A rare complication of the Sistrunk's procedure in thyroglossal duct remnant: tracheal injury

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Management of thyroglossal duct remnants (TGDRs) is a common clinical concern in childhood. A girl with an unusual complication of tracheal injury secondary to the Sistrunk's procedure is presented and discussed with regard to the relevant literature on airway injuries during the procedure. Surgical resection is an optimal choice of therapy in TGDRs and may be associated with high morbidity, especially recurrence. Although the Sistrunk's procedure is a safe and successful technique, life-threatening complications should be kept in mind during the management of TGDRs. *Ann Pediatr Surg* **10:130–132**
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Introduction

Management of the congenital neck lesions (CNLs) is a common clinical concern in infants and children. The differential diagnosis includes congenital, inflammatory, and neoplastic lesions. The physicians caring for these children should be aware of different presentations as these lesions are known to be complicated by infection. An orderly and thorough examination of the neck with the understanding of embryology and anatomy of the region, and the natural history of a specific lesion concerning the CNL, will facilitate the diagnosis. Treatment modalities are different depending on their nature, symptoms, and location of the lesions.

Thyroglossal duct remnants (TGDRs) are the most common CNLs encountered in pediatric practice [1]. Early referral of these patients to pediatric surgeons for timely surgical treatment is recommended. With a correct preoperative diagnosis and an appropriate surgical management, it is possible to prevent possible complications.

A girl with an unusual complication of tracheal injury secondary to the Sistrunk's procedure during the management of TGDR is presented and discussed with regard to the relevant literature on airway injuries observed at the time of the surgical treatment of these lesions.

Case

A 3.5-year-old girl presented with a midline upper neck mass that was mobile with tongue protrusion and swallowing since her birth. No inflammation or mucoid discharge was detected, and no fistulous opening was encountered in the physical examination. She had associated Turner syndrome in addition to TGDR and was in follow-up in the Department of Pediatric Endocrinology. The physical examination revealed otherwise healthy but underdeveloped child.

Ultrasonography (US) of the neck revealed a cystic mass of 11 mm in diameter, parasagittal in location, with dense contents inside (Fig. 1). Radioisotope thyroid scanning was performed as a normal thyroid gland was not demonstrated on preoperative US and was found to be normal. MRI confirmed the cystic mass positioned as thyrohyoid in location (Fig. 2). Elective surgical excision was performed by the Sistrunk's procedure, which involved removal of the central portion of the hyoid bone together with the cystic mass. There was no fistulous tract behind the hyoid bone located upward or downward in direction.

The technique of our surgery includes transverse incision at the approximate level of the hyoid bone. Superior and inferior myocutaneous flaps are developed deep to the platysma exposing the cyst. The notch of the thyroid cartilage and the thyrohyoid membrane is recognized. Dissection continues superiorly along the membrane to the hyoid bone. The inferior and superior margins of the hyoid are identified and skeletonized lateral to the cyst and posterior surface of the hyoid is defined. The hyoid bone is lifted with a hemostat and transected with confidence at the level of the lesser cornu. After the hyoid is resected, dissection continues superiorly toward the foramen cecum. A double-gloved finger is inserted into the oral cavity and is used to identify the area of foramen cecum. The specimen is cross-clamped and divided just deep to the lingual mucosa, and the stump is oversewn with 4/0 polyglycolic acid. The wound is thoroughly irrigated, no drain is placed, and anatomic closure is performed.

The patient was extubated and transferred to the ICU where all of a sudden she developed a marked stridor, severe respiratory distress, hoarseness, and extensive subcutaneous crepitus that dispersed throughout the body. She was retransferred to the operating room, and after consultation with ENT specialist, it was decided to perform an emergent tracheoscopy, which revealed a perforation of 2 mm in diameter located in the subglottic area, 1 cm under the vocal cords on the anterior surface of the trachea. It was thought that tracheal injury was presumably secondary to the Sistrunk's procedure. Conservative treatment with tracheal intubation and mechanical ventilatory support with deep sedation of 6 days' duration was uneventful. Control MRI of the neck

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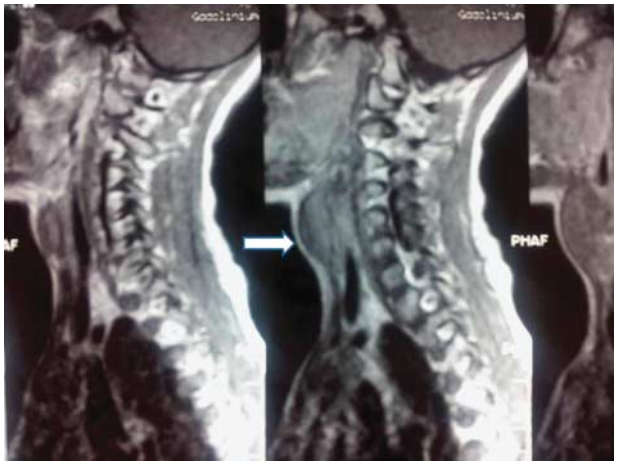
Fig. 1

Sonographic image of the cystic mass (arrow) in the patient with thyroglossal duct remnants.

Fig. 2



MRI of the cystic mass (arrow) positioned as thyrohyoid in location.

taken on the eighth postoperative day revealed a pretracheal air collection that was aspirated with a needle. Histopathologic findings of excised mass confirmed the thyroglossal duct cyst, the presence of the hyoid bone, and a fistulous tract with squamous epithelium. With 19 days of hospital stay and a follow-up period of 53 months, the postoperative course is eventless and the patient is well with no stridor.

