

Case Report

# Collaural Fistula: Usually Misdiagnosed, Inadvertently Ill Treated and Ideally Microscopically Excised

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## ABSTRACT

First pharyngeal arch anomalies present usually as cysts, sinuses and fistulae, with an incidence of <10%. These anomalies need a high degree of clinical suspicion and once diagnosed a expertise for adequate management as misdiagnosis is not uncommon with subsequent further inappropriate management. Collaural fistula is a fistulous tract connecting external ear to the collar or neck region, which may as a persistently discharging sinus.

We here present a case of 30 year old male patient who presented with a ulcer below left ear lobule in upper neck with history purulent discharge since childhood along with intermittent serosanguinous discharge from external auditory canal, initially managed as abscess, and further diagnosed as collaural fistula. He was managed with microscopic excision.

Hence, a persistently discharging neck sinus should raise a suspicion of it being a collaural fistula, which is ideally treated with microscopic excision after confirming the diagnosis with help of MR fistulogram.

**Keywords:** Collaural fistula, Fistula, Neck sinus, Pharyngeal arch.

## INTRODUCTION

The external ear is a derivative of 1<sup>st</sup> pharyngeal arch. Its anomalies usually present as cysts, sinuses, or fistulae. At the same time, their incidence is less than 10% among all the pharyngeal arch anomalies. A collaural fistula is a tortuous tract along an imaginary line between the tragus and the hyoid bone, running in close relation to the facial nerve. Its rarity and diverse presentation often lead to misdiagnosis and inappropriate treatment, which ends up in recurrence along with local infection of the tract. Misdiagnosis in ordinary clinical practice may include inflammatory lesions of the preauricular area and tumors, but distinct clinical features can be derived from embryological development for an appropriate diagnosis.<sup>1-4</sup> This paper reports a case in order to emphasize upon correct diagnosis and management of collaural fistula with microscopic excision in otorhinolaryngology.

## CASE REPORT

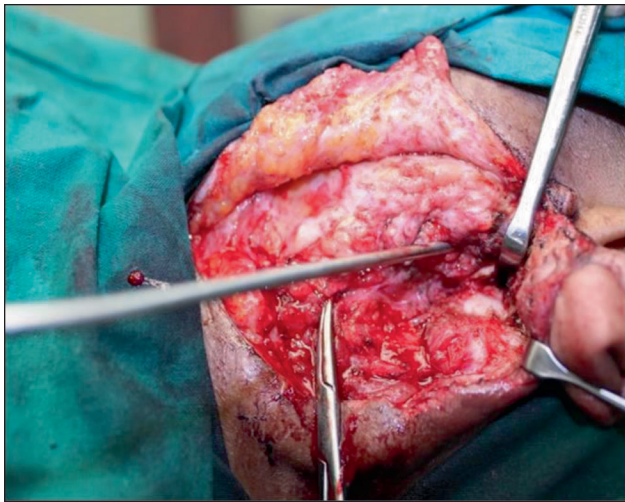
A 30-year-old male patient presented with complaints of ulcers below the left ear lobule in the upper neck, with a history of discharge since childhood, along with intermittent discharge from the left external auditory canal (EAC). Discharge from the neck was purulent, whereas it was serosanguinous from EAC. He was earlier managed as a case of neck abscess and underwent incision and drainage 6 months prior at a local hospital by a general surgeon. In view of misdiagnosis and inappropriate treatment, remission and relief were not achieved. Following the incision and drainage, he had a recurrence of discharge over the next 3 months.

On physical examination of the neck, there was a discharging sinus. A fistulous opening was found in the left EAC, 1 cm from the opening, with a scar anterior to the sternocleidomastoid muscle. An magnetic resonance (MR) fistulogram showed a fistular tract in the skin surface in the infraauricular upper neck region surrounded by focal abscess

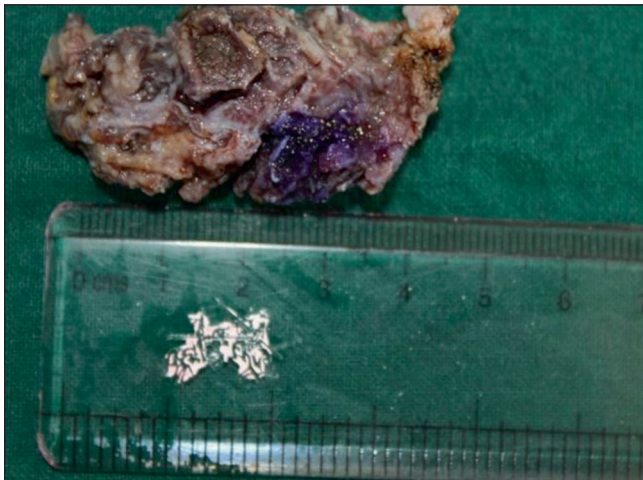
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**Figure 1:** Intraoperative picture showing the excision of the fistulous tract.



**Figure 2:** Fistulous tract excised in-toto.

collection in the left retroparotid and premastoid space with adjacent myofascitis found, and it was reported as a collaural fistula. An endoscopy of the larynx showed no abnormality.

