

Complete second branchial cleft anomaly presenting as a fistula and a tonsillar cyst: An interesting congenital anomaly

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Abstract

Branchial cleft anomalies make up 30% of all pediatric neck masses, but complete second branchial cleft anomalies are extremely rare. We report an unusual case of a complete second branchial cleft anomaly that presented as a draining neck fistula and a tonsillar cyst in an otherwise healthy 3-month-old girl. At the age of 7 months, the patient had been experiencing feeding difficulties, and there was increasing concern about the risk of persistent infections. At that point, the anomaly was excised in its entirety. Our suspicion that the patient had a complete second branchial cleft anomaly was confirmed by imaging, surgical excision, and histopathologic analysis.

Introduction

Branchial cleft anomalies account for approximately 30% of all pediatric neck pathologies.¹ They are often described as either fistulas, cystic masses, or sinus deformities, depending on the presentation. Approximately 95% of these anomalies are derived from the second branchial arch and are incomplete; they often present within the first 2 decades of life.² Complaints of intermittent mucoid drainage, recurrent inflammatory cellulitis, and/or abscess formation along the anterior border of the sternocleidomastoid muscle are common in patients with second branchial cleft anomalies.³ Evidence in the literature supports upper respiratory tract infections as the exacerbating cause of such conditions in most cases.

The embryologic migration pattern of the second branchial arch tract originates in the tonsillar fossa, travels over third arch structures, and terminates in the middle to lower two-thirds of the lateral portion of the neck.⁴ Second branchial cleft anomalies are often described as remnants along this migration pathway; they rarely present as a complete tract. Furthermore, there is very limited literature describing these anomalies presenting as a tonsillar cyst with a complete fistulous tract.

In this article, we report an unusual case of a complete second branchial cleft anomaly that presented as a draining neck fistula and a tonsillar cyst.

Case report

A 3-month-old girl with an uncomplicated birth history presented with a 3-week history of a persistent green malodorous drainage from a small external opening on the right side of her neck. Her mother originally assumed that the moisture represented either saliva or milk residue. She reported no recent sick contacts, feeding difficulties, or respiratory problems.

Physical examination revealed an area of erythema surrounding an actively draining pinpoint opening at the anterior border of the lower third of the right sternocleidomastoid muscle. On examination of the oral cavity, a well-defined cystic mass was discovered at the superior aspect of the right tonsillar fossa. Computed tomography demonstrated an elliptical cyst-like mass at the level of the soft palate, with a fistulous tract traveling anterior to the carotid arteries and opening anteriorly to the right sternocleidomastoid muscle.

The patient was treated with antibiotics for 2 weeks and scheduled for surgical excision of a second branchial cleft fistula and tonsillar cyst, which would be performed

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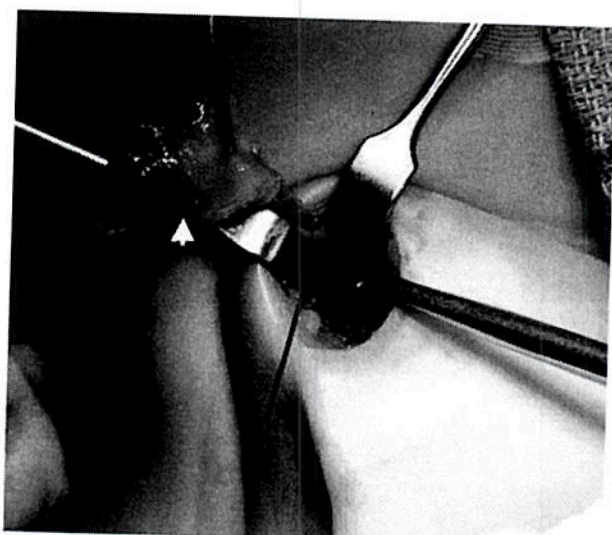


Figure 1. Intraoperative photo shows the dissection of the right branchial anomaly and fistulous tract. The tan rubbery structure (arrowhead) and the fistulous tract (arrow) are seen.

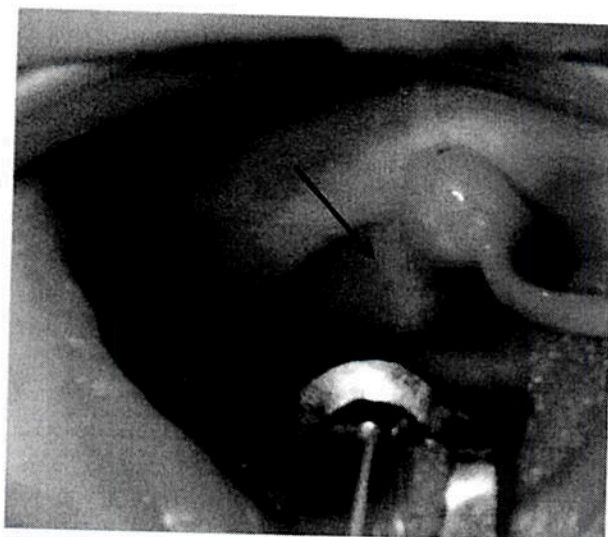


Figure 2. Photo shows the right tonsillar cyst (arrow) in situ.

once she reached the age of 10 months. However, 4 months after resolution of the acute infection, the now 7-month-old child began to experience feeding difficulties. Further investigation revealed that the tonsillar cyst had increased in size and was now encroaching on and deviating the uvula. Therefore, we decided to perform surgery the following week.

