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## A case report on an ectopic accessory parotid fistula in an adult: A dilemma of acquired or congenital origin with delayed manifestation

Produl Hazarika

*Department of ENT, NMC Specialty Hospital, Abu Dhabi, UAE, [produl\\_ent@rediffmail.com](mailto:produl_ent@rediffmail.com)*

Seema Elina Punnoose

*Department of ENT, NMC Specialty Hospital, Abu Dhabi, UAE, [produl\\_ent@rediffmail.com](mailto:produl_ent@rediffmail.com)*

Rajeev Chaturvedi

*Department of Radiology, NMC Specialty Hospital, Abu Dhabi, UAE, [produl\\_ent@rediffmail.com](mailto:produl_ent@rediffmail.com)*

Suneet Kaur Hora

*Department of Pathology, NMC Specialty Hospital, Al Ain, UAE, [produl\\_ent@rediffmail.com](mailto:produl_ent@rediffmail.com)*

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## Case report

# A case report on an ectopic accessory parotid fistula in an adult: A dilemma of acquired or congenital origin with delayed manifestation

Produl Hazarika\*, Seema Elina Punnoose, Rajeev Chaturvedi, Suneet Kaur Hora

Email: produl\_ent@rediffmail.com

## Abstract

To our knowledge, this is the second known reported case of ectopic accessory parotid gland fistula in an adult in literature. Congenital parotid duct fistula itself is a rare entity, but even rarer is the occurrence of the fistula in an otherwise healthy adult. A sequential pictorial account has depicted the surgical excision of the fistula in its entirety and histopathology confirmed the parotid fistula. Two months post procedure, the patient continues to be symptom free. We used search engines such as PubMed, Medline, Google Scholar, and Wiley Online library to search published literature.

**Key words:** Accessory parotid gland, congenital parotid fistula, ectopic fistulous duct

## Introduction

Fistula of parotid gland is almost always acquired and cause of the fistula is most often due to trauma, infection, and tumor of either benign or malignant in nature. Congenital fistulas are extremely rare and manifest in childhood with a fistulous tract in the cheek near the angle of the mouth and are believed to be arising from an ectopic accessory parotid gland. We herein report a case of left sided spontaneous parotid fistula in a healthy adult, which we feel is of congenital origin, but has manifested later in adulthood.

## Case report

A 25-year-old young male of Pakistani national came to our ENT clinic in NMC Specialty Hospital, Abu Dhabi, on 13 March 2017 with a history of intermittent watery mucoidal discharge from

**Produl Hazarika<sup>1\*</sup>, Seema Elina Punnoose<sup>1</sup>, Rajeev Chaturvedi<sup>2</sup>, Suneet Kaur Hora<sup>3</sup>**

<sup>1</sup>Department of ENT, NMC Specialty Hospital, Abu Dhabi, UAE

<sup>2</sup>Department of Radiology, NMC Specialty Hospital, Abu Dhabi, UAE

<sup>3</sup>Department of Pathology, NMC Specialty Hospital, Al Ain, UAE

\* Corresponding Author

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Figure 1a: Showing the opening of salivary fistula

left side of his cheek away from the left angle of mouth for the past three months. The quantity of secretions increased during consumption of food. There was no associated pain or bleeding. There was no history of any infection, trauma, any kind of swelling, or tumor. He describes his discharge as watery, not foul smelling, not blood stained and slightly mucoidal. It started spontaneously without any pain or fever. He visited other local hospitals for the same problem, where wound dressing and other

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supportive treatment was offered, but had not found any remission in the symptom of discharge. Clinical examination revealed a fistulous opening with associated minimal indurated margin around 5.5 cm away from the left angle of mouth which is lying 1.1 cm below the imaginary line connecting the angle of mouth and tragus (Figure 1a, b). Oral cavity examination showed both Stenson's duct opening was in normal position. Culture of the secretions from the fistula came as sterile and no growth of organisms was seen. Salivary amylase estimation of the secretion could not be done because of the



Figure 1b: Showing the relation of left angle of mouth to the opening of the fistula in the cheek

non-availability of this facility in our institute. MRI face and neck scan with contrast done on 25 March 2017 showed a well-defined area of enhancing tissue in the left subcutaneous cheek region anterior to the masseter, lateral to buccinators, and inferior to the Stenson's duct with a fistulous tract extending from this region and opening into the oral cavity opposite to the left second upper molar just adjacent to the normal parotid duct opening (Figure 2a, b, c). Surgical excision of the fistula was performed on 7 May 2017. The steps of the surgical procedures (Figure 3a, b, c, d, e, f) further confirmed the existence of the fistula from cheek to buccal cavity. A curvilinear skin incision around the external fistulous opening was given and the skin along with the fistula was dissected out using bipolar cautery. The fistulous tract along with the skin was dissected out from the buccinator muscle and transposed into oral cavity in toto. Skin incision closed in two layers. Histopathology showed stratified squamous

epithelium overlying sub-epithelium showing skin appendages and fistulous tract lined by granulation tissue. No evidence of tuberculosis or Crohn's disease seen. No evidence of dysplasia or malignancy seen. Post-operative follow-up done two months later, on 10 July 2017 revealed a well healed cheek wound with no evidence of recurrence (Figure 4).

