DOI: 10.7860/JCDR/2024/66851.19013

Case Report

Ear, Nose and Throat Section

Collaural Fistula on the Left-side of the Face: A Case Report

SAGAR GAURKAR[1], CHANDRAVEER SINGH[2], JASLEEN KAUR[3], PRASAD T DESHMUKH[4]

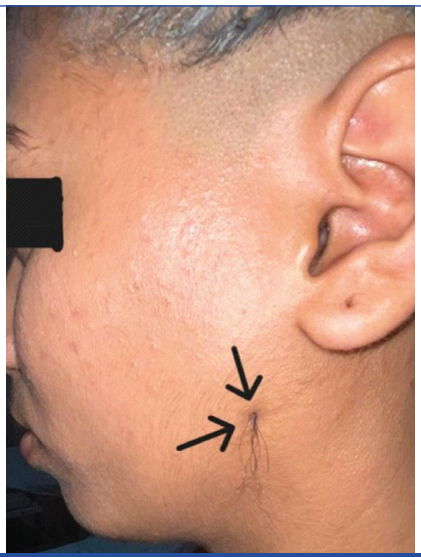
ABSTRACT

Collaural fistula, also known as cervico-aural fistula, is a rare anomaly that accounts for less than 8% of all branchial cleft anomalies. These fistulas result from incomplete closure of the first branchial cleft during embryonic development. A 21-year-old female exhibited depression on the left-side of her face since childhood, accompanied by a fistula. She had a history of intermittent discharge and a tuft of hair emerging from it. Additionally, the patient reported left ear discharge and an opening in the floor of the External Auditory Canal (EAC). Diagnostic imaging, including X-ray fistulogram, ultrasound, and Magnetic Resonance Imaging (MRI) neck fistulogram, played a crucial role in confirming the diagnosis of a collaural fistula. Surgical intervention involved excision of the fistulous tract and reconstruction of the EAC floor. The patient received antibiotic therapy, resulting in successful postoperative outcomes. The present case highlights the importance of recognising and appropriately managing collaural fistulas. When these fistulas are discovered early, along with a solid grasp of regional embryology and anatomy, there is a chance for better treatment outcomes.

Keywords: Branchial cleft anomalies, Fistulogram, Sinus

CASE REPORT

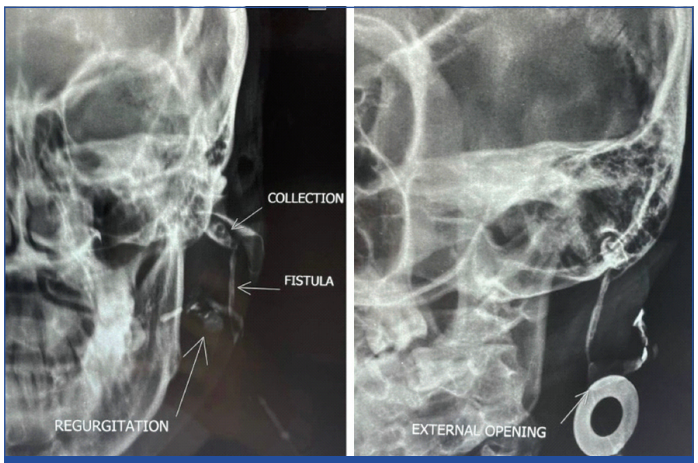
A 21-year-old woman presented with complaints of depression on the left-side of her face since birth, along with a fistula located approximately 2 cm in front of the left ear. The patient reported a tuft of hair emerging from the fistula and occasional episodes of left ear discharge. The clinical examination revealed a sinus opening on the left-side of her face. Upon otoscopic examination, the tympanic membrane appeared normal, and there was an opening on the floor of the EAC that directed inferiorly. Clinically, a collaural fistula with an unusual opening 2 cm above the mandible was diagnosed [Table/Fig-1].



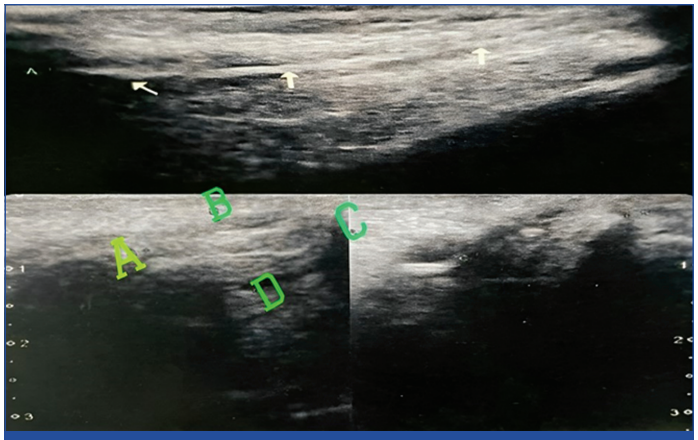
[Table/Fig-1]: A tuft of hair coming out from the fistula (black arrow) on the left side of the face.