Discussion

CNLs in children constitute one of the most intriguing areas of pediatric pathology and can produce diagnostic and therapeutic challenges for clinicians and surgeons. In the absence of infection, identification of a cystic mass in a child should raise the diagnosis of congenital malformations related to abnormal embryogenesis of the thyroglossal duct.

TGDR is by far the most common form of CNLs, accounting for up to 70–90% of such lesions [2,3]. The true incidence of TGDR is not known with certainty and in fact many are never detected clinically [2]. It is commonly observed in children or adolescents, and in a meta-analysis its incidence was found to be higher in children than in adults [4]. Equal distribution among male and female patients has been reported in most of the reviews [4–6]. Main presentations are that of a midline neck mass as in our patient or infection as a single or a recurrent event [5].

There are six diagnostic methods in the preoperative evaluation of TGDR. These include US, computed tomography, MRI, fine needle aspiration (FNA), radioisotope thyroid scanning, and thyroid function test [7]. US is the most common test ordered for children [8]. It is readily available, noninvasive, and offers information of both the TGDR and thyroid gland [9]. Although the tract could not be completely visualized in our patient, US was useful to confirm that the palpable mass was cystic. The absence of a normal appearing thyroid gland in US prompted us to obtain preoperative thyroid scintigraphy, which was reported to be normal. Concerning FNA, although the diagnostic sensitivity of 62% and a positive predictive value of 69% are reported, FNA is not popular for diagnosing TGDR in children [7,10]. For this reason, our patient did not undergo FNA for preoperative diagnosis of TGDR.

The most common location for the cystic mass in TGDR is close proximity to the hyoid bone with an incidence of 66%, but other locations including lingual, suprahyoid, suprasternal, or within the thyroid gland have also been reported [4,11]. Our patient shows similarity with most of the children in the literature as the cyst was adjacent to the hyoid bone.

Complications after the Sistrunk's procedure may be divided into minor and major categories as shown in Table 1. Although the incidence of minor complications after the Sistrunk's procedure has been noted up to 29%, these problems are relatively insignificant and may be treated easily in an outpatient setting [12]. Even though possible major complications of TGDR surgery are rare, recurrence, which is the most commonly investigated major complication after the Sistrunk's procedure, is reported to be 2.6–5% [13–15]. Even in a most recent independent review article, it was reported that one of the 10 operated children experienced

Table 1 Possible complications of the Sistrunk's procedure

Major complications
Recurrence
Abscess or hematoma requiring drainage
Transfusion
Inadvertent entry into the airway
Tracheotomy
Nerve paralysis
Hypothyroidism
Malignancy
Death
Minor complications
Seroma
Local wound infection
Stitch abscess
Wound dehiscence

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recurrence after the Sistrunk's procedure, and vast majority of recurrent cases were diagnosed within a few weeks after initial surgery [16]. It is reported that fewer than 1% of patients with TGDRs may have malignant tissue, usually well differentiated thyroid carcinoma [17]. Fortunately, there was no malignant transformation or recurrence during the follow-up period in our patient.

Although much has been written about TGDR, little has been reported regarding the airway complications associated with the Sistrunk's procedure. Inadvertent entry into the airway is one of the major complications associated with the Sistrunk's procedure and, if untreated, may produce fatal consequences. Absent from the literature are sporadic verbal accounts of major anterior laryngotracheal injury that may occur when the rigid framework of the anterior neck is misidentified. To the best of our knowledge, there is one pediatric case series with four children describing major laryngotracheal injury complicating the Sistrunk's procedure in the English language literature [18]. The thyroid cartilage was mistaken for the hyoid bone in all of the children in that report, and laryngotracheoplasty with the usage of cartilage grafts was performed on these children and all of them ultimately required tracheotomy [18].

As all of the children reported in the literature with airway injuries associated with the Sistrunk's procedure had thyroid cartilage injury to some extent [18], our patient was fortunate to have had a rather small perforation of 2 mm in diameter located in the subglottic area on the anterior surface of the trachea, with the thyroid cartilage being intact. A conservative approach of tracheal intubation with ventilatory support was the treatment modality in our patient. Our case is unique in that, apart from a successful conservative treatment of airway injury, to the best of our knowledge, there are no cases of Turner syndrome associated with TGDR in the English language literature.

Avoiding major laryngotracheal injury during thyroglossal duct excision begins with proper preoperative planning and the perioperative utilization of anatomic landmarks to guide resection. Before surgical treatment, the physician should palpate to locate the cyst in relation to the cricoid and thyroid cartilages and the hyoid bone. In the operating room, safe TGDR excision begins with preservation of the cyst and its tract during dissection in the anterior neck. Before removal of the mid-portion of the hyoid bone, one should palpate the cricoid cartilage, the cricothyroid membrane, the thyroid cartilage, the thyrohyoid membrane, and finally the hyoid bone. Special attention must be paid to the relationship between the TGDR, its tract, and the hyoid notch [12]. In young patients, the ossified hyoid bone may over-ride the top of the thyroid cartilage placing the cartilage at risk for misidentification and resection, and surgeons must remain oriented to midline cervical anatomy [18].

Conclusion

Surgical resection is an optimal choice of therapy in TGDR and its management may be associated with high morbidity, especially recurrence. Early referral of these patients for pediatric surgeons and accurate and timely surgical treatment is suggested. Although the Sistrunk's procedure is a safe and successful technique with low complication rates, rare and sometimes life-threatening complications as in our patient with tracheal perforation should be kept in mind during the management of these children.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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