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Case Report

Management of a recurrent third branchial arch fistula using combined endoscopic and open approach

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ABSTRACT

Branchial cleft anomalies are rare, congenital lesions, that occur due to the maldevelopment of the branchial apparatus, during fetal maturation. Anomalies of the third and fourth arch contribute to a very small proportion of these. Most cases are detected in childhood, either as a cyst, sinus or fistula in the neck. Some of the common treatment approaches to third branchial cleft fistulas include endoscopic cauterization, open cervical fistulectomy and, in cases of recurrence, selective neck dissection. Here, we present a case of recurrent branchial fistula of the third arch, wherein we used a combined cervical and intraoral approach, conserving the thyroid cartilage, to achieve a complete excision of the fistula tract.

Keywords: Branchial arch fistula, Third arch fistula, Recurrent branchial fistula

INTRODUCTION

Branchial cleft anomalies are congenital lesions of the neck, which are a result of some defects in the branchial apparatus occurring during embryonic development. 17% of all neck swellings in children are branchial arch anomalies.[1] Second arch anomalies constitute the majority, with the third and fourth arch anomalies being extremely rare and are often subject to misdiagnosis and mismanagement. Third branchial arch fistulas present with an external opening in the mid-to lower part of the neck, and an internal opening, at the pyriform sinus. Management requires a comprehensive radiological workup to determine the exact course of the fistulous tract.

Complete surgical excision of the tract is required to prevent recurrences in the future. Recurrent fistulas are difficult to deal with and usually require a combined oral and cervical approach to tackle them.

CASE REPORT

A 27-year-old female, presented to plastic surgery outpatient department with complaints of salivary leak from an opening in the neck. The patient had a history of right sided neck swelling, noticed at the age of 7 years, for which she underwent surgery. The swelling recurred after 2 months, and the patient underwent a second surgery for the same. Soon after the second surgery, the patient noticed salivary leak from a small opening in the right side of neck.

On physical examination she had a surgical scar on the upper right side of the neck, laterally. A small fistulous opening was seen at the medial end of the scar, through which salivary leak could be appreciated (Figure 1).

A computed tomography sinogram scan (CT sinogram) of the neck was carried out but it was not able to accurately trace the tract. Our radiologist tried again with an oral contrast, and this time we were able to identify a fistulous

tract with external opening in the right upper neck, anterior to sternocleidomastoid muscle. The tract traverses inferiorly towards the lower border of the thyroid cartilage, with a possible extension of the tract superiorly along the lateral wall of larynx into the pyriform fossa (Figure 2).

Figure 1: Clinical photograph of the patient showing a horizontal scar on the right side of neck with a dimpling at the medial end marking the site of leakage of saliva.

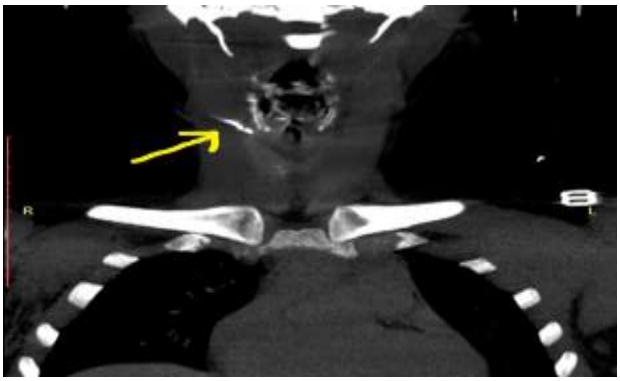

Figure 2: Computed tomography scan (coronal section) of the neck, post oral contrast, showing the sinus tract extending from the lower border of the ala of thyroid cartilage to the external opening on the neck.

We were called in to be part of the surgical team, as the tract was going medial to the thyroid lamina into the pyriform fossa. Patient was taken up for a combined approach (cervical and intra-oral) excision of the fistula.

