Unusual association of diseases/symptoms

A rare case of a concurrent large thyroglossal duct cyst with a base of tongue haemangioma

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Summary

An 85-year-old lady presented with a large midline neck mass. After 8 years of steady growth, the previously asymptomatic mass began to cause stridor and dysphagia. The patient's comorbidities included a previous partial glossectomy for haemangioma of the tongue, chronic obstructive pulmonary disease, congestive cardiac failure and obesity (body mass index >30). CT neck revealed the midline mass was cystic in nature, most likely a thyroglossal duct cyst. This mass was closely related to an angiomatous malformation involving the tongue, floor of mouth and left parotid. Fine needle aspiration cytology was consistent with a colloid goitre characterised as Thy-1. Due to her extensive comorbidities, surgical resection of the midline mass was deemed to be a high-risk procedure. A Sistrunk's procedure was performed. Dissection proved difficult due to the intimately related base of tongue haemangioma. Histopathology confirmed it to be a benign thyroglossal duct cyst. She made an uncomplicated postoperative recovery.

BACKGROUND

► This case report demonstrates an extremely rare association of a huge thyroglossal duct cyst (TGDC) and a base of tongue haemangioma. To our knowledge, this is the first reported case of TGDC in the literature associated with a tongue base haemangioma. The management of the case was both challenging and unusual due to significant patient comorbidities, and the complex anatomical relationships of the dual pathologies in the neck.

CASE PRESENTATION

An 85-year-old lady, who had undergone a partial glossectomy in 2001 for a haemangioma of the tongue, first presented to clinic in July 2009 with a midline neck mass. It had been enlarging over a period of 8 years, during which time she remained asymptomatic. She was, however, found to be hypothyroid and commenced on levothyroxine 75 µg once a day.

Medical history included chronic obstructive airway disease, hypothyroidism, obesity (body mass index >30), previous congestive cardiac failure, previous partial glossectomy for a haemangioma of the tongue.

This midline neck mass increased progressively in size and a clinical diagnosis of a thyroglossal cyst was made. Due to the patient's comorbidities, the mass, presumed a thyroglossal cyst, had been managed by 4 monthly aspirations in clinic over a year. Despite the increasing size, it was not until March 2010 when she began to develop symptoms of dysphagia and stridor especially worse at night. At this point, the possibility of surgical resection was explored.

On examination, there was a large midline neck mass measuring 10 × 8 cm in the widest dimensions which was firm, non-tender, mobile, not attached to overlying skin and no skin changes. The thyroid gland was indistinguishable

from the mass on palpation with no retrosternal extension. There was a separate palpable haemangioma in the submental region with overlying bruising of the skin which was non-tender, non-pulsatile and clinical in continuity with the midline cystic mass at the level of the hyoid bone (figure 1).

There was no palpable cervical lymphadenopathy.

The oropharynx showed evidence of the previous partial glossectomy with no pharyngeal oedema. Flexible nasendoscopy revealed a narrow airway consistent with her symptoms.

INVESTIGATIONS

Fine needle aspiration of the mass revealed abundant colloid and macrophages, and there were no epithelial cells present. It was consistent with a colloid goitre, characterised as Thy 1.

CT scan with contrast showed an angiomatous malformation involving the tongue, floor of mouth and left parotid. The midline mass in the neck measured 8.2 cm in the transverse diameter and 7.2 cm antero-posterior diameter with a 6 cm cephalo-caudate extension (figure 2). It appeared cystic, most likely to be a thyroglossal cyst closely related to the angiomatous malformation (figure 3).

DIFFERENTIAL DIAGNOSIS

Differential diagnoses include

- Thyroglossal duct: cancer, TGDC
- Thyroid: an ectopic thyroid, thyroid cancer, lingual thyroid, thyroid chondroma
- Lymphadenopathy: lymphoma, metastatic cancers of the upper aero-digestive tract
- Infective causes: tuberculosis, abscesses
- Lymphatics: cystic hygroma, lymphangioma
- Cystic: epidermoid cyst, dermoid cyst
- Thymus: enlarged thymus

Figure 1 Preoperative view of the large thyroglossal cyst and skin changes overlying base of tongue haemangioma.

- Vascular: vascular malformations of the great neck vessels, haemangioma
- Muscular: involvement of sternocleidomastoid, strap muscles.

TREATMENT

Preoperative

After a detailed anaesthetic review and patient consent, it was decided that this was a feasible yet high-risk procedure

to undertake a surgical resection. The patient was aware of the risks which were mainly difficult intubation, bleeding, injury to nerves (superior and recurrent laryngeal), vocal cord palsy and the specific anaesthetic risks relating to her high body mass index and comorbidities.

Relevant blood tests including full blood count, electrolytes and urea, thyroid function tests, clotting profile, group and save. We ensured the patient was euthyroid preoperatively.

Figure 2 CT scan of the neck showing a large midline thyroglossal duct cyst.

Intraoperative

The procedure was done under general anaesthesia with the patient supine and neck hyperextended.

A horizontal midline incision was made along a neck crease and dissection of tissues after raising subplatysmal skin flaps. The huge cystic mass was mobilised, decompressed and a Sistrunk's procedure performed (figure 4). This involved removal of the TGDC with the middle third of the hyoid bone.

