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Atypical thyroglossal duct cyst with intra-laryngeal and para-glottic extension

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A B S T R A C T

Thyroglossal duct cysts (TDC) are the most common congenital neck masses. Although they are anatomically closely related to the larynx, intra-laryngeal extension is very rare. We present a case, review the literature and discuss the challenges of intra-laryngeal TDC. A 55-year-old man presented with a neck mass associated with dysphagia. Computer Tomography neck scan showed a midline cyst extending to the pre-epiglottic space with partial obliteration of the right pyriform sinus and narrowing of the larynx. The cyst was excised en-bloc via Sistrunk procedure. Intra-laryngeal TDC are surgically challenging due to risk of perforation into the aerodigestive tract.

1. Introduction

Thyroglossal duct cysts are the most common congenital neck masses, accounting for 70% of congenital neck abnormalities and 40% of primary neck masses [1,2]. They most commonly present as a painless anterior midline neck mass in the 2nd–3rd decade of life. They arise due to failure of involution of the thyroglossal duct. During embryonic development, the thyroid anlarge migrates from the foramen caecum to its final pre-tracheal position via the epithelium-lined thyroglossal duct. The duct usually atrophies by the 10th week of gestation. However, portions of the duct may persist at any point between the foramen caecum and thyroid isthmus whereby inflammation, infection or mucous retention results in cystic expansion and the formation of a thyroglossal cyst. Hence, thyroglossal cysts may be located at the level of the hyoid (15–50%), supra-hyoid (20–25%) or most commonly infra-hyoid (25–65%) [2].

Although thyroglossal cysts are anatomically closely related to the larynx, intra-laryngeal extension is rare with only 18 reported cases over the last 30 years. We present the 19th case, review the literature and discuss the challenges of intra-laryngeal thyroglossal cysts.

2. Case report

A 55-year-old man presented with 5-year history of anterior neck mass which was significantly enlarging over the past year causing intermittent dysphagia. He did not have dyspnoea or hoarseness. Physical examination revealed a 4 × 5 cm anterior infra-hyoid neck mass that elevated on deglutition and tongue protrusion. Flexible nasopharyngoscopy under local anaesthesia in the office was normal. Computer Tomography (CT) scan of the neck showed a cystic multi-loculated septated 5.7 × 3 cm midline lesion splaying the thyroid lamina, extending posterior to the hyoid bone to the pre-epiglottic space, supraglottic region and right aryepiglottic fold with partial obliteration of the right pyriform sinus and narrowing of the larynx (Figs. 1 and 2). There was no thyroid or cricoid cartilage erosion. The thyroid gland was visualised and was separate from the lesion.

Intra-operatively, a multi-lobulated thyroglossal cyst was seen above the thyroid cartilage exerting mass effect on an intact thyrohyoid membrane causing the membrane to invaginate into the pre-epiglottic space (Fig. 3). The cyst was carefully dissected from the pyriform sinus mucosa without breaching the pharyngeal wall and excised en-bloc with the central portion of the hyoid bone via a Sistrunk procedure (Fig. 4). A tracheostomy was not required as the laryngeal structures were intact. Patient's post-operative recovery was uneventful and he could resume normal diet after the surgery. His neck drain was removed and he was discharged 4 days post-operatively. Histopathology showed a cyst lined by pseudostratified ciliated columnar and squamous epithelium containing scattered aggregates of benign thyroid follicles, confirming the diagnosis of thyroglossal cyst.

