published cases, reflecting the novelty of the TORS approach in LT as well as the rarity of the diagnosis itself. In this report, we describe the first LT patient in Scandinavia treated with TORS.

**Case presentation**

A 20-year-old patient presented at a secondary hospital with a foreign body sensation in the throat and eating difficulties, and experienced difficulty breathing, especially when lying flat; symptoms that had become more distinct the weeks before first consultation. At clinical examination, a large round mass at the base of the tongue was noticed (Figure 1). TSH was elevated. Primary work-up included a CT-scan (Figure 2) and ultrasound examination. The tumour in the base of the tongue was well defined and cervical thyroid was missing. LT was suspected.

The patient was referred to a tertiary hospital for further diagnostic work-up including thyroid scintigraphy and LT4 treatment was started. The symptoms escalated despite LT4 treatment, and active treatment was considered. Radioactive iodine therapy was discussed but deemed insufficient for the intended purpose and unsuitable due to expected side effects. Open surgery was also considered but deemed inappropriate in regard to symptoms by both patient and managing doctor. Agreement was reached on referral to another tertiary hospital for evaluation for possible TORS-assisted treatment. After referral, treatment options were once more considered and discussed with the patient, and it was decided to opt for TORS-assisted surgical treatment.

At surgery, a video laryngoscope-assisted nasal intubation was performed to optimize the procedure and hence reduce the risk of intubation-related injury of the LT. 250 mg of hydrocortisone and 3 g of benzylpenicillin were intravenously administered. A suture was placed in the front of the tongue for easier manipulation. Exposure was established with a Crowe-Davis mouth gag. Utilizing a Da Vinci Xi robotic system (Intuitive Surgical, Sunnyvale, CA), a robot-assisted dissection was performed with monoo- and bipolar electrocautery starting frontally (Figure 3). The frontal and lateral portions were relatively avascular while the caudal part was more vascularized, in line with the pre-operative radiologic work-up. The larger vessels were handled with bipolar electrocautery without need for clips. Resection was performed en bloc, macroscopically radical without extra margins, and minimal peri-operative bleeding was noted. Thorough electrocautery of the wound cavity was performed and a local hemostatic agent, Surgiflo (Ethicon, Raritan, NJ), was applied. The console time for the procedure was 52 min. The airway was not compromised, and the patient was extubated at the end of surgery. A nasogastric tube was placed and the patient was referred to a post-operative ward for 4 h, and after fiberoendoscopic examination further referred to a common ward. The patient was discharged without nasogastric tube 4 days post-surgery with post-operatively adjusted LT4 treatment. Histopathology verified normal thyroid tissue, with the expected exception that C-cells were not encountered, and no signs of malignancy were found.

At follow-up 4 months after surgery, the patient experienced complete relief of prior symptoms, there was no globus sensation, and the patient could eat normally and did not experience any breathing difficulties in any settings. The only remaining complaint was an occasional retention of food while eating which could be relieved immediately by drinking. A follow-up program for thyroid hormone levels and LT4 treatment had been established. Clinical examination showed a normalised anatomy (Figure 4).

**Discussion**

This article reports, to our knowledge, the first TORS procedure for LT in Scandinavia. Furthermore, it highlights the great potential for this technique in the
management of this rare condition; a technique utilized in this condition for over a decade, but yet with still only a few reported cases.

The diagnosis of LT can be associated with pronounced symptoms, and thus warrant treatment. Due to the high morbidity associated with surgical treatment in the historical context, LT4 and radioactive iodine ablation have been treatment options of choice, though options with expected incomplete response and especially in the latter case risk of pronounced adverse effects. The patient reported was treated with LT4 but nonetheless experienced worsening of symptoms. Radioactive iodine ablation was considered but deemed inappropriate in the specific case.

In contrast to open surgery, TORS provides a more non-invasive access route with associated far less morbidity. A further advantage with the TORS approach, partly reflecting this, is that time under anesthesia can be anticipated to be significantly shorter. In the presented case, the operating time was short, which is in consistency with similar cases [9,11,12,22], and thus in sharp contrast to the expected time required for open surgery.

The major potential drawbacks in utilizing TORS are the risk of bleeding events and the risk of airway obstruction. Significant bleeding related to TORS utilized in surgery for oropharyngeal cancer has been reported to occur in approximately 5.5% of cases [24]. It should be emphasized however, that even if cases of LT can be expected to be well vascularized, TORS for LT is still a less extensive procedure than the majority of TORS procedures, which are primarily utilized in cancer treatment such as for oropharyngeal
cancer, with hence associated larger resections, exposure of major vessels and risk of airway edema. A further drawback to consider for TORS is system availability, associated with the large cost for surgical robotic systems and the training of console surgeons. However, procedures like LT treatment will never be the main usage for such systems, or the main cases for TORS console surgeons, and should rather be viewed upon as supplementary benefits of a technique used for other causes.

In the described case, no adverse effects were noted. The airway could be handled with extubation at the end of procedure. At follow-up, the patient was relieved of prior symptoms; a marked difference to the pronounced symptoms at presentation, and this was in line with clinical findings. We advocate TORS as a very promising means to be considered when encountering the rare condition of symptomatic LT.

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Informed consent

Written informed consent for publication, including imaging and clinical information, was obtained from the patient.

Disclosure statement

No potential conflict of interest was reported by the author(s).

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