A fistulogram of the left parotid region was performed using an X-ray [Table/Fig-2]. One millilitre of iodinated non ionic contrast medium was injected through the external opening in the left cheek region. Radiographs were obtained before and during the contrast injection. The precontrast control radiograph showed no significant abnormalities in the bone or soft tissue. After the injection of contrast, a 33.9 mm long and 1.6 mm thick fistulous tract was observed, extending superiorly to the external opening in the left parotid region, laterally to the left mandibular ramus and left mastoid bone, and terminating in a collection measuring 12.7×6.6×9.3 mm (ML×CC×AP). Continued injection of contrast resulted in the regurgitation of the contrast. The ultrasound examination [Table/Fig-3] revealed a fistulous tract extending superiorly and medially toward the left external ear. The fistula terminated in an echogenic structure measuring 23.9×12.3×22.9 mm, which lacked vascularity. MRI imaging [Table/Fig-4] confirmed the presence of a collaural fistula, with a thick caliber tract extending from the external opening in the left pre-auricular region. The tract followed the lateral margin of the left parotid gland, curving posteromedially to communicate with the inferior part of the EAC. Notably, a tiny air focus was observed within the tract near the EAC. These imaging findings supported the diagnosis of a collaural fistula with an unusual presentation on the left-side of the face.

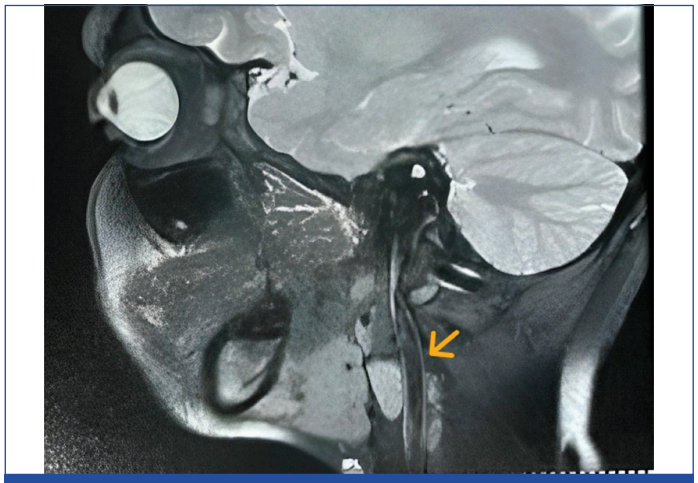
The patient underwent surgical intervention under general anaesthesia. An elliptical incision was made around the fistula opening, followed by the injection of dye for visualisation. The fistulous tract was carefully dissected from the surrounding tissue. A modified Blair's incision was made, extending from the left root of the helix, tragus, and earlobe



[Table/Fig-2]: X-ray left parotid region fistulogram) suggested an ectopic fistulous tract with the possibility of involvement of accessory left parotid gland.

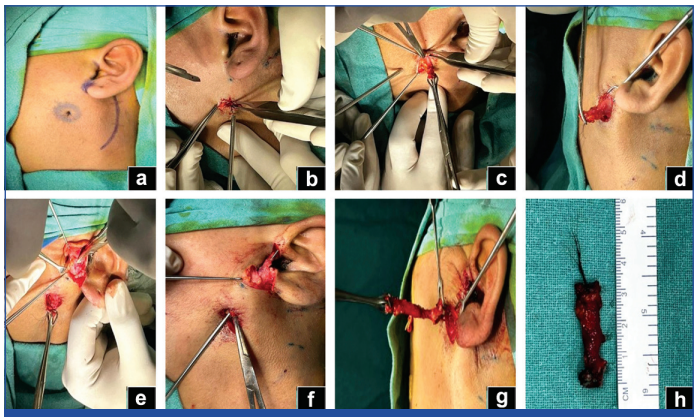


[Table/Fig-3]: Shows Ultrasonography (USG) of parotid region. A long fistulous tract (shown by white arrows) is seen extending from the external opening in the left infra-auricular region extending superiorly medial to the left external ear near the superior part of the left parotid gland. The fistula is seen terminating in an avascular echogenic structure of size 23.9×12.3×22.9 mm (marked by A, B, C, D).



