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CASE REPORT

Complete congenital third branchial fistula on right side

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The incomplete branchial fistula is not an uncommon congenital anomaly of branchial apparatus, but a complete one is rare. Fistulas form when the mesenchyme separating the cleft and pouch involutes, and the cleft and pouch unite.

More than 90 percent of branchial cleft anomalies arise from the second branchial cleft system and 8 percent from the first branchial cleft system. Anomalies arising from the third and fourth branchial cleft system rarely occur. Third and fourth branchial pouch fistulas are collectively referred to as pyriform sinus fistulas. Branchial cleft sinuses with external openings are usually associated with the first and second branchial cleft arches, and those with internal openings are usually associated with the third and fourth arches.

Almost all cases reported were incomplete fistulas and were on the left side, which probably is due to the asymmetric development of branchial apparatus. However, here we report a case of complete congenital third branchial fistula on the right side with its management.

A 16-year-old male patient presented with a history of mucus discharge from the right side of the neck since birth, which increased on taking food. He has no family history of similar complaints. On examination, a 3-mm diameter opening at the junction of the middle and the lower one-third of the anterior border of the right sternocleidomastoid was found. The rest of the physical examination was unremarkable. A fistulogram with anterior and lateral views (Fig 1) clearly revealed the tract extending cranially and posteriorly, communicating with the hypopharynx.

Under general anesthesia, the external opening was probed carefully, and subsequently methylene blue dye was injected to delineate the tract, which spilled into the pyriform sinus and revealed the internal opening. A transverse elliptical incision was made around the external opening, and dissection continued carefully on either side of the tract, which was greatly facilitated by passing a probe (infant feeding tube) along the tract. The tract was found to run posterior to the common and internal carotid arteries. A continuous intact fistulous tract of 7 cm length was excised in toto (Fig 2).

The postoperative period was uneventful. Histopathology opined it as squamous cell epithelium. The patient had no recurrence on 3-year follow-up.

DISCUSSION

A typical lateral cervical fistula is present at birth. Males and females are equally affected and some fistulas occasionally appear to be hereditary. The sinuses usually present with recurrent episodes of neck abscess or acute suppurative thyroiditis, a perithyroid swelling, or a retropharyngeal abscess.

A fistula formed from the third branchial arch has its external opening in the same area as the second branchial fistula. The tract passes deep to the platysma, ascending along the common carotid sheath, passing behind the common and internal carotid artery. The tract crosses the hypoglossal nerve but will not ascend above the glossopharyngeal nerve or the stylopharyngeus muscle and is superficial to the superior laryngeal nerve. The internal opening is in the pyriform sinus.

Fourth branchial fistula also originates from the pyriform sinus, but in contrast they course inferior to the superior laryngeal nerve. Complete fourth branchial apparatus anomalies have never been conclusively demonstrated. Theoretically, the tract would loop around the right subclavian artery on the right or the aortic arch on the left into the mediastinum, and course superiorly to the upper esophagus.

The third pouch fistula drains anteriorly to the fold made by the internal laryngeal nerve; the fourth pouch fistula drains posterior to this fold. Other criteria that have been used to differentiate third from fourth pouch remnants are the location of the internal opening into the pyriform fossa (third at cephalad and fourth at apex, that is, caudal part of the fossa, or even in the proximal esophagus) and the presence of thymic tissue (third pouch) or thyroid tissue (fourth pouch).¹

Third and fourth branchial abnormalities have been reported in all age groups; they have also been diagnosed in utero. These abnormalities can be especially dangerous in neonates if they rapidly enlarge through a fistulous tract

during swallowing. Such an enlargement can lead to tracheal compression and airway compromise.¹

In cases of complete fistula, fistulogram or barium contrast study can delineate the course of the anomaly with clear sufficiency. Diagnostic accuracy is improved by using thin contrast material and by performing the test in the noninfected state, because edema may prevent contrast from entering the tract. Sensitivity of barium-swallow imaging is as high as 80 percent for diagnosing a pyriform sinus fistula, and some have recommended the use of carbonated beverages instead of barium for diagnosis.² Ultrasonography also can show gas within the area, which is pathognomonic of a pyriform sinus fistula.³ In difficult cases both CT and MR studies are advocated, but CT is the preferred method.⁴

Complete surgical excision of the tract is the treatment of choice. If the fistula tract is long, some would use stepladder incisions. But in our case plastic surgical skills allowed extirpation with single incision. Vertical skin incisions should be avoided because the resultant scar is cosmetically unappealing.

In the neonatal period, surgery can be postponed until 3 to 6 months of age. The delay allows the child to grow and ideally precedes a first upper respiratory infection. Recurrence rate after surgery is 3 percent, which increased to 21 percent with previous attempts of surgery and 14 percent with history of infection. Treatment by chemocauterization of the internal



Figure 1 Fistulogram, right lateral view.

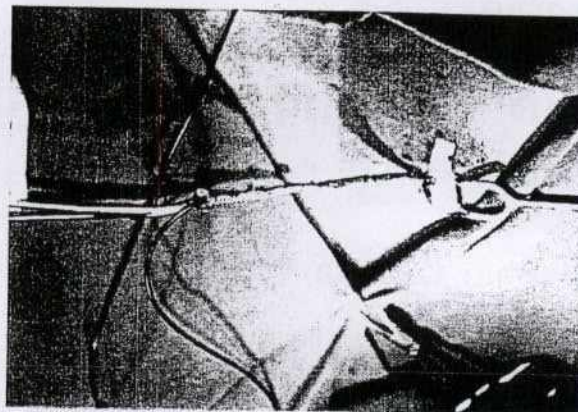


Figure 2 Perioperative photograph showing excision of complete tract.

opening has been reported with encouraging results,⁵ but long-term efficacy of this method is yet to be proven.

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FINANCIAL DISCLOSURE

None.

REFERENCES

1. Liberman M, Kay S, Emil S, et al. Ten years of experience with third and fourth branchial remnants. *J Pediatr Surg* 2002;37:685-90.
2. Bar-Ziv J. Pyogenic thyroiditis in children due to a fourth branchial pouch fistula. *Pediatr Radiol* 1996;26:88-90.
3. Elahi MM, Dubé P, Manoukian JJ, et al. Partial thyroidectomy and selective neck dissection in the management of pyriform sinus fistulae. *J Otolaryngol* 1997;26:57-63.
4. Park SW, Han MH, Sung MH, et al. Neck infection associated with pyriform sinus fistula: imaging findings. *Am J Neuroradiol* May 2000; 21:817-22.
5. Kim KH, Sung MW, Koh TY, et al. Pyriform sinus fistula: management with chemocauterization of the internal opening. *Ann Otol Rhinol Laryngol* 2000;109:452-6.