He was further planned for left microscopic collaural fistula excision under general anesthesia. Methylene blue was injected into the tract and direction of the fistulous tract note [Figure 1]. The fistulous tract was dissected under microscopic vision using a sharp dissection with a scissor to the outer EAC around the tympanomastoid suture [Figure 2]. Further canalplasty was done after excision of the tract in toto and wound closure in 2 layers. EAC was packed with merocel, and the patient was discharged on postoperative day 2.

## DISCUSSION

Pharyngeal arch deformities have been classified as Type 1 and Type 2 by Arnot in 1971, with Type 1 being the ones

involved in the parotid region and Type 2 being the ones involving the anterior cervical region. These deformities have also been classified histologically by Work in 1972.<sup>5,6</sup> Here, Type 1 being ectodermal in origin due to duplication of membranous EAC and tract running parallel to EAC and Type 2 being both ectodermal and mesodermal in origin containing both skin and adnexal structures within.

As said earlier, any inflammatory process in the region of Poncet's triangle should raise the suspicion of collaural fistula, as it is the site where it typically arises.<sup>7</sup> The Poncet's triangle is bounded by EAC superiorly, mental region anteriorly, and hyoid bone inferiorly.

Choi and Zalzal studied, that the most common presenting feature of collaural fistula was discharge from sinuses, cervical mass, and repeated infections. Otorrhea was the most common otological symptom, and the collaural fistula should be suspected in cases of recurrent otorrhea in the absence of chronic otitis. Any inflammatory process in the region of Poncet's triangle should immediately raise an index of suspicion. The anomaly is seen twice as often in women (69%) than in men (31%) with left-side preference (64%). The patients may also present with non-healing ulcers of the neck and EAC. The most common site for collaural fistulas includes the cervical region (35%), parotid (35%), and preauricular region (24%). Only 44% of patients presented with an opening in the auditory canal EAC.<sup>5,8-10</sup>

Although otorrhea is the most common symptom, a complete examination should be done, which may reveal a fistulous opening in the EAC, seen only in 44% of the patients. Accurate diagnosis of the brachial cysts around the parotid gland is difficult clinically without complete surgical exploration, as said by Trijia.<sup>10-12</sup>

For diagnosis and workup, a fistulogram is done and needed, along with a computed tomography temporal bone (CT Temporal Bone) is needed to identify the facial canal along with magnetic resonance imaging neck (MRI Neck) to locate the proximity to the facial nerve. Methylene blue is injected into the tract in the opening in the neck (collar) region, and dye is seen coming out in the EAC. CT-Fistulogram can yield more accurate results regarding the course of the fistula. It is a useful diagnostic tool in the case of a sinus or fistula without the signs of cyst formation or inflammation. Radio-opaque sinuogram demonstrates the extent of the tract and confirms the position of the upper end.<sup>13-15</sup> Laryngeal endoscopy is required to rule out any other pathologies.

The differentials to be evaluated include any abscess, mycobacterial adenitis, lymphoepithelial cyst, and venolymphatic malformations.

The patient should be managed with surgical exploration and complete excision of the fistulous opening, along with the skin and cartilage involved. Split skin grafting along with shunting is required if > 30% of the skin of EAC is involved and excised. In the case of middle ear structures,

reconstructive surgery will be required.<sup>3</sup> The complications include facial nerve palsy and recurrence. Facial nerve injury is more prominent in the case of a Type 2 fistula, where the facial nerve is identified at the stylomastoid foramen. In case the stylomastoid foramen is also involved, the facial nerve should be traced proximally into the temporal bone. Facial nerve identification is aided with the use of a microscope for the surgery, which, along with facial nerve identification, aids in better visualization of the tract and achieving hemostasis.

Prasad S., in 2017 reported the use of modified parotid incision for opening of the fistula and tract dissection. Using a tragal pointer, the facial nerve was identified and noted to be superficial to the tract, where, as in our case report, microscopic excision aided the easy identification of the facial nerve. The granulation tissue was noted by Prasad S due to repeated incisions and drainage in the past, which can be better evaluated under a microscope, aiding a better cosmetic result.<sup>16</sup> In a resource-limited setting, a collaural fistula has been operated on and reported by Penjor D. in Bhutan, where a nasogastric tube had been used as a guide probe without any imaging in view of fibrosis secondary to repeated infections along with limited resources but carried a high risk of facial nerve injury. In resource-limited settings, circular incision is used, which carries with it a high morbidity along with cosmetic challenges.<sup>17</sup> Examination under the microscope is extremely essential, especially in case the sinus in EAC is not visualized, as noted by Shalima PS. They also note superficial parotidectomy can be planned in case of recurrent collaural fistula but carries a very high-risk facial nerve injury, which can be devastating in itself.<sup>18</sup>

## CONCLUSION

The first brachial arch anomalies should be kept in mind for any periauricular and cervical region inflammatory lesions. Whose remission depends largely upon complete excision, which in turn needs an understanding of embryogenesis along with knowledge of varying anatomical presentations. This knowledge is also useful for the identification and salvaging of facial nerves.

For the purpose of management, a CT fistuogram helps with the fixation of the diagnosis along with noting the direction of the fistula and planning the further course of action. While treating, microscopic examination and excision are essential for identification of the tract, along with for better hemostasis salvaging the facial nerve, and mastoid tip.

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