[Table/Fig-4]: MRI neck sagittal view shows a thick calibre hyperintense tract (shown by yellow arrow) with an external skin opening in the left pre-auricular region (parotid gland region). Imaging features are consistent with the left first branchial cleft fistula.

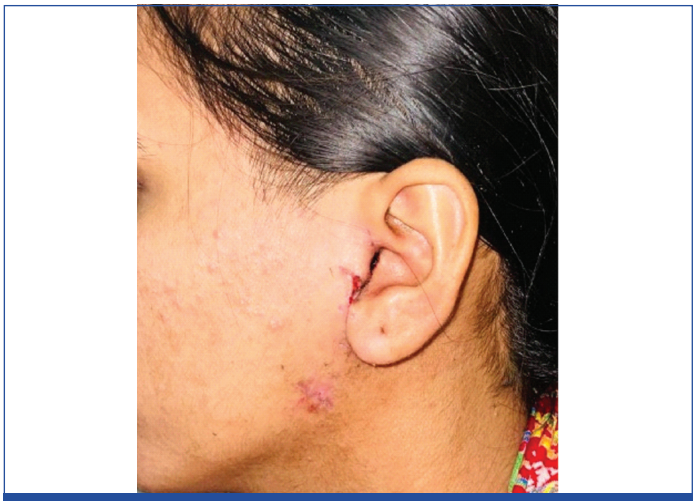
up to the mastoid tip. Dissection involved the layers of the skin, subcutaneous tissue, and superficial musculoaponeurotic fascia. The fistulous tract was traced and separated from the surrounding tissues, extending up to the level of the EAC floor. Complete excision of the sinus tract, measuring approximately 3.5×1 cm, along with the tuft of hair, was performed [Table/Fig-5a-h]. The excised specimen was sent for histopathological examination. Histopathological examination revealed a stratified squamous epithelial lining and focal granulation tissue with a tuft of hair. Fibrous tissue proliferation around the tract suggested a chronic inflammatory response. A negative suction corrugated drain was inserted and secured. The wound was closed



[Table/Fig-5]: a) Shows the marking of an elliptical incision around the fistula and modified Blair incision; b-d) Shows dissection of the sinus tract from elliptical incision and pointer inserted from fistula, traversing up to floor of EAC; e and f) Shows dissection of fistula from floor of EAC; g) Shows the dissected sinus tract, and h) Shows the sinus tract measuring approximately 3x1 cm along with a tuft of hair coming out of it.

in two layers. The cartilage of the EAC was meticulously sutured, and the patient received postoperative antibiotic therapy. Parotid dressing was applied. The patient was discharged after drain removal on the 5th postoperative day. Suture removal was performed on day 7.

The patient's postoperative course was uneventful, with the resolution of left ear discharge and the discharging sinus over the left-side of the face. Regular follow-up visits were planned to assess the progress of wound healing and ensure the long-term stability of the reconstructed EAC floor [Table/Fig-6].



[Table/Fig-6]: Healed scar after one month of follow-up.

DISCUSSION

Understanding branchial cleft anomalies requires a good understanding of the embryology of the branchial apparatus. The branchial apparatus forms during the fourth week of pregnancy and consists of four distinct sets of mesodermal arches, separated by endothelial- and ectodermal-lined pouches on the inside and outside, respectively. Additionally, two rudimentary arches also form, although they are not visible on the embryo's surface [1]. Abnormal persistence of remnants from the branchial apparatus can lead to branchial anomalies, which typically present as cysts, sinuses, or fistulas. A sinus is characterised as a tract with a blind end that can arise from the branchial apparatus [2]. These sinuses may be connected to the pharynx (branchial pouch sinuses) or the skin (branchial cleft sinuses) [2]. On the other hand, a fistula is defined as a communication between two surfaces that have undergone epithelialisation. In the case of a congenital branchial fistula, it would involve a connection between a persistent pouch and a cleft [3,4].