This was a difficult dissection as, superiorly, the cystic mass abutted the remaining base of tongue haemangioma. Extreme care was taken to avoid the vascular tissue. Due to the size of the cyst removed, excision of redundant skin was performed (neck lift), two drains were left in situ and the wound closed with clips.

Postoperative

She made an uneventful recovery with no complications.

OUTCOME AND FOLLOW-UP

Histology

Macroscopy

Deflated cystic structure was measuring $11 \times 4 \times 3$ cm and weighing 75 g. This appeared unilocular with a corrugated internal surface. A strip of bone (hyoid) was present along one edge.

Microscopy

Widespread sampling confirmed a unilocular cyst, partially lined by ciliated, respiratory-type glandular epithelium

interspersed with a vigorous cholesterol granulomatous response. Mantling this was a zone of laminated, organising fibrovascular granulation tissue with an occasional congerie of colloid-containing thyroid follicles.

There was no suppuration, no discrete granulomatous inflammation or giant cell transformation.

Local excision was complete with no evidence of malignant transformation.

Conclusion

Sistrunk's procedure – benign thyroglossal duct cyst.

She was reviewed in the out-patient clinic with a satisfactory outcome and good wound healing. There were no neurovascular deficits and there has been no recurrence.

DISCUSSION

TGDC

The thyroid gland forms as a diverticulum from the floor of the pharynx (tuberculum impar) at a site that later becomes the foramen caecum of the base of the tongue. The thyroid gland then descends along the thyroglossal duct in the midline of the neck. This duct normally involutes; however, when involution does not occur this increases the potential for development of a TGDC.¹

TGDCs may be present in any part of the thyroglossal tract commonly beneath the hyoid, in the region of the thyroid cartilage and above the hyoid bone – usually painless neck swellings in the midline which move upwards on protrusion of the tongue as well as on swallowing (because of the attachment of the tract to the foramen caecum).²

Figure 3 CT scan of the neck showing large thyroglossal duct cyst in close relation to the angiomatous malformation at the base of the tongue.

Figure 4 Large thyroglossal duct cyst excision using Sistrunk's procedure.

TGDCs occur with equal frequency in males and females and are the most common mass found in the midline of the neck in children.¹ TGDCs have been well reported in the literature in several case series.^{3–9}

Manifestations

TGDCs usually present clinically as congenital cysts, infected cysts or sinuses. Less commonly reported manifestations in the literature include acute airway obstruction,^{10–11} carcinoma (usually papillary),^{12–22} misdiagnosed as laryngomalacia in children²³ and following radiation therapy to the neck.²⁴ Atypical presentations also include the floor of the mouth,²⁵ intralaryngeal extension,^{26–27} as a thyroid nodule in the lateral neck,²⁸ cutaneous extrusion,²⁹ cutaneous blisters,³⁰ as a retro/pharyngeal abscess³¹ and in the mediastinum.³²

Known concurrent associations

To our knowledge, this is the first reported case of a concurrent huge TGDC with a base of tongue haemangioma in the literature. Known concurrent associations with TGDCs in the literature include sublingual/lingual thyroid,^{33–34} epidermoid cyst,³⁵ thyroid carcinoma,^{36–37} fistula,⁵ inferior parathyroid cyst³⁸ and lingual tonsil hypertrophy.³⁹

Tongue haemangioma

Vascular malformations comprise a significant portion of angiodyplastic lesions in the head and neck but haemangioma of the base of tongue is extremely rare.^{40–42} Depending on the size and symptoms caused, management could be conservative, medical therapy or surgical excision. Medical therapy includes use of steroids (systemic or intralesional), β -blockers and injection of sclerosing agents.⁴³ Surgical care includes embolotherapy, laser therapy, cryosurgery and excision. In this case, a partial glossectomy was employed.

Management

Sistrunk's procedure has been recognised as the surgical treatment of choice for TGDCs particularly as it significantly reduces the risk of recurrence.⁴⁴ It involves excision of the thyroglossal duct tract and the middle third of the hyoid bone. Recognised complications include neurovascular injury, surgical emphysema and airway injury.⁴⁵ Sistrunk's was performed in this case for a huge TGDC with a satisfactory outcome albeit with complex anatomical relationships to a tongue base haemangioma.

Stealth surgery has also been described (for lesions not as huge as in this case report) for neck masses.⁴⁶ Other

described options include endoscopic carbon dioxide laser procedure⁴⁷ and percutaneous ethanol injection.⁴⁸

Competing interests None.

Patient consent Obtained.

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Learning points

- This is a very rare association of a huge thyroglossal duct cyst with a base of tongue haemangioma.
- This case report demonstrates a satisfactory outcome following a Sistrunk's procedure despite a complex anatomical association in the neck.
- In similar dual neck pathologies, especially those with vascular associations, extreme care should be taken to prevent intraoperative complications.

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McNamara K, Olaleye O, Smith J, Karamchandani D, Watkinson J. A rare case of a concurrent large thyroglossal duct cyst with a base of tongue haemangioma. *BMJ Case Reports* 2011;10.1136/bcr.10.2010.3393, date of publication

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