Intraoperatively, the right neck opening was marked, and a size 00 lacrimal probe was advanced into the fistulous tract to determine its orientation. A 3.0 × 1.5-cm elliptical incision was made through the platysma, around the fistula opening and the area of induration. Once the subplatysmal level was reached, we identified a tan, rubbery, 2.2 × 1.5-cm cystic structure that was encompassing the fistulous tract. This was bluntly dissected from the surrounding tissue boundaries and was traced superiorly and deep to the anteroinferior sternocleidomastoid muscle and anterolaterally to the carotid arteries (figure 1).

At the level of the hyoid bone, intraoral finger pressure was applied to the right tonsillar fossa to enable further dissection superiorly. The fistula was ligated with 2-0 silk sutures inferior to the tonsillar base. This

specimen was delivered from the neck incision.

The tonsillar cyst was dissected from the superior pole of the tonsillar fossa inferiorly between the tonsillar pillars and under the tonsillar tissue to the base of the fossa (figure 2). This specimen was then removed in fragments. The defect created at the base of the fossa was closed with 4-0 Vicryl. The fistula excision site was closed in an interrupted multilayer fashion, and a Penrose drain was placed under a compression dressing. No respiratory, feeding, or wound complications were observed postoperatively or at the 6-month follow-up visit.

Histopathologic examination of the branchial fistula and the tonsillar cyst revealed that they represented multiple large cystic spaces lined with ciliated columnar cells and mucous cells (figure 3). These findings were consistent with branchial remnant histology.

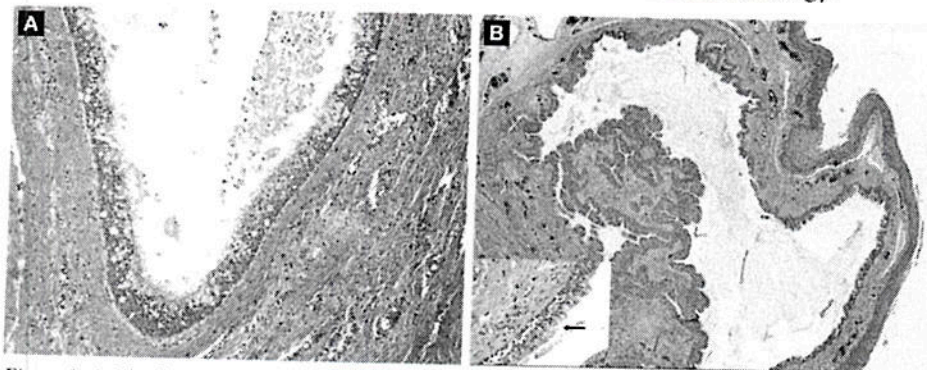


Figure 3. A: The fistulous tract is lined with respiratory epithelium. Note the dense fibrous wall (hematoxylin and eosin, original magnification ×100). B: The tonsillar cyst is lined with columnar epithelium focally and mucous cells (hematoxylin and eosin, ×40). Inset: The ciliated epithelium (arrow) is seen at higher magnification (×400).

Discussion

According to the literature, congenital branchial anomalies usually present in early childhood or adolescence, and they show no predisposition to either sex.¹ These abnormalities are formed from branchial remnants that persist through embryologic development. Histologically, they are lined with ciliated columnar epithelium, squamous epithelium, or both.¹

The course of a classic branchial cleft fistula is one that begins at the lower anterolateral two-thirds of the neck as an external opening. This tract runs deep to the platysma along the carotid sheath. It can be located laterally, deep to, or between the internal and external carotid arteries before they terminate in the supratoral fossa. Along this course it passes over the hypoglossal nerve and below the stylomandibular ligament.^{3,5}

Surgical intervention for these anomalies is usually performed only when a patient is symptomatic or to prevent recurrent infections.³ Enlargements of these anomalies can also result in respiratory compromise, torticollis, and/or dysphagia.¹ Recurrence rates for branchial fistulas after surgical intervention have been reported to range from 4 to 10%.⁶ For this reason, surgery should include a complete excision of the fistulous tract.

Our case illustrates the known developmental migration of second branchial arch anomalies while demonstrating an unusual finding of a complete branchial fistula presenting as an intraoral tonsillar cyst. It was with imaging, surgical exploration, and histopathologic confirmation that we determined that these two entities represented a single second branchial cleft anomaly. To the best of our knowledge, the literature on second branchial arch pathologies does not contain any previously reported case of a complete draining fistulous tract with a connection to a large superior pole tonsillar cyst.

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