First branchial cleft anomalies are the least frequent among these abnormalities, accounting for less than 8% of all branchial cleft anomalies. In 1972, they were classified into Type-I and Type-II. Type-I abnormalities, which originate from ectoderm, present as cystic masses. Type-II anomalies, originating from both mesoderm and ectoderm, present as cysts, sinuses, or fistulas [3]. Collaural fistula, the least common type of first branchial cleft anomaly, falls under Type-II. It involves a connecting fistulous tract, typically located at the angle of the jaw, between the floor of the EAC and the neck. The fistulous tract may be superficial, deep, or wedged between branches of the facial nerve. In the described case, the fistula is situated 2 cm above the mandible. It is important to note that while branchial cleft anomalies are more commonly observed in children, they can also present in adults [5]. Penjor D and Kitamura M reported a case with a sinus opening on the upper neck, positioned at the level of the hyoid bone anterior to the sternocleidomastoid muscle [5]. In contrast, present case presents a unique and atypical appearance, with the sinus opening and associated hair tuft located 2 cm above the mandible. This distinctive location sets present case apart, highlighting a variation in the presentation of this condition.

Otorrhoea, which refers to recurrent or chronic discharge from the ear, is the most common otological symptom associated with

branchial cleft anomalies. It is important to consider this condition when recurrent or chronic otorrhoea occurs in the absence of chronic otitis. Although only 44% of patients with branchial cleft abnormalities show a sinus or fistula opening in the EAC, it should be noted that even if one exists, it may not be obvious. Sichel JY et al., observed that in certain cases, first branchial cleft defects were connected to a myringal web, which is an epidermal structure that extends from the floor of the EAC to the umbo of the tympanic membrane [6]. It is widely acknowledged that there is no direct relationship between the myringal web and the sinus tract or fistula, and it is uncommon for the tract to affect the tympanic membrane or middle ear [2,7].

Accurately diagnosing first branchial cysts in and around the parotid gland can be challenging without surgical exploration. One anatomical reference point used for localisation is Poncet's triangle, which is the typical area where first branchial cleft cysts or their sinus orifice are found. Poncet's triangle is defined by the EAC above, the mental region anteriorly, and the hyoid bone inferiorly. It serves as a useful guide for identifying the approximate location of these cysts [6].

Branchial cleft anomalies are diagnosed using a comprehensive medical history, physical examination, and a high degree of suspicion. Imaging procedures such as Computed Tomography (CT) and magnetic Resonance Imaging (MRI) with fistulograms can help define the lesion and determine how the sinus tract connects to the facial nerve. Complete surgical excision is the only effective treatment for branchial cleft abnormalities to prevent recurrence. Some authors advise performing a superficial parotidectomy while exposing and monitoring the facial nerves. However, in settings with limited resources and the absence of such facilities, preoperative imaging tests can still be useful in defining the lesion. Intraoperative facial nerve monitoring is also advised to reduce the risk of facial nerve damage [1,8].

Mandal MM et al., documented a case involving a fistula extending between the floor of the EAC and the nasopharynx. Notably, the fistulous tract ran superior and medial to the trunk of the facial nerve [9]. This specific anatomical trajectory adds a distinctive element to their reported case, emphasising the importance of detailed anatomical considerations in such presentations.

Dutta A and Vallur S reported a case involving a 32-year-old male who presented with first branchial cleft anomalies on both sides. The patient had discharging cutaneous openings on the right-side and a cystic swelling on the left-side [10]. The present highlights the rare occurrence of bilateral first branchial cleft anomalies and the diverse clinical manifestations associated with such anomalies.

Pol SA et al., documented a case involving a four-year-old girl who had two sinuses surrounding the lobule of the right pinna. Under general anaesthesia, a modified Blair incision with an elliptical rim around the sinuses was performed, revealing three tracts. These tracts were located below, behind, and in front of the right ear lobule. The tract beneath the ear lobule ran parallel to the floor of the cartilaginous EAC, displaying duplication of the canal and reaching the osteocartilaginous junction along the floor [11]. The entire tract was successfully removed. In contrast to study by Pol SA et al., patient in the present study was 21-year-old with a single fistula, and following a modified Blair incision, the fistulous tract was traced extending up to the level of the EAC floor [11].

It is worth noting that misdiagnosis of first branchial cleft anomalies is common, highlighting the importance of considering these anomalies and conducting appropriate investigations to ensure accurate diagnosis and appropriate management.

CONCLUSION(S)

Collaural fistulas are rare anomalies that pose diagnostic challenges due to their diverse and atypical presentations. Misdiagnosis and inappropriate treatment are not uncommon, highlighting the need for awareness among clinicians. Accurate diagnosis, achieved through meticulous clinical evaluation and diagnostic imaging, is crucial for appropriate management. Surgical intervention, tailored to the individual case, remains the mainstay of treatment. The present case report emphasises the rarity of collaural fistulas and the significance of accurate diagnosis and appropriate management to ensure optimal patient outcomes.

