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CASE REPORT

Robotic resection of lingual thyroglossal duct cyst in an infant

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Abstract
Thyroglossal duct cysts (TDCs) are the most common congenital midline neck masses. Lingual TDC is a rare variant that emerges as an isolated cyst at the tongue base. Unlike conventional procedures aiming removal of cyst, duct, and hyoid bone via open surgical access, a transoral cystectomy is mostly sufficient in cases with lingual TDC. We present a case describing a 3-month-old infant patient with lingual TDC who was successfully treated with transoral robotic surgery. The cyst wall was completely excised with no complications or obvious bleeding occurred. The operating time was 10 min. He had an uneventful postoperative course. Six months postoperatively, he is free of symptoms with no evidence of recurrence. Surgical treatment of lingual TDC in an infant is possible with transoral robotic approach and minimal risk of complication. Further studies are strongly needed to confirm the safety of robotic surgery in pediatric population.

Keywords Lingual thyroglossal duct cyst · Transoral robotic surgery · Infant

Introduction

Thyroglossal duct cysts (TDCs) are the most common congenital midline neck masses [1]. The thyroglossal duct occurs during the embryological development of the thyroid gland. It is the embryological remnant of the path that the thyroid gland descends from the foramen caecum of the tongue base to the neck region. The thyroglossal duct closes off between 5th and 16th gestational weeks after the thyroid gland reaches its final position in the pretracheal region. Any alteration in this process could lead to persistent ducts and development of cysts or fistulas [1].

TDCs can be seen anywhere along the course of thyroglossal duct, predominantly at the hyoid bone level. Lingual TDC is a rare variant that emerges as an isolated cyst at the base of tongue and does not exhibit continuity. It constitutes 3.8% of all TDCs [2]. Unlike TDCs in other localizations, it is often symptomatic. If not treated, it may cause fatal upper respiratory tract obstruction especially in newborns [3].

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Traditionally, the most common surgical technique in the management of TDCs is the Sistrunk procedure [1]. This procedure involves excision of the cyst together with hyoid bone and fistula tract. As the lingual TDC does not persist with a duct and because of its localization, transoral approach is more suitable in this group of patients [2].

Transoral robotic surgery (TORS) is a new minimally invasive approach that is increasingly used in the surgical treatment of benign and malignant lesions of the oropharynx. However, studies regarding the efficacy and safety of TORS in the pediatric population are rather limited [4, 5]. In this report, we presented a rare case of lingual TDC in an infant patient treated with TORS. To the best of our knowledge, this is the second infant case in which lingual TDC is treated with robotic surgery.

Materials and methods

A 3-month-old male infant presented to our clinic with complaints of respiratory distress, dysphagia, and frequent lower respiratory tract infections. The patient had stridor and nutrition problems since birth, and that complaints increased gradually in recent times. Flexible nasopharyngoscopy showed a well-defined mass extending from the tongue base to the epiglottis (Fig. 1a–c). A computed tomography (CT) scan revealed a 3 × 3 cm hypodense mass, located at the

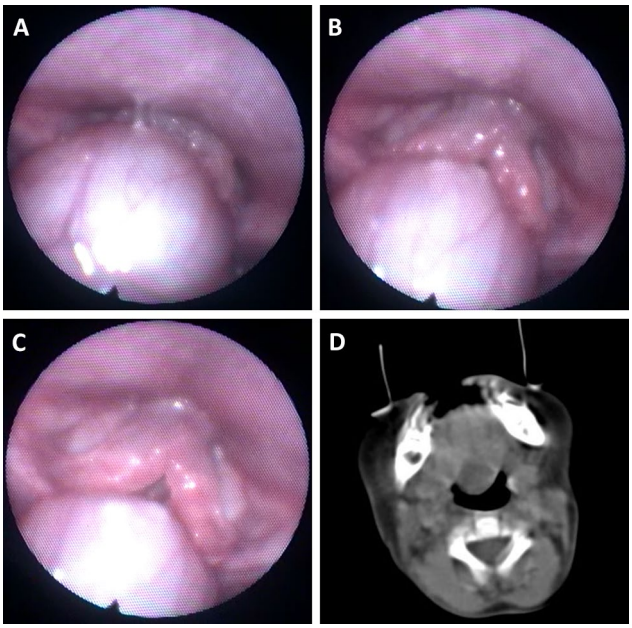

Fig. 1 **a–c** Flexible nasopharyngoscopic view of the cyst. **d** Computed tomography view of the cyst on axial plane

tongue base (Fig. 1d). The thyroid gland was in its normal position. The infant underwent TORS, after providing a written consent from his parents.

