Stridor and lingual thyroglossal duct cyst in a newborn

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Introduction

Thyroglossal duct cysts (TDC) generally occur in the neck. The tongue is a rare site of thyroglossal duct cyst in newborns. To our knowledge, fewer than 50 cases have been described at this site [1–4]. We report the case of a newborn infant referred for stridor in whom a lingual TDC was diagnosed. The clinical, radiographic and histological features leading to the diagnosis and the treatment are described and compared with those reported in the literature.

Case report

A one-month-old baby girl was referred with stridor since the first week of life that became worse in the prone position. The parents reported feeding difficulties (eight 50 ml bottles per day) associated with failure to thrive (weight: 3480 g). On examination, the infant presented minimal supravclavicular and suprasternal tugging. Ambulatory nasal endoscopy (2.2) demonstrated short aryepiglottic folds...
and the infant was admitted to hospital with continuous monitoring.

On the following day, upper airway endoscopy was performed in the operating room under general anaesthesia. The larynx was poorly visible on laryngoscopy due to the presence of a regular, round, soft mass underneath a well vascularised mucosa, situated in the foramen caecum and obstructing the airway. Nasal endoscopic intubation was performed due to intraoperative desaturation caused by upper airway obstruction by the mass in the prone position. No tracheobronchial abnormality was observed. Contrast-enhanced computed tomography (CT) of the head and neck was performed urgently due to suspected angioma of the base of the tongue. CT showed a low-density, cystic lesion of the base of the tongue with no contrast enhancement, measuring $12 \times 13 \times 11$ mm, protruding into the oropharynx and slightly lateralised to the left (Fig. 1 A and B). The thyroid was in a normal position. Marsupialization of the cyst was performed in the operating room under suspension laryngoscopy by protecting the airways with the endotracheal tube (Fig. 2 A–C). The cyst incision was performed by laser, completed by curved scissors, releasing thick secretions. Haemostasis was ensured by application of swabs impregnated with epinephrine saline and the child was extubated after endoscopy.

The postoperative course was uneventful with resolution of stridor and resumption of normal feeding. Nasal endoscopy performed four days after the operation showed satisfactory healing allowing the child to be discharged from hospital. Histological examination revealed the presence of thick squamous epithelium, suggestive of a thyroglossal duct cyst. No recurrence was observed one year after the operation, at which time the child weighed 10 kg with a normal diet, in the process of diversification.

**Discussion**

TDCs are the most common congenital tumours of the neck, representing 40% of all congenital tumours in this site [5]. During the fourth week of embryonic development, the rudimentary thyroid gland, situated at the base of the tongue in the foramen caecum, migrates to a pretracheal position in the base of the neck. TDCs are due to an anomaly of closure of the thyroglossal duct between the thyroid gland and foramen caecum before the tenth week of intrauterine life, as secretory cells can persist throughout the course of the thyroglossal duct inducing accumulation of mucus and formation of a TDC. According to the literature, 65 to 85% of all TDCs are situated at or below the hyoid bone, while only 1 to 2% are located in the foramen caecum [6,7]. Recently, Bukart et al. published a series of 16 lingual TDC, representing 8.5% of all TDC seen in their department [4].

The clinical presentation in newborns differs from that observed in children or adults, as lingual TDCs in newborns usually cause respiratory distress or even fatal upper airways obstruction [1,8,9]. These infants often present respiratory distress with stridor and are referred with suspected laryngomalacia [10]. Nasal endoscopy is not always able to establish the diagnosis of lingual TDC when it is performed in the seated position, as the base of the tongue may be difficult to examine. However, nasal endoscopy confirms the normal morphological appearance of the larynx, thereby excluding the diagnosis of laryngomalacia. Direct laryngoscopy in the operating room is indicated in this clinical setting and demonstrates a bulging mass at the base of the tongue. CT of the neck is useful for the aetiologic work-up or when there is a persistent doubt about the diagnosis. It demonstrates a round, well-circumscribed, low-density lesion at the base of the tongue, that may obstruct the upper airways. CT may also be useful to exclude differential
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The main risk associated with these lesions is progressive enlargement leading to death from upper airway obstruction [9]. TDC must be rapidly treated by surgery to avoid respiratory complications. Two surgical techniques have been described in the literature. The first technique consists of cyst aspiration by direct laryngoscopy performed in the case of severe, acute respiratory distress when tracheal intubation is impossible due to poor exposure of the larynx [3,10]. The second technique consists of endoscopic marsupialization of the cyst under general anaesthesia usually performed with endotracheal intubation. According to the literature, the recurrence rate after endoscopic marsupialization is 17% [4].

Conclusion

Lingual thyroglossal duct cysts are very rare and, in newborns, can be confused with laryngomalacia due to their nonspecific symptoms. They present as a bulging mass at the base of the tongue obstructing the upper airway and are responsible for severe respiratory distress that can sometimes be fatal. Direct laryngoscopy and neck CT scan are essential for diagnosis. Surgical treatment must be performed as rapidly as possible. Surgical resection by marsupialization is the treatment of choice with a low recurrence rate.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

References