REFERENCES

[1] Waldhausen JHT. Branchial cleft and arch anomalies in children. Semin Pediatr Surg. 2006;15(2):64-69.
[2] Houck J. Excision of branchial cysts. Oper Tech Otolaryngol-Head Neck Surg. 2005;16(3):213-22.
[3] Bajaj Y, Tweedie D, Ifeacho S, Hewitt R, Hartley BEJ. Surgical technique for excision of first branchial cleft anomalies: How we do it. Clin Otolaryngol Off J ENT-UK Off J Neth Soc Oto-Rhino-Laryngol Cervico-Facial Surg. 2011;36(4):371-74.
[4] Rea PA, Hartley BEJ, Bailey CM. Third and fourth branchial pouch anomalies. J Laryngol Otol. 2004;118(1):19-24.
[5] Penjor D, Kitamura M. Collaural fistula (Work Type-II first branchial cleft anomaly) with prolonged morbidity: A case report. SAGE Open Med Case Rep. 2021;9:2050313X211014515.
[6] Sichel JY, Halperin D, Dano I, Dangoor E. Clinical update on Type-II first branchial cleft cysts. The Laryngoscope. 1998;108(10):1524-27.
[7] Tham YS, Low WK. First branchial cleft anomalies have relevance in otology and more. Ann Acad Med Singapore. 2005;34(4):335-38.
[8] Ash J, Sanders OH, Abed T, Philpott J. First branchial cleft anomalies: Awareness is key. Cureus. 2021 Dec 24 [cited 2023 Nov 17]; Available from: https://www.cureus.com/articles/79756-first-branchial-cleft-anomalies-awareness-is-key.
[9] Mandal MM, Panchal AJ, Kher SB, Dudhani HG, Shah VD. Unusual case of congenital fistula between external auditory canal and nasopharynx. Int J Otorhinolaryngol Head Neck Surg. 2021;7(5):912.
[10] Dutta A, Vallur S. An unusual case of bilateral first branchial cleft anomaly: A case report with review of literature. Indian J Otolaryngol Head Neck Surg. 2020;72(3):381-84.
[11] Pol SA, Singhal SK, Gupta N, Sivanandan SP. Recurrent collaural fistula: A challenge. Int J Otorhinolaryngol Head Neck Surg. 2019;5(5):1404.

PARTICULARS OF CONTRIBUTORS:

1. Associate Professor, Department of Otorhinolaryngology, Jawaharlal Nehru Medical College, Datta Meghe Institute of Higher Education and Research (Deemed to be University), Sawangi (M), Wardha, Maharashtra, India.
2. Assistant Professor, Department of Otorhinolaryngology, Jawaharlal Nehru Medical College, Datta Meghe Institute of Higher Education and Research (Deemed to be University), Sawangi (M), Wardha, Maharashtra, India.
3. Postgraduate Resident, Department of Otorhinolaryngology, Jawaharlal Nehru Medical College, Datta Meghe Institute of Higher Education and Research (Deemed to be University), Sawangi (M), Wardha, Maharashtra, India.
4. Professor, Department of Otorhinolaryngology, Jawaharlal Nehru Medical College, Datta Meghe Institute of Higher Education and Research (Deemed to be University), Sawangi (M), Wardha, Maharashtra, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Sagar Gaurkar,
Associate Professor, Department of Otorhinolaryngology, Jawaharlal Nehru Medical College, Datta Meghe Institute of Higher Education and Research (Deemed to be University), Sawangi (M), Wardha-442107, Maharashtra, India.
E-mail: shaggy6486@gmail.com

PLAGIARISM CHECKING METHODS: [Jain H et al.]
- Plagiarism X-checker: Aug 05, 2023
- Manual Googling: Oct 19, 2023
- iThenticate Software: Dec 02, 2023 (18%)

ETYMOLOGY: Author Origin

EMENDATIONS: 6

AUTHOR DECLARATION:
- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? Yes
- For any images presented appropriate consent has been obtained from the subjects. Yes

Date of Submission: **Aug 04, 2023**
Date of Peer Review: **Oct 06, 2023**
Date of Acceptance: **Dec 05, 2023**
Date of Publishing: **Feb 01, 2024**