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CASE REPORT / ПРИКАЗ БОЛЕСНИКА

Thyroglossal duct cyst as a cause of dyspnea in a two-year-old child

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SUMMARY

Introduction Thyroglossal duct cysts are developmental, epithelial lesions localized in the neck's median line. They occur mainly in children and adolescents; however, they also occur in one-third of patients older than 20 years of age. The symptoms dependent on the size and location along the path of the thyroglossal duct.

Case outline This article presents a case, with diagnostic imaging and surgical treatment, of a two-year-old girl hospitalized due to dyspnea, caused by a large cyst localized at the base of the tongue during an upper respiratory tract infection. Before surgery, biochemical examinations, diagnostic imaging were performed to exclude ectopic thyroid tissue. Surgery was performed applying Sistrunk's procedure, which entailed excising the cyst's tissue at its origin.

Conclusion In patients presenting with thyroglossal duct cysts, upper respiratory tract infections increase the probability of discovering previously existing cysts. A cyst localized around the foramen cecum can cause inspiratory and expiratory dyspnea.

Keywords: thyroglossal duct cysts; dyspnea; children; Sistrunk's procedure

INTRODUCTION

Thyroglossal duct cysts (TDC), representing the most commonly occurring congenital lesions of the neck (around 70%), are epithelial lesions located in the mid-sagittal plane of the body [1, 2]. They occur primarily in children and adolescents (approximately 7% of the population) irrespective of sex [3]; however, only one in three of patients may be older than 20 years [4, 5].

At around the third week of embryonal development, in the *foramen cecum* region at the base of the tongue, the thyroid gland bud forms, which subsequently descends the neck, creating the thyroglossal duct. It achieves its final position at about six weeks and regresses in the eighth week. A cyst is a result of seromucous secretions through the persistent duct. Considering its embryological derivative, the cyst remains in communication with the body of the hyoid bone [6].

We present a case of a two-year-old child admitted to the hospital because of stridor, who was then diagnosed with lingual TDC. Clinical and radiographic features leading to a diagnosis are described and equated with those which are reported in the literature.

CASE REPORT

A two-year-old girl was presented to the Emergency Department with symptoms of acute laryngeal obstructive (inspiratory and expiratory) dyspnea noticed by the parents a few days before admission. Medical history revealed fever up to 39°C, as well as difficulty in swallowing solid foods. The parents became alarmed by the appearance of dyspnea and stridor. The girl was admitted to Pediatric Department. Laboratory studies on admission showed elevated inflammatory markers: C-reactive protein (CRP, norm: 0–5 mg/l) – 53.22 mg/l, leukocytosis (WBC, norm: 4–10 × 10^3/ul) – 23.23 × 10^3/ul. The remaining laboratory parameters were found to be within normal limits. On initial physical examination, a greyish-blue smooth mass was found at the base of the tongue, which blocked the laryngopharingeal view (Figure 1). Amoxicillin with clavulanic acid was administered intravenously (50 mg + 5 mg/kg every eight hours). A rapid improvement of the patient's condition was observed, along with a decrease in dyspnea. Inflammatory markers returned to normal. Due to sustained stridor, the child was referred for a diagnostic follow-up to the Laryngology Department. Magnetic resonance imaging (MRI) of the neck revealed a thin-walled homogenous lesion in the median plane, measuring 2 × 1, 1 × 2 cm, with

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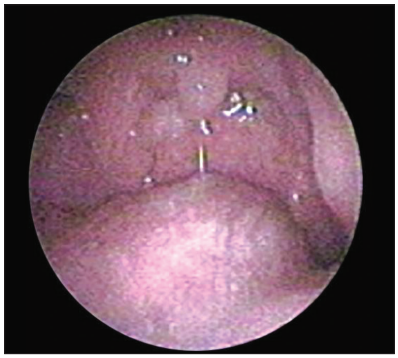


