

CASE REPORT

Laryngeal perforation during Sistrunk procedure

Leonor Matos*, Lúcia Carvalho, Alexandre Alves and Mário Nora

General Surgery Department, Centro Hospitalar Entre Douro e Vouga, Santa Maria da Feira, Portugal

*Correspondence address. General Surgery Department, Centro Hospitalar Entre Douro e Vouga, 8th floor, R. Dr. Cândido Pinho 5, Santa Maria da Feira 4520-211, Portugal. Tel: +351 256 379 700; Fax: +351919507788; E-mail: leonormatos@hotmail.com

Abstract

Thyroglossal duct cyst (TGDC) is the most common congenital neck anomaly in childhood, although it can be diagnosed at any age group. Ultrasound of the thyroid gland and the neck structures is an appropriate tool for diagnosis. The authors aim to report a rare case of a 22-year-old male with a laryngeal perforation during a TGDC excision and raise awareness to this potentially life-threatening complication. The Sistrunk procedure is the surgical treatment of choice with a low morbidity and recurrence rates.

INTRODUCTION

Thyroglossal duct cyst (TGDC) is the most common cause of midline congenital cyst formation in the neck, accounting for >75% of such lesions [1].

The thyroid gland begins its embryological development in the 3rd week of fetal life at the level of the foramen cecum and descends in the neck, anterior to the hyoid bone. During this descent, the thyroglossal tract is established. If this duct does not regress, a remnant may persist and contribute to the formation of a TGDC.

It is imperative to comprise this knowledge to make a correct preoperative diagnosis and proper surgical treatment to prevent future complications and recurrence.

The authors report a case of laryngeal perforation in a 22-year-old man who had undergone a Sistrunk procedure. The perforation was successfully repaired by direct closure with absorbable suture.

CASE REPORT

A 22-year-old man with no relevant past medical history presented a midline upper neck mass since his birth. It was a small mass, mobile with tongue protrusion and swallowing, without

inflammation, mucoid discharge, or any fistulous opening. Ultrasonography of the neck revealed a 7 mm cystic mass in the midline, underlying to the geniohyoid muscles, in the proximity of the hyoid bone.

Elective surgical excision was performed, a Sistrunk procedure, which involved removal of the central portion of the hyoid bone along with the cystic mass. The cystic mass was firmly adhered to the anterolateral wall of the larynx, which made the dissection more difficult. There was no fistulous tract behind the hyoid located upward or downward in direction.

Our surgical technique includes transverse incision at the approximate level of the hyoid bone. Superior and inferior myocutaneous flaps are developed deep to the platysma to expose the cyst. The notch of the thyroid cartilage and the thyrohyoid membrane are identified and dissection continues superiorly along the membrane to the hyoid bone. The margins of the hyoid are identified and outlined lateral to the cyst and the muscles attached to the center of the bone are resected. The hyoid is lifted and transected at the level of the lesser cornu. From the hyoid up to the level of the foramen cecum, the duct with the surrounding tissue is then removed, as needed.

During isolation of the cystic mass, we noticed a 3 mm right anterolateral laryngeal perforation, which prompts us to warn the

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anesthesia team. Afterwards, due to the found perforation, we quickly decided to close primarily with absorbable suture.

To support our decision, an otorhinolaryngologist was called to the operating room and we proceeded with the surgery. The wound was thoroughly irrigated, no drain was placed, and the incision was closed in subcutaneous and cutaneous planes.

The patient was extubated and transferred to our department where he had an uneventful postoperative course. He was discharged on the 4th day and evaluated at the postoperative consultation, without any noteworthy event. No control exam was required. Histopathologic findings of excised mass confirmed the TGDC and the presence of the hyoid bone.

DISCUSSION

TGDC is the most common congenital neck anomaly in childhood [1]. Despite being frequently highlighted in pediatric population, a significant percentage of them are diagnosed in the second decade of life, and they can also present later in adulthood [1].

TGDC originates from persistent epithelial remnants of the thyroglossal duct that are present during the descent of the thyroid gland from the foramen cecum to its final position in the anterior neck [2].

They are commonly located in the midline in close proximity to the hyoid bone or along the course of the embryological thyroid duct [3]. Primary carcinoma is rare, representing around 1% of cases [4, 5].

TGDC usually presents as a painless midline neck swelling that moves with deglutition and protrusion of the tongue [1, 6]. Failure to anticipate its possibility may be related with the performance of an inadequate surgical technique, which is associated with significant recurrence rates.

Despite the lack of consensus regarding preoperative imaging in patients with TGDC, ultrasound remains the method of choice [1, 7].

Open surgical excision—Sistrunk procedure—is the mainstay of surgical approach. This consists of transcervical cystectomy, resection of the central part of the hyoid bone and resection of the foramen cecum, if needed, depending on the location of the cyst and the path of the thyroglossal tract [3, 8].

Potential major complications, such as nerve paralysis, hypothyroidism, blood transfusion, hematoma/abscess, which require surgical drainage or death, are rare [9].

One of the most feared complications is inadvertent entry into the airway. To minimize this risk, the surgeon must use meticulous surgical technique, remain oriented to midline cervical anatomy, identify key anatomical landmarks and choose wisely the time of surgery, specifically if there has been a recent infected cyst. If laryngotracheal injury occurs, its characteristics—size, depth, location, injury extent and mechanism—and the patient's respiratory status—have to be dully considered in order to define the best surgical option.

Surgical alternatives include direct repair as performed in our patient, strap muscle flap or cartilage interposition graft. Transection of the sternohyoid and the sternothyroid muscles ('strap muscles') at their insertion, while preserving their origin, allow for 180° rotation. By preserving the muscular origin and carefully dissecting to achieve rotation, these muscle flaps act as a well-vascularized patch, helping to close larger defects [10]. Alternatively, conchal cartilage graft can be used in selected cases, with individual adaptation of the size of the graft to the perforation. Although technically demanding, it is a surgical option to overcome these challenging circumstances.

In conclusion, TGDCs are the most common cause of midline congenital cyst formation in the neck, appearing mainly in the pediatric group. Its location, clinical presentation and embryology origin must be taken into account to make an accurate preoperative diagnosis and an appropriate surgical technique. The Sistrunk operation is the standard surgical treatment. Although it has a low morbidity and recurrence rates, rare and sometimes life-threatening complications may occur, as presented in our patient with a laryngeal perforation, and should always be kept in mind during the management of these patients.

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