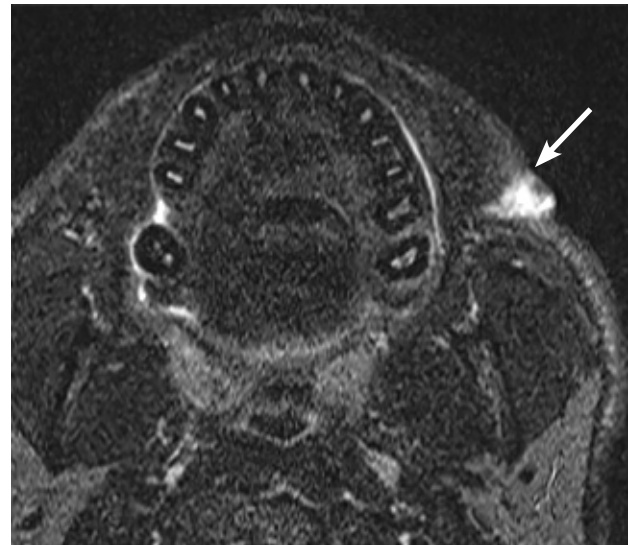


Figure 2a: Axial T2 image showing a linear hyper intense T2 fistulous tract (arrow) extending from the left cutaneous cheek surface and opening into the oral cavity opposing the left second molar.



Figure 2b: Axial T2 image (arrow head) showing the normal parotid duct opening in the oral cavity just adjacent to the pathological fistulous tract (small arrow) opposite the left upper second molar.



Figure 2c: Coronal STIR (short tau inversion recovery sequence) - Image showing hyper intense (big arrow) tissue in the subcutaneous left cheek which leads into a fistulous tract (small arrow) opening into oral cavity at left upper second molar level.

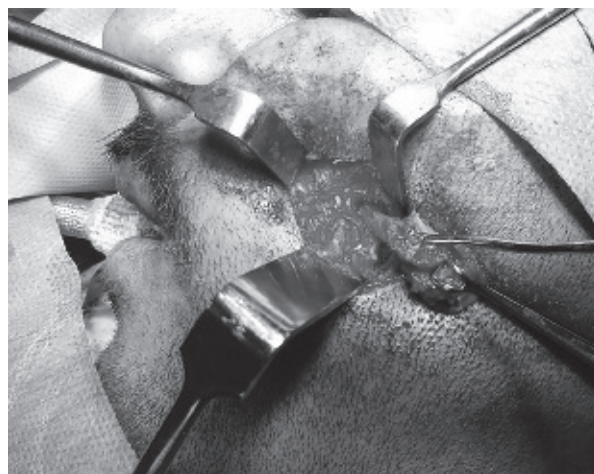


Figure 3c: More deeper dissection of the fistulous tract through buccal pad of fat and buccinator muscle

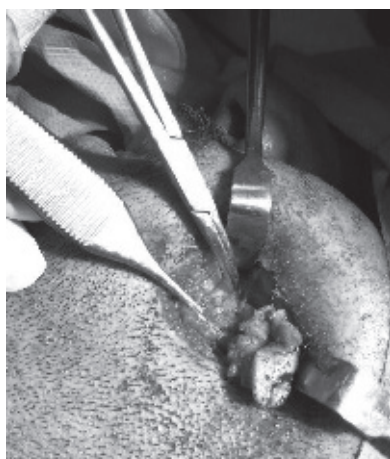


Figure 3a: A curvilinear incision around the opening of the fistula along with the skin

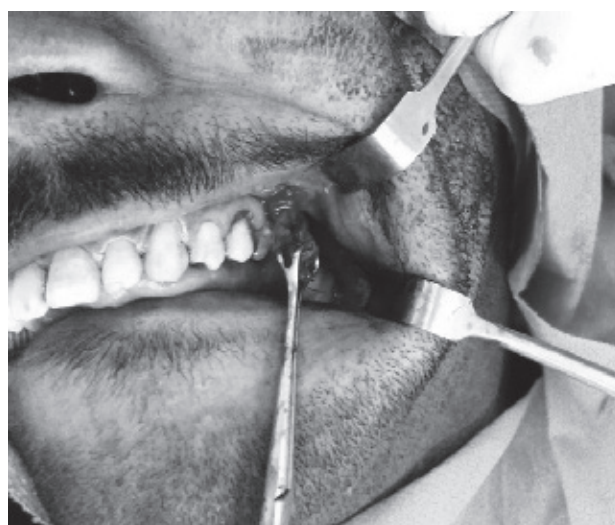


Figure 3d: Delivery of the fistulous tract through oral cavity



Figure 3b: Dissecting fistulous tract deeper - probe showing the intact fistula tract