Figure 1. The endoscopic image of the thyroglossal duct cyst

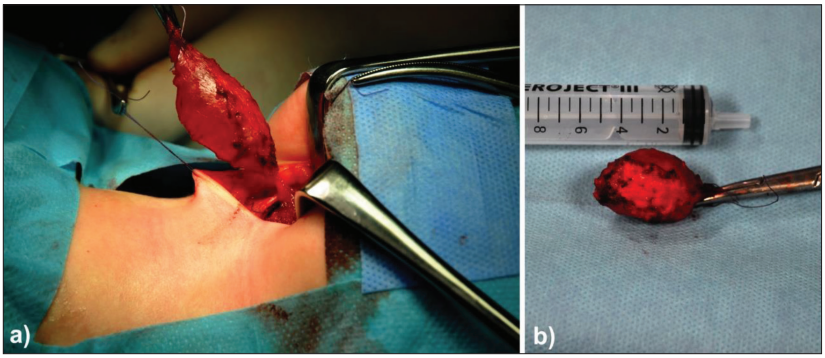


Figure 2. a) Thyroglossal duct cyst during surgical excision procedure; b) excised thyroglossal duct cyst

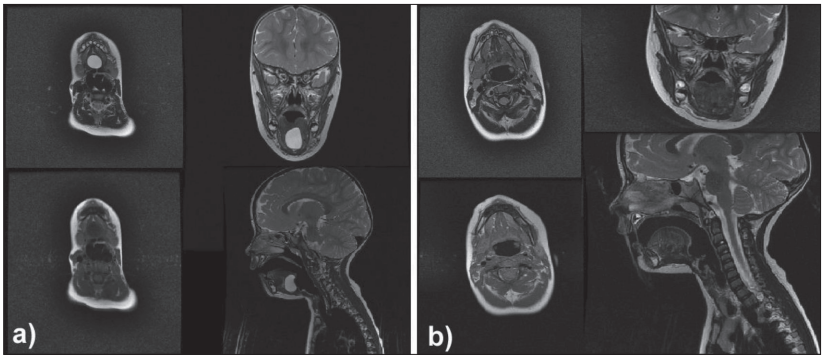


Figure 3. a) Magnetic resonance imaging (MRI) visualisation of the thyroglossal duct cyst; b) MRI of the surgical area after one year of follow-up

cyst-like characteristics in the laryngopharynx. Above, it communicated with the *foramen cecum* at the basis of the tongue and distally extended towards the hyoid bone, with a demarcated thyroglossal duct (Figure 2a). An ectopic thyroid tissue was excluded on diagnostic imaging (ultrasonography, MRI), and the presence of a properly developed thyroid gland in its anatomic position was confirmed. A provisional diagnosis of a lingual TDC was made, and the patient was referred for operative management. An anterosuperior cervical approach was applied. Intraoperative findings were consistent with a lingual TDC. The lesion was removed in its entirety from the level of the hyoid bone to the *foramen cecum* at the base of the tongue. The surgery included resection of the body of the hyoid bone (Figure 3). The postoperative course was uneventful, and the girl was discharged on the fourth postoperative day. Histological examination confirmed the diagnosis of a TDC. At follow-up review, the girl did not present any recurrence of the thyroglossal duct remnants (Figure 2b).

All procedures performed involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

DISCUSSION

The lesion usually appears in the median line of the neck in its upper 1/3 part and rarely occurs at the base of the tongue (around 2.1%) [6, 7], the suprasternal notch (approximately 10%) [8], or within the thyroid gland parenchyma [9]. It presents as a smooth, painless, and soft growth occurring in the neck at the thyrohyoid membrane level, with proximity to the adjacent hyoid bone [10].

The cause of the development of this type of lesion is unknown. One theory proposes the lymphatic tissue's infectious hyperplasia, remaining with the thyroglossal duct, leading to its closure and forming a cyst [11]. It seems that the infectious factor present in the respiratory tract and teeth, especially in children, plays a key role. Other factors predisposing to the formation of this type of lesion in adults may include pregnancy and childbirth, as well as an autosomal dominant inherited genetic factor [12].

The symptoms depend on the location and size of the lesion and may include dyspnea, dysphagia, or speech difficulties [7]. The presence of a lesion on the base of the tongue can be hazardous due to potential obstruction of the laryngeal and pharyngeal airways. This mechanism is compared with the "ball valve effect" between the cyst and the laryngeal inlet. Secondary to the respiratory tract obstruction, the patient may present with stridor, raspy respiration, and the recruitment of auxiliary respiratory muscles. Large lesions in this location in newborns and infants may be lethal [13].