3. Discussion

A summary of the literature review findings is shown in Tables 1 and 2. Including our current case, there are only 19 such cases reported in the literature. Mean age at presentation was 40 years old (range 2–76 years old) and males were more commonly affected (94%). The cyst was usually located at the infra-hyoid region; but 4 patients did not have any neck mass clinically. Symptoms experienced by patients were

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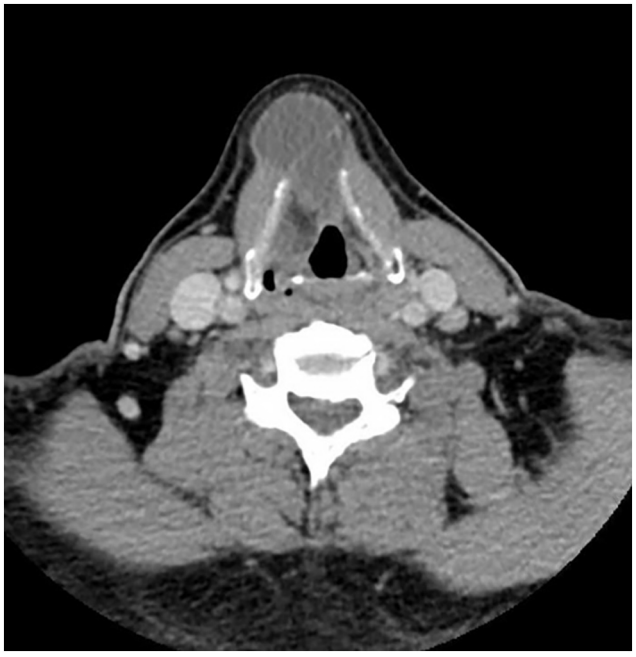

Fig. 1. Multi-loculated septated cyst splaying the thyroid lamina and extending into pre-epiglottic space.

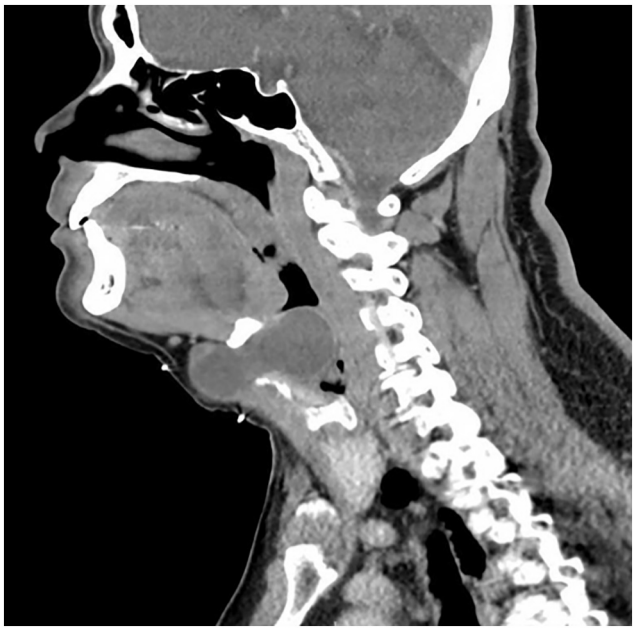

Fig. 2. Cyst causing partial obliteration of the right pyriform sinus and narrowing of the larynx.

hoarseness (8 cases), dyspnoea (5 cases), dysphagia (4 cases), stridor (2 cases) and foreign body sensation (1 case). Most intra-laryngeal extension were supraglottic. Laryngeal erosion included erosion of thyroid cartilage (4 cases), thyrohyoid membrane (2 cases), cricoid cartilage (2 cases) and cricothyroid membrane (1 case). Tracheostomy was done in patients with difficult intubation due to airway narrowing or cases whereby post-operative airway oedema was anticipated. Sistrunk procedure was performed in all cases and there was no reported recurrence.

Intra-laryngeal extension may be due to massive enlargement of the cyst over a long period of time, weakness over the laryngeal structures or malignant transformation of thyroid tissue within the cyst. Cases

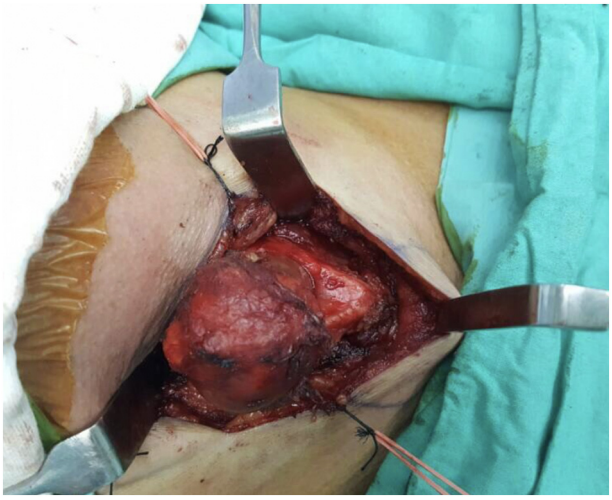

Fig. 3. Multi-lobulated thyroglossal cyst above the thyroid cartilage notch.