Following oral intubation, the patient's head was positioned in a supine position at 30° extension. To support neck extension, shoulder supports were applied. A Feyh-Kastenbauer® laryngeal retractor (Gyrus Medical, Maple Grove, MN) could not be inserted because of small mouth of the infant. The tongue was pulled forward with the help of a suture, and Farabeuf retractors were used to provide exposure of the tongue base.

The Da Vinci XI® robotic surgical system (Intuitive Surgical Inc., Sunnyvale, CA) was approached to the patient from the left side. The robotic arm number three was equipped with a 30° robotic camera; number two with fenestrated bipolar forceps; and number four with EndoWrist® monopolar cautery. An assistant surgeon was positioned on the patient's right to assist aspiration.

A cortical incision was made on the mucosa of the tongue covering the cystic lesion (Fig. 2a). During the dissection, the cyst was ruptured and a mucoid content was aspirated. Subsequently, the dissection was continued down to the lingual face of the epiglottis and vallecular mucosa, and the cyst wall was completely excised with no complications or obvious bleeding occurred (Fig. 2b–c).

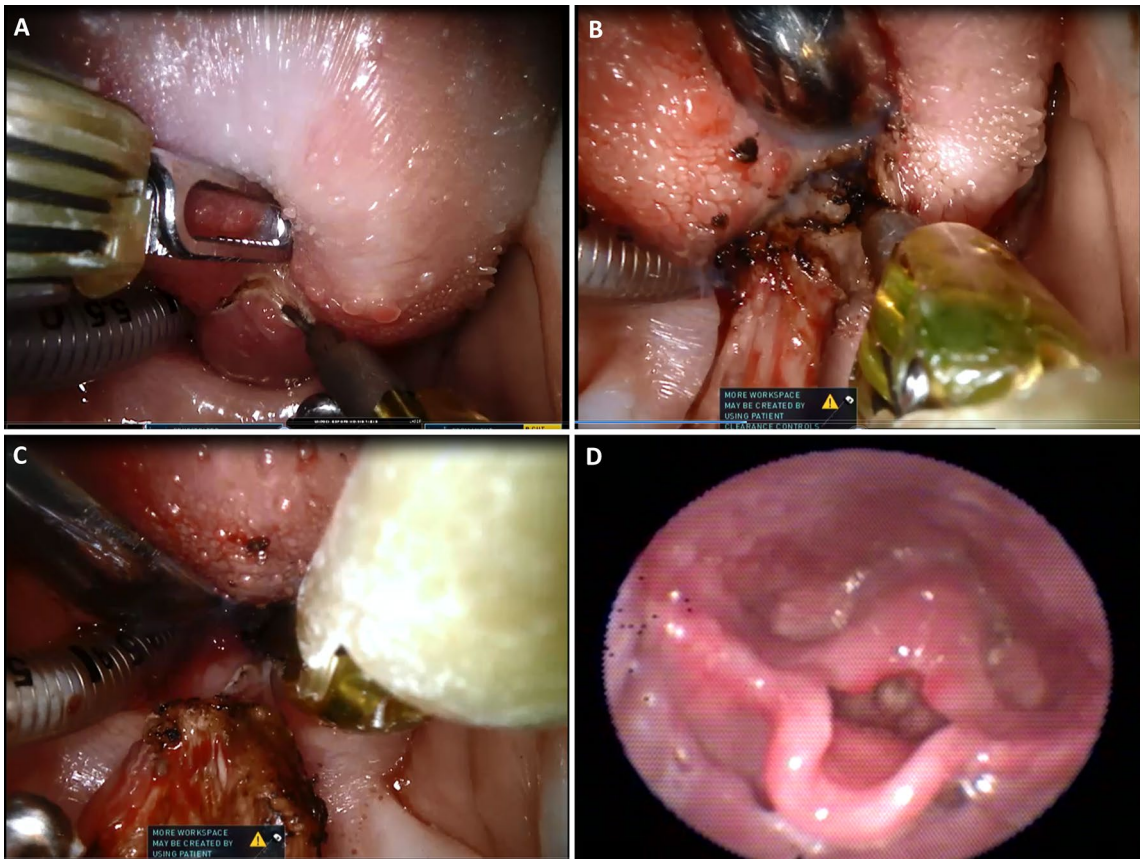

Fig. 2 **a–c** Cortical incision, dissection, and resection steps of the robotic surgery. **d** Flexible nasopharyngoscopic view 6 months after surgery

Results

The operating time was 10 min. The infant was followed up for 1 day in the intensive care unit. On the second day, he was taken to the ward and oral feeding was started. He had an uneventful postoperative course and was discharged home on postoperative day 7. Histopathologic examination was consistent with lingual TDC. Six months postoperatively, he is free of symptoms with no evidence of recurrence (Fig. 2d).

Discussion

Treatment of lingual TDCs differs from TDCs in other localizations. Unlike conventional Sistrunk procedure which aims to remove cyst, duct, and hyoid bone via open surgical access, a transoral cystectomy is mostly sufficient in cases with lingual TDC [2]. In the largest series, Burkart et al. [6] reported the outcomes of 16 patients with lingual TDC treated with endoscopic transoral cystectomy. The authors noted that all patients recovered without complication and have shown no evidence of recurrence during a median follow-up of 3.7 years.

