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CASE REPORT

Unusual coexisting thyroglossal duct cyst and second branchial cleft fistula in an adult☆

Coexistência incomum de cisto do ducto tireoglosso e fístula da segunda fenda branquial em adulto

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Introduction

Thyroglossal duct cysts are the most common congenital neck masses, followed by branchial cleft anomalies.[1] Thyroglossal duct cysts are three times more prevalent than branchial cleft anomalies.[2] However, to our knowledge, coexisting thyroglossal duct cyst and branchial cleft anomaly has been reported only once in the literature.[3] Herein, we present the second case report of coexisting thyroglossal duct cyst and second branchial cleft fistula in a 34 year-old male.

Case report

A 34 year-old male presented with recurrent anterior and lateral neck mucoid discharge since childhood. The patient reported no relevant medical history except for a bilateral tonsillectomy 25 years ago. The physical examination revealed two external openings on the patient's neck. One was located at the hyoid level; the other was located along the anterior border of the lower third of the sternocleidomastoid muscle (SCM). No palpable cervical mass or subcutaneous tract, and no active inflammation were present around the lesions. Computed Tomography (CT) scan of the neck revealed an 8.2 cm elongated rim-enhancing cystic lesion along the left anterior neck, extending from the anterior border of the lower third of the SCM to the tonsillar fossa (Fig. 1). CT scan showed no fistulous track or cyst at the hyoid level, and no connection between the two external openings. The pharyngoesophagogram showed no leakage from the pharynx.

Based on these observations, the patient was diagnosed with a left second branchial cleft fistula and a likely thyroglossal duct cyst. As the first surgical step for the branchial cleft fistula, a transverse elliptical incision was made around the external opening. With gentle traction on the fistula, the dissection proceeded extending to its cephalic portion. After a stepladder incision, the fistulous tract was dissected to the tonsillar fossa, where it was ligated and separated (Fig. 2). We performed the Sistrunk's operation for the fistula at the hyoid level under the working diagnosis of a thyroglossal duct cyst (Fig. 2). Intraoperative pharyngoscopy revealed no pharyngeal opening. Pathological examination of the lesions revealed thyroglossal duct cyst and branchial cleft fistula. The patient's postoperative course was uneventful. At 14 months follow-up, there has been no recurrence, and the patient remains asymptomatic.

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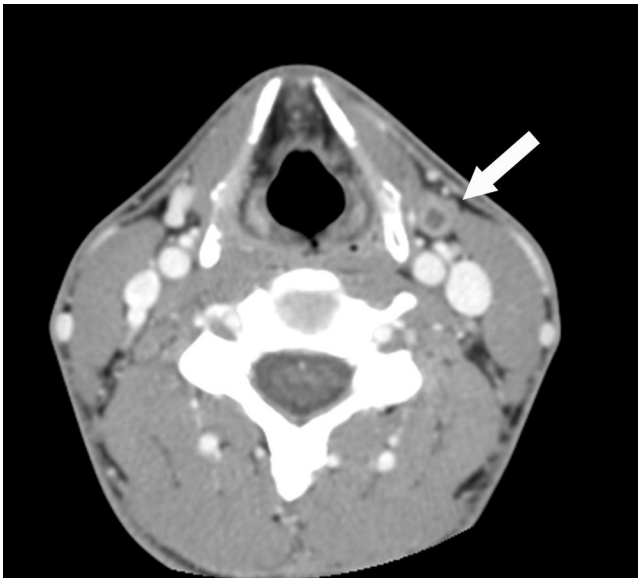

Figure 1 Axial computed tomography scan of the neck shows an 8.2 cm elongated rim-enhancing cystic lesion (arrow) along the left anterior neck.

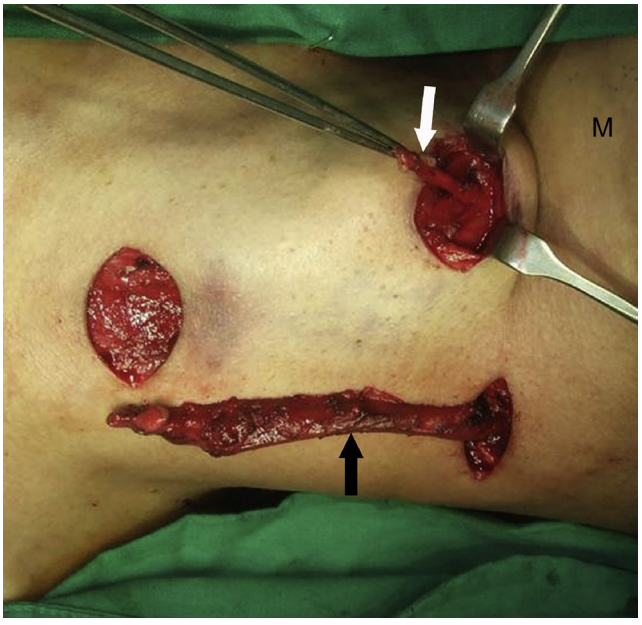

Figure 2 Intraoperative finding shows a dissected left second branchial cleft fistula (black arrow) by stepladder incisions, and a thyroglossal duct cyst (white arrow) with attached hyoid bone. M, mandible.

Discussion

Thyroglossal duct cysts are the most common cause of congenital neck masses.[1] They usually present as midline neck masses and can be found at any point between the foramen cecum and suprasternal notch.[4] They are painless, enlarge slowly, and may be associated with a fistula or sinus tract.[4] In this case, thyroglossal duct cyst presented as a fistula. Branchial cleft anomalies can present as cysts, sinuses, cartilaginous remnants, or fistula; they account for approximately 30% of all congenital head and neck lesions, with the majority being second brachial cleft anomalies.[1,2,5] Overall, cystic lesions are more common than fistulae in patients with second branchial cleft anomalies.[6] The current patient presented with a fistula with a small external opening along the anterior border of the lower SCM.

Ultrasound, CT, or, magnetic resonance imaging (MRI) may be helpful to evaluate both thyroglossal duct cysts and branchial cleft anomalies.[1,4] The definitive treatment of thyroglossal duct cysts and branchial cleft anomalies is complete surgical excision of the entire tract. After diagnostic CT scan in this case, the authors performed surgical excision of the entire tract by the Sistrunk's operation and stepladder incision.

There were some unique characteristics to this case. Firstly, this was the second reported case of unusual coexisting thyroglossal duct cyst and second branchial cleft fistula in an adult.[3] This finding highlights that thyroglossal duct cysts and branchial cleft fistula are encountered in adults. Therefore, coexisting thyroglossal duct cysts and branchial cleft fistula should be considered in the differential diagnosis of neck lesions, even in adults. Secondly, the patient did not present with a history of infection and previous surgical procedure for congenital neck lesions. There is a possibility of an asymptomatic thyroglossal duct cyst and branchial cleft fistula occurrence without palpable neck lesions in adults. Thirdly, if thyroglossal duct cyst is suspected in an adult, the Sistrunk's operation, removing the central portion of the hyoid, is the optimal choice of therapy not only for aesthetic reasons, but also for management of recurrent infections and the potential danger of malignancy.[1,6]

Conclusion

Coexisting thyroglossal duct cysts and branchial cleft fistula should be considered in the differential diagnosis of neck lesions, even in an adult.

Conflicts of interest

The authors declare no conflicts of interest.

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