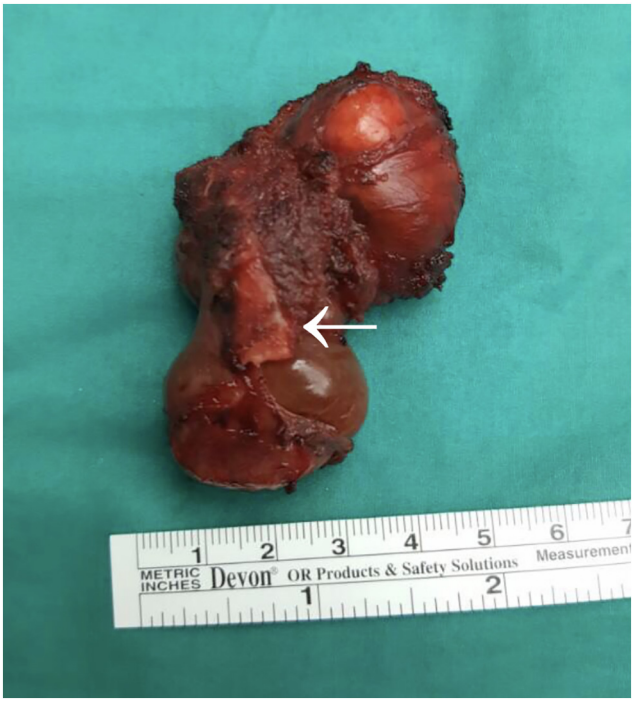

Fig. 4. "Dumbbell-shaped" thyroglossal cyst attached to hyoid bone (arrow).

arising from longstanding enlargement typically present as "dumbbell-shaped" lesions, whereby the cyst expands on both sides of the hyoid bone and pushes through an intact thyrohyoid membrane into the pre-epiglottic space [10]. This was evident in our patient, as seen from the "dumbbell-shaped" resected cyst (Fig. 4).

Hoarseness, dysphagia or dyspnoea in a patient with a neck mass that is clinically a thyroglossal cyst should raise suspicion of intra-laryngeal extension. Flexible nasopharyngoscopy and CT or Magnetic Resonance Imaging (MRI) neck should be performed to evaluate airway, assess the nature and extent of the cyst. They pose a diagnostic challenge as laryngeal cysts and neoplasms have similar presentations; a cystic lesion with normal mucosa within the paraglottic space is suggestive of laryngocele or saccular cyst. This has implications on surgical management as saccular cysts may be excised via endolaryngeal approach, which should not be performed in intra-laryngeal thyroglossal cysts as the end-on view of the laryngoscope may make dissection difficult and may not be able to provide a distinct plane

Table 1
Clinical presentation of patients in literature review.