Using Kleinsasser laryngoscope, a dimpling of the mucosa was noted on the lateral wall of the right pyriform fossa, indicating the opening of the fistulous tract. We used a 5 FR (French gauge) infant feeding tube with a metallic guidewire inserted inside so that it could be used to dilate the opening of the tract, as well as guide the tube into to the tract to the maximum extent possible. A cervical skin incision was given along the existing surgical scar. The external opening of the fistulous tract was carefully dissected circumferentially and followed medially (Figure 3). Cricothyroid muscle had to be cut and the tract was followed superiorly by retracting the inferior border of thyroid ala. The tract was noted to be reaching up to the cricoid cartilage and then turning upwards to enter the pyriform fossa just behind the inferior horn of thyroid cartilage. The tract was traced upto the stent and superiorly, the intra-oral stent in the tract was reached from the external incision. The tract was delineated along its entire length, ligated with transfixion sutures applied close to the pyriform mucosa and the fistulous tract was excised out in toto (Figure 4). Cricothyroid muscle was ligated back. Ryle's tube feeding was started for the patient post-operatively and was removed after 10 days. On follow up after 4 months, external wound has healed well and patient is doing well with no complaints. Patient has been on regular follow ups post-surgery and has reported no new symptoms.

Figure 3: Intra-operative clinical photograph showing the delineated sinus tract.

Figure 4: Clinical photograph of the excised sinus tract.

DISCUSSION

Second and third branchial cleft anomalies have very similar presentations. Both present with a swelling in the neck that are deep to the sternocleidomastoid muscle, but on surgical exploration the difference between the two become apparent due to the difference in the course of their tracts.[2]

Second branchial cleft anomalies constitute an overwhelming 90-95% of all the branchial arch anomalies, whereas the third and fourth arch anomalies are extremely rare and constitute around 2-8% and 1-4% of the total respectively.[1,3,4]

It has been noted that around 90% of the third branchial cleft fistulas occur on the left side, whereas only about 10% of them are reported to occur on the right side.[1,4] Our patient had a right sided fistula.

Most commonly third and fourth arch fistulas present with an external opening in the anterior border of the sternocleidomastoid, and the inner opening at the base of the pyriform sinus in case of third arch fistulas and at the apex of pyriform sinus in case of fourth arch fistulas.[1,5,6]

The recommended treatment of third branchial anomalies, warrant a complete excision of the entire fistulous tract.[7] Conventionally, this treatment consisted of surgical sinus tract excision with or without a hemithyroidectomy.[5-7]

Endoscopic cauterization of the sinus tract represents a minimally invasive therapeutic alternative to open-neck surgery.[7]

Recurrence of third branchial fistula after surgery is usually a consequence of incomplete excision of the tract during previous surgeries. Recurrent fistula surgeries are even more complex as is lot of inflammatory and adhesive tissue that needs to be dealt with.[7-9]

In our case, our patient had a history of two previous surgeries in the past, and this was a third time recurrence. She had a discharging sinus opening at the anterior border of sternocleidomastoid on the right side with the internal opening at the base of pyriform fossa.

Intraoperatively, we viewed the pyriform fossa using a Kleinsasser suspension laryngoscope and cannulated it using an infant feeding tube with a guide wire inserted inside it to provide some rigidity. This tube made it easier for the internal opening to be traced when approached through the cervical incision.

We also found that the tract was coursing close to the posterior part of the lamina of the thyroid cartilage, but through careful dissection we were able to completely free the tract from the cartilage and hence leave it completely intact.

The success of this surgery was due to a collaborative effort from all three departments, ENT, plastic surgery and radiology. Radiology played an important role as they helped us trace the course of the entire tract, which helped us ensure that it is completely dissected out, so that we can give the patient a good quality of life.

CONCLUSION

Our aim in reporting this case is to highlight the importance of having a high index of suspicion for branchial arch anomalies in children with neck swellings, as it could help prevent fistula formations and multiple surgeries. In this instance we also would like to highlight using a guidewire, or a makeshift one like we did with the infant feeding tube with the guidewire inserted inside to give it some strength, that would assist in delineating the internal part of the tract. We also would like to highlight that it is not necessary to remove part of the thyroid cartilage in all cases of third and fourth branchial fistula, as careful and meticulous dissection can ensure that no part of the tract is left behind, thus preventing any possibility of recurrence in the future.

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