In differential diagnosis, it is imperative to have in mind a fact about an ectopically placed thyroid gland – more than 90% of ectopy cases present at the tongue's base [14]. In about 5% of cases, thyroid tissue can be found in the cyst wall [9]. The differential diagnostics should include dermoid cysts and steatocystomas. They are usually situated superficially, similarly to lipomas, and present with weakly demarcated borders. More medially localized lesions, which originate from the pharyngeal grooves, could indicate a fistula's presence instead of a TDC. The remaining lesions occurring at the midline position are thyroid nodules, hypertrophy of the pyramidal lobe of the thyroid, lymphadenopathy, parotid tumors, or lymphatic malformations.

Ultrasonography is the diagnostic imaging of choice and is characterized by a high sensitivity and specificity

(higher than 90%) [15]. This examination allows for the visualization of the cystic structure of the thyroid gland. However, it does not provide information on its relation with the surrounding tissues, especially the hyoid bone. Scintigraphy and computed tomography allow for proper identification of the thyroid gland [16, 17]. MRI confirms a diagnosis of the TDC and its close correlation with the hyoid bone. It also provides objective data for measuring the lesion and depicts the exact location [15, 18]. Fine-needle aspiration (FNA) biopsy is often used to confirm or exclude the presence of lesions with cystic characteristics. Still, it is challenging to administer the procedure without anesthesia in the pediatric population [19].

Most TDCs manifest following upper respiratory tract infections or secondary to their inflammation. Treatment should be commenced with broad-spectrum antibiotics targeting the oral cavity's flora and subsequently concluded with Sistrunk's operative procedure [20, 21]. Operating in an infectious episode is contraindicated due to the high risk of recurrence of the lesion [22, 23]. The incision and drainage of the lesion can be considered if an abscess is not reacting to pharmacologic treatment only. Operative management is markedly less challenging in the absence of fibrotic changes or cutaneous fistulas [24]. An alternative to operative treatment, described in the literature, is sclerotherapy with intralesional ethanol administration. This procedure can only be implemented in cases where the neoplastic lesions surrounding the duct had been excluded. However, the literature reports that this method's success is 1/3 of patients, with a high recurrence percentage [25].

Surgical resection of a cervical cyst is the method of choice. As one of the first pioneers, Schlange described how a resection of the lesions and the body of the hyoid bone is performed in one block. This method has decreased the recurrence of symptoms by about 20%. Subsequently, in 1920, Sistrunk modified the technique and expanded it, including the excision of the TDC, the middle part of hyoid bone, and the surrounding tissue along the path of the thyroglossal tract. It is worth noting the possibility of the occurrence of ramifications or doubling of the thyroglossal duct around the hyoid bone, which may impair the surgeon's ability to recognize it intraoperatively. Leaving a fragment of the duct may cause a recurrence of symptoms. For this reason, Horisawa et al. [26] recommend removing the root of the lesion in one block, with sparing of a small margin of the surrounding tissues. Recurrence of this disease is observed in around 5% of cases [8].

Concerning patients with undiagnosted TDCs, an upper respiratory tract infection may increase the probability of intense clinical manifestation of a highly localized TDC and hasten the diagnosis. Endoscopic examination may allow better diagnosis of difficult cases of dyspnea in children. In every pediatric patient presenting with an acute episode of dyspnea, TDC should be included in the differential diagnosis.

Conflict of interest: None declared.

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Циста тироглосалног канала као узрок диспнеје код двогодишњег детета

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САЖЕТАК

Увод Цисте штитњаче су развојне, епителне лезије, локализоване у средњој линији врата. Јављају се углавном код деце и адолесцената, али и код трећине болесника старијих од 20 година. Симптоми зависе од величине и локације лезије.

Приказ болесника Овај чланак представља случај дијагностичког снимања и хируршког лечења двогодишње девојчице хоспитализоване због диспнеје, изазване великом цистом локализованом у дну језика, током инфекције горњих дисајних путева. Пре операције извршени су биохемијски прегледи, дијагностичка слика, како би се искључило ектопично ткиво штитњаче. Операција је изведена применом Систранкове методе, која је подразумевала изрезивање ткива цисте из локалитета порекла.

Закључак Код болесника са цистама тироглосалног канала, инфекције горњих дисајних путева повећавају вероватноћу откривања претходно постојећих циста. Циста око локалитета *foramen cecum* може изазвати диспнеју и током удисања и током издисања.

Кључне речи: цисте тироглосалног канала; диспнеја; деца; Систранков поступак