Case	Author	Age	Gender	Neck mass, duration	Location	Symptom(s), duration	Abnormal nasoendoscopy
1	Slotnick et al. [3]	33	M	(+) 3 months	Infrahyoid	NP	(+)
2	Slotnick et al. [3]	42	M	(+) 6 months	Infrahyoid	NP	(+)
3	Slotnick et al. [3]	59	M	(+) 15 years	Infrahyoid	(+) 3 years	(+)
4	Cumberworth and Bradley [4]	45	M	(+) NP	Infrahyoid	(+) 4 months	(+)
5	Shaari et al. [5]	44	M	(+) 10 years	Infrahyoid	(+) 1 year	(+)
6	Mokhtari et al. [6]	41	M	(−)	Infrahyoid	(+) 6–8 months	(+)
7	Lübben et al. [7]	62	M	(+) 5 months	Suprahyoid	(+) 6 months	(+)
8	Quah and Lim [8]	42	M	(+) 1 year	Infrahyoid	(+) 1 year	NP
9	Loh et al. [9]	42	M	(−)	Infrahyoid	(+) 1 month	(+)
10	Soliman and Lee [10]	76	F	(+) 1 year	Infrahyoid	(+) 1 year	(+)
11	Sari et al. [11]	2	NP	(−)	Infrahyoid	(+) 1 year	(+)
12	Nicollas et al. [12]	2	M	(+) NP	Infrahyoid	(+) 1 month	(+)
13	Keleş et al. [13]	60	M	(+) NP	Infrahyoid	NP	(+)
14	Karlatti et al. [14]	28	M	(+) 2 months	Infrahyoid	NP	NP
15	Bando et al. [15]	50	M	(−)	Infrahyoid	(+) 1 month	(+)
16	Verma et al. [16]	27	M	(+) 1 month	Infrahyoid	(−)	NP
17	Aslıer et al. [17]	22	M	(+) 1 year	Infrahyoid	(−)	(−)
18	Touati and Ammar [18]	31	M	(+) 2 months	Infrahyoid	NP	NP
19	Our case 2017	55	M	(+) 5 years	Infrahyoid	(+) 1 year	(−)

NP – not provided in case report.

Table 2
Imaging, surgical findings and clinical outcome of patients in literature review.

Case	Author	Intralaryngeal extension	Laryngeal erosion	Tracheostomy	Recurrence, follow-up period
1	Slotnick et al. [3]	Supraglottic	(−)	(−)	NP
2	Slotnick et al. [3]	Supraglottic	(−)	(−)	NP
3	Slotnick et al. [3]	Supraglottic	(+)	(+)	NP
4	Cumberworth and Bradley [4]	Supraglottic	(−)	(−)	(−) 2 years
5	Shaari et al. [5]	Supraglottic	(+)	(−)	NP
6	Mokhtari et al. [6]	Supraglottic	(+)	(+)	NP
7	Lübben et al. [7]	Supraglottic	(−)	(+)	NP
8	Quah and Lim [8]	Supraglottic	(−)	(+)	NP
9	Loh et al. [9]	Supraglottic	(−)	(−)	NP
10	Soliman and Lee [10]	Supraglottic	(−)	(−)	(−) > 1 year
11	Sari et al. [11]	Subglottic	(+)	(−)	(−) 1 year
12	Nicollas et al. [12]	Subglottic	(+)	NP	NP
13	Keleş et al. [13]	Supraglottic	(+)	NP	(−) 1 year
14	Karlatti et al. [14]	Supraglottic	(+)	NP	NP
15	Bando et al. [15]	Supraglottic	(−)	(+)	(−) 3 years
16	Verma et al. [16]	Supraglottic	(−)	(−)	(−) 6 months
17	Aslıer et al. [17]	Supraglottic	(−)	(−)	(−) 6 months
18	Touati and Ammar [18]	Supraglottic	(+)	NP	(−) 6 months
19	Our case 2017	Supraglottic	(−)	(−)	(−) 2 years

NP – not provided in case report.

of the cyst [9]. Sistrunk procedure should be done instead to minimize risk of recurrence.

Intra-laryngeal thyroglossal cysts are also challenging to excise surgically as there is usually only a thin layer of mucosa between the cyst and airway or pharynx. Dissection has to be done carefully to avoid incomplete excision or entry into the aerodigestive tract.

4. Conclusion

Intra-laryngeal extension of thyroglossal cysts may result from massive enlargement of the cyst over a long period of time, weakness over the laryngeal structures or malignant transformation. They are a major concern as they may encroach on the larynx and pyriform sinus resulting in aerodigestive tract compromise. They may also pose a diagnostic challenge whereby laryngeal cysts and neoplasms are possible differentials. Surgery can be challenging due to the risk of perforation into the aerodigestive tract.

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