Collaural fistula (Work Type II first branchial cleft anomaly) with prolonged morbidity: A case report

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Abstract
Collaural fistula is a very rare Work Type II first branchial cleft anomaly in which there is a complete fistulous tract between external auditory canal and the neck. Misdiagnosis and mismanagement can lead to prolonged morbidity and complications due to repeated infections. We present a case of an 18-year-old lady with a recurrent discharging sinus on her neck for 4 years. She has been treated with repeated incision and drainage and multiple antibiotics in the past. Otoscopic examination revealed an opening on the floor of the left external auditory canal. A diagnosis of an infected collaural fistula was made. Complete excision of the fistulous tract was done after treatment of the active infection. On follow-up, there was no further recurrence at 1 year. Sound knowledge of embryology of branchial anomalies with good history and examination is important to make correct and early diagnosis to prevent morbidity.

Keywords
Branchial anomaly, branchial cleft, collaural fistula, neck abscess, otolaryngology

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Introduction
Branchial cleft anomalies are the second most common head and neck congenital lesions seen in children of which second cleft origin makes up 95% of the anomalies.1,2 First branchial cleft anomalies are rare and account for <8% of all branchial anomalies.3 Third and fourth cleft anomalies are rarer.1,2 First branchial cleft anomalies are results of incomplete obliteration of the cleft between the first and second arches and they present as cyst, sinus or fistula depending on the degree of obliteration.2 Collaural fistula is the least common of first branchial cleft anomalies in which there is a communicating fistulous tract between floor of external auditory canal and the neck at the angle of mandible.4 The principle of management includes early diagnosis, controlling infection and complete surgical excision of the tract with preservation of the facial nerve.5

Case report
An 18-year-old lady presented to ENT clinic with a discharging sinus on the left side of her neck. She gave a history of recurrent neck abscess with pus discharge for over 4 years. She had to undergo incision and drainage for about 10 times in the past and had taken repeated course of different antibiotics. Her infection however subsided only temporarily and recurred each time with pus discharge, pain and swelling. She did not have any ear symptom. On examination, there was a sinus opening on her upper neck at the level of hyoid bone anterior to sternocleidomastoid muscle (Figure 1).

Some pus was draining out from the sinus at the time of first presentation to us and the surrounding skin was scarred. Otoscopic examination revealed an opening on the floor of the external auditory canal directed inferiorly (Figure 2). Tympanic membrane was found to be normal. A clinical diagnosis of infected collaural fistula was made. Fistulogram was attempted but we could not inject the contrast which could be due to fibrosis from recurrent infection. Imaging studies were not done due to long waiting time in a resource-poor setting. Excision of the sinus tract was performed under general anaesthesia after the acute infection has settled.
course of oral antibiotic. Circular skin incision was made encompassing scarred skin around the sinus opening. A small size nasogastric tube was used as a guide probe and dissection was done with a wide incision for good exposure (Figure 3).

The sinus tract was traced upwards towards the opening at the external ear canal. We found the sinus tract running superficial to the facial nerve. The upper end of the sinus tract opened at the cartilaginous part of the external auditory canal. We ligated the upper end of the tract, and the sinus tract was excised completely (Figure 4).

The wound was closed with a drain. Post operatively, the surgical wound healed well and she did not have further recurrence of infection at 1-year follow-up.

Discussion

To understand branchial cleft anomalies, sound knowledge of the embryology of branchial apparatus is necessary. Four well-defined sets of mesodermal arches divided by endothelial-lined pouches internally and ectodermal lined clefts externally appear during the fourth week of gestation called branchial apparatus. Two additional rudimentary arches also appear which are not visible on the surface of the embryo. Abnormal persistence of branchial apparatus remnants leads to branchial anomalies which present as cysts,
First branchial cleft anomalies are rare and account for <8% of all branchial cleft anomalies. Work in 1972, classified first branchial cleft anomalies into Type I and Type II. Type I anomalies are ectodermal and present as a cystic mass while Type II anomalies are ectodermal and mesodermal in origin and present as a cyst, sinus or fistula. Collaural fistula is Work Type II first branchial cleft anomaly and the least common of the first cleft anomalies. It has a communicating fistulous tract between floor of external auditory canal and the neck at the angle of mandible. The fistulous tract may lie superficial, deep or between the branches of facial nerve. In our case, the tract was superficial to the facia nerve. Although more common in children, branchial cleft anomalies can present in adults. Our case was 18 years old at the time of the diagnosis although she had presented much earlier.

Diagnosis of branchial cleft anomalies depends on good history and examination, and high index of suspicion. Computerized tomography (CT) and magnetic resonance imaging (MRI) scans are useful to evaluate and define the lesion as well as to evaluate the relationship of the sinus tract to the facial nerve. We could not do imaging studies in our case due to very long waiting time. definitive treatment is complete surgical excision to prevent recurrence. Some authors recommend superficial parotidectomy with facial nerve exposure and facial nerve monitoring. In our resource-poor setting, we did not have privilege of such facility. However, we recommend pre-operative imaging studies to delineate the lesion and the use of intra-operative facial nerve monitor to minimize facial nerve damage. Misdiagnosis of first branchial cleft anomalies is common, and in our case, diagnosis was delayed by 4 years. Misdiagnosis with repeated incision and drainage has led to prolonged morbidity and unnecessary hospital visits. Repeated infection and surgery will lead to scarring which makes excision difficult and risky for facial nerve injury intra-operatively.

Conclusion

In conclusion, collaural fistula is a very rare Work Type II first branchial cleft anomaly. Delay in diagnosis leads to repeated infections and prolonged morbidity. Sound knowledge of embryology of branchial anomalies with high degree of suspicion is essential for early diagnosis. We recommend thorough head and neck examination including careful otoscopy to avoid misdiagnosis.

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Ethical approval

Our institution does not require ethical approval for reporting individual cases.

Informed consent

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