Robotic surgery is a novel surgical approach that has recently been used in the field of otorhinolaryngology. It was initially used as a minimally invasive technique in the treatment of head and neck cancers. Over time, with increasing evidences of its efficacy and safety, indications for robotic surgery have extended to different fields such as surgical treatment of obstructive sleep apnea [7]. However, there is still lack of sufficient data regarding the use of robotic surgical system particularly in the pediatric airway. In a preliminary study, Rahbar et al. [8] assessed the application and safety of TORS in five pediatric patients with laryngeal cleft and in four pediatric cadaver larynxes. The authors reported that use of the surgical robot on cadaver larynxes provided great dexterity and precision, and relatively easy suturing. However, despite two patients with laryngeal clefts having successful treatment with TORS, the surgical robot could not be used on three out of five pediatric patients due to limited transoral access. On the other hand, in one of the largest series of TORS in the pediatric population which included 16 children with a wide range of ages (14 days to 15 years) and weights, Zdanski et al. [5] reported that all patients had successful robotic access, and no children had conversions to open or conventional endoscopic approach.

To date, only three case reports described successful use of TORS specifically in the treatment of lingual TDCs [9–11], of which only one reported a pediatric case [11]. In that report, Kayhan et al. [11] demonstrated the marsupialization of lingual TDC with TORS in a 2-month-old infant. The authors reported that no complications were observed, all symptoms were completely resolved and the infant was discharged on postoperative day 3. Our report is the second in which lingual TDC is treated via TORS in an infant. Both in the previous report [11] and in our report, the most challenging problem faced in application of robotic surgery to an infant was the limited surgical field due to the small anatomy. In addition, robotic surgical systems have not been specifically designed for pediatric population. Therefore, operating in a small area with standard instruments designed for adults makes the procedure more difficult. Smaller size of instruments and/or single port technique may facilitate TORS in this group of patients.

In conclusion, surgical treatment of lingual TDC in an infant patient can be performed successfully with transoral robotic approach and minimal risk of complication. However, further studies are strongly needed to confirm the safety of robotic surgery in pediatric population.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval This article does not contain any studies with human participants or animals performed by any of the authors.

Informed consent Written informed consent was obtained from the patient for publication of this case report/any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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