

*Case Report***Double Thyroglossal Duct Cyst Derived from a Single Tract:
A Rare Presentation***Ehsan Khadivi¹, Hossein Payedar Ardekani²**Abstract****Introduction:**

Thyroglossal duct cysts are one of the most common congenital pathologic findings in children's cervical area. This type of cyst can be located anywhere between the base of the tongue and the sternal manubrium.

Case Report:

We report the case of a patient with a double thyroglossal cyst, located inferior to the hyoid bone. The 2 cysts were connected by a common permeable tract, which confirms that an evolution failure of the embryonic remnants of the thyroglossal duct has been responsible for the development of such a cyst. Our case is the second case of double thyroglossal duct cysts and the first of double cysts located in the neck outside the thyroid gland.

Keywords:

Congenital, Mass, Thyroglossal duct cyst

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Introduction

Thyroglossal duct cysts comprise approximately one-third of congenital neck masses in children (1).

Most thyroglossal duct cysts are seen in the midline and below the level of the hyoid bone; this is because the tract passes through the hyoid bone. Some cysts may present laterally and are not infrequently found superior to the hyoid bone or even as low as the level of the thyroid gland. Other unusual presentations of thyroglossal duct cysts include formation on either side of the hyoid bone as dumbbell-shaped lesions and rarely, as cystic lesions in the larynx (2).

Reviewing the literature, we found only one report of double cysts derived from one thyroglossal duct, whereas one of them was in the hyoid region and the other in the thyroid gland (3).

Case report

We report a healthy 14-year-old female patient with history of 1 year complaint of an anterior neck mass at the level of the hyoid bone. The mass has varied in size during this period due to upper respiratory infections. On physical examination we palpated two separate anterior neck masses below the hyoid bone, their size being 2.5×1.5 and 2×1.5 cm; the mass was cystic, non tender and mobile with touch and tongue movements. A contrast enhanced neck CT scan also showed two masses near the hyoid bone (Fig1).

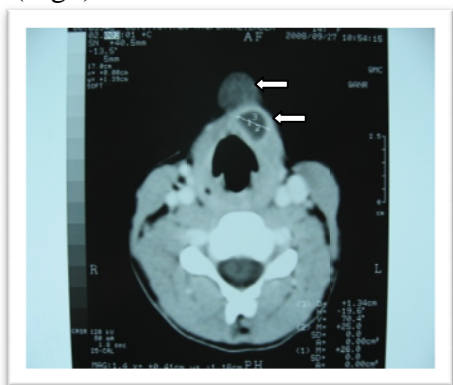


Fig 1: CT scan with contrast shows two separate cystic lesions, one is more superficial than the other (white arrows).

The patient underwent a transcervical approach with a horizontal midline incision above the mass in a skin crease; two cysts were dissected from the adjacent soft tissue. They were two cysts with a common duct passing through the hyoid bone (Fig 2). A standard Sistrunk operation was performed and the specimen was sent for pathological examination.



Fig 2: Surgery specimen. Two inferahyoid cysts with joined tracts. The hyoid bone can be seen.

The pathologist reported two cystic parts covered with ciliary pseudostratified cylindrical epithelium with regions of ulceration and infiltration of inflammatory polymorphic cells and plenty of foamy macrophages plus fibrosis and hemorrhage in the cyst wall, compatible with double thyroglossal cysts.

Discussion

Thyroglossal duct cysts and dermoids are the most common midline lesions of the neck in children (4). The thyroglossal duct cyst derives from the remainings of the embryonic thyroglossal duct, anywhere between the foramen cecum to the thyroid gland (5). Two third of thyroglossal duct anomalies are diagnosed within the first 3 decades of life, with more than half being identified before the age of 10. The most common presentation is a painless cystic neck mass on the midline near the hyoid bone. Although they are most commonly found immediately adjacent to the hyoid bone (66%), they can also be located between the tongue and hyoid, between the hyoid and the pyramidal lobe, within the tongue, or even within the thyroid (6).

Thyroglossal duct remnants often involve the central part of the hyoid, necessitating removal of the midportion of the hyoid to minimize the risk of recurrence. Surgical excision is the treatment of choice (5).

Al-Khatib et al in a 12-year study in Jordan, reported a total of 2063 neck mass lesions; of these, 252 (12%) were congenital masses. These cases were distributed as 166 (66%) midline, 55 (22%) lateral and 31 (12%) entire neck masses. The most frequent mass was the thyroglossal duct cyst (fistulas) (53%), followed by cysts (fistulas) of the branchial apparatus (22%), dermoid cysts (11%), hemangiomas (7%), and lymphangiomas (6%). The majority of branchial arch anomalies (85%) were of the second arch. The mean age of patients was 16 years, with the greatest number of cases (38%) in the first decade of life. The male-to-female ratio was 1:1.2, with most lesions affecting females (7).

Lin et al compared thyroglossal duct cysts between adults and children in a total of 84 patients (32 children, 52 adults) and reported that no significant difference in sex was observed in either group. In comparison to children, left-sided and infrahyoid cyst locations were more prevalent in adult patients. The cyst sizes were also significantly larger in adults (8).

In the study of 62 thyroglossal cysts by Sathish et al atypical features were reported in 5/62 cases. 2/5 were adolescents with a short history whereas 3 were below the age of 5 with an onset since infancy; 4/5 were females; 1/5 had had a Sistrunk operation earlier and 2/5 had had a redo surgery prior

to cure. 3/5 had grossly identifiable tracts during surgery, one with a midline course and two deviating laterally and opening into the left pharyngeal wall. The final histological diagnosis was a mixed thyroglossal-dermoid cyst, thyroglossal duct anomalies with aberrant pharyngeal communication, isthmus thyroglossal 'cold' cyst and a tuberculous cyst (9).

Other rare reported presentations and locations are thyroglossal cyst within the mediastinum (10), breath-holding-like spells in an infant with lingual thyroglossal duct cyst and intralaryngeal extension (11,12).

Our case is the second report of two thyroglossal cysts connected by a single tract. In the study of Pueyo et al, double thyroglossal duct cysts in one patient is discussed in whom one cyst is located in the hyoid region and the other in the thyroid gland (3).

The two cysts in our patient were connected by a permeable tract which confirms that an evolution failure in the embryonic remains of the thyroglossal duct has resulted in the cyst formation.

Although our case is the second report of a double thyroglossal duct cyst, but due to the specific location of the cysts, both below the hyoid bone and outside the thyroid, it differs from the other reported cases.

Conclusion

In spite of the fact that double thyroglossal cyst is not a common form of presentation, its consideration in the differential diagnosis of neck masses is highly recommended.

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