Figure 3e: Showing the course of the tract after complete excision of the tract

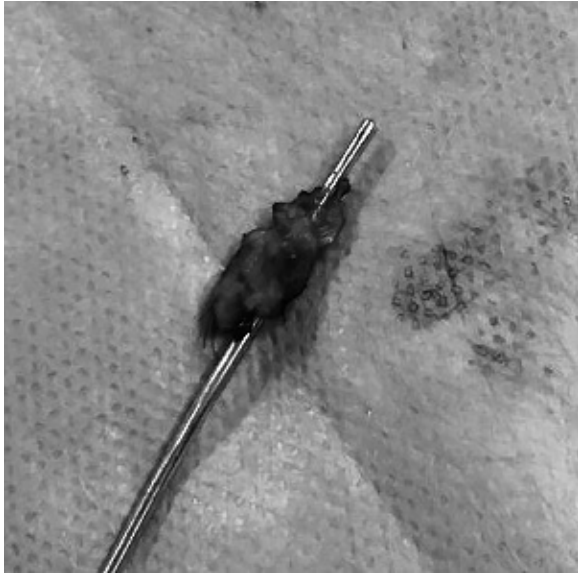


Figure 3f: Dissected specimen of the entire fistula

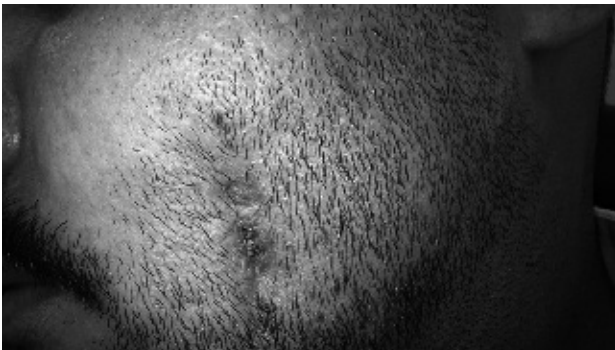


Figure 4: Post-operative photograph showing the healed skin incision

## Discussion

An oro-facial fistula is a pathological communication between the cutaneous surface of the face and oral cavity. Dental infection, salivary gland diseases, neoplasm, and developmental lesion are the common causes for oro-cutaneous fistula. Acquired oro-cutaneous salivary gland fistulas are commonly found in parotid gland because the course of the parotid duct comes close to the cutaneous surface crossing the outer surface of the masseter muscle. Congenital parotid duct fistulas are extremely rare and only ten such cases have been reported so far in literature, out of which nine are in children and one in adult. These fistulas were found to be arising from an accessory parotid gland. The congenital fistulae found in the region of the face and neck results due to a failure in the normal involution process of the branchial arches during embryonic development.

The most common site for an accessory parotid tissue is overlying the masseter muscle with the ductal system emptying into main Stensen's duct of parotid gland<sup>1</sup>, whereas ectopic accessory parotid fistula has its own independent ductal system.<sup>2</sup> Frommer (1977)<sup>3</sup> in their study of anatomical dissections found that 21% of human cadaveric dissections revealed a clearly detached accessory parotid gland at a variable distance from the main parotid gland. The accessory parotid gland was found in close relation to the Stensen's duct, because the duct passes along the lateral aspect of the masseter muscle and is usually seen positioned on or above the duct. Congenital parotid fistula arising from the ectopic accessory gland is an extremely rare condition and the fistulous opening is always located at the facial skin of the cheek near the angle of the mouth. Such congenital fistulas to manifest in adulthood are extremely rare and only one such case has been reported earlier in a 43-year-old man in English literature.<sup>4</sup> Our patient did not give any history of infection or trauma. It was spontaneous in origin presenting with a clear watery fluid in relation to the process of consumption of food. The location of the fistulous opening was near the left angle of mouth anterior to masseter, lateral to buccinator, and inferior to the Stensen's duct as described by Dutta (2017).<sup>5</sup> Thus, we would like to term our case as an adult ectopic accessory congenital parotid gland fistula based on the outset of history, MRI findings, and operative findings. MRI of face and neck with contrast shows a small well-defined area of enhancing tissue in the left subcutaneous cheek region away from the normal parotid gland lateral to the masseter muscle and was considered to represent an ectopic parotid gland with a separate ductal system of its own. Jernstrom P, Prietto C A (1962)<sup>6</sup> reported that ectopic salivary glands present as a swelling of the subcutaneous tissue and consisted of sinuses that generally open externally, secreting an odorless liquid similar to saliva, especially during meals. The reason for the late presentation could be the absence of the saliva-draining to external fistula, allowing the lesion to persist unnoticed.<sup>5</sup> Our surgical procedure also traces the tract very clearly. Histopathological examination has confirmed the dissected surgical specimen as a parotid fistula.

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