Atypical Location of the Facial Nerve in a Patient with a First Branchial Cleft Fistula

Case Report

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Abstract

First branchial cleft anomalies are rare. Its estimated incidence is one in 100,000. Clinically, patients present with recurrent otorrhea, periauricular swelling, and/or flowing fistula in the neck. Surgical removal of the tract is considered the best treatment option for the first branchial cleft fistula. Due to the close relationship between the fistula tract and the facial nerve branches, facial nerve injury is one of the not uncommon complications of this surgery. Different variations in the relationship between the fistula tract and the facial nerve trunk and its branches have been mentioned in the literature. In this study, we presented the case of an atypical course of the facial nerve in a nine-year-old pediatric patient who underwent first branchial cleft fistula surgery, and discussed the importance of anatomic variations and measures to be taken to prevent facial nerve injury.

Keywords: Branchial cleft, facial nerve, anatomy, congenital, surgery, pediatric, case report

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Introduction

First branchial cleft anomalies are rare, as the estimated incidence is one in 100,000 (1). Clinically, patients have recurrent swelling, hyperemia, pain, and inflammation whit periauricular swelling and/or flowing fistula in the neck.

Surgical removal of the tract is considered the best treatment option for first branchial cleft fistulas (2). Due to the close relationship between the fistula tract and the facial nerve branches, the parotid gland, and the external acoustic canal, the most common postoperative complications are parotid gland sialorrhea, facial paralysis, and external acoustic canal stenosis (3). Different variations in the relationship between the fistula tract and the facial nerve trunk and its branches have been mentioned in the literature (Figure 1).

The fistula tract, which is associated with the external auditory canal, usually passes through the superficial surface of the facial nerve. However, the probability of the fistula tract being located under the facial nerve or between its branches increases in younger patients (4).

The risk of nerve damage also increases in the surgery of these cases; therefore, these conditions should be taken into consideration, especially in patients presenting with the first branchial cleft fistula at a young age. Using intraoperative nerve monitoring helps surgeons identify the facial nerve and reduces the risk of nerve damage during the operation.

In this case report, we present a pediatric patient who had a first branchial cleft fistula with an atypical location of the facial nerve (Figure 2).

Case Presentation

A nine-year-old female patient presented with occasional discharge and swelling in front of the left ear lobe, which was reported to have been present since infancy. In the clinical examination, a hyperemic lesion with a draining hole was observed at the level of the angle of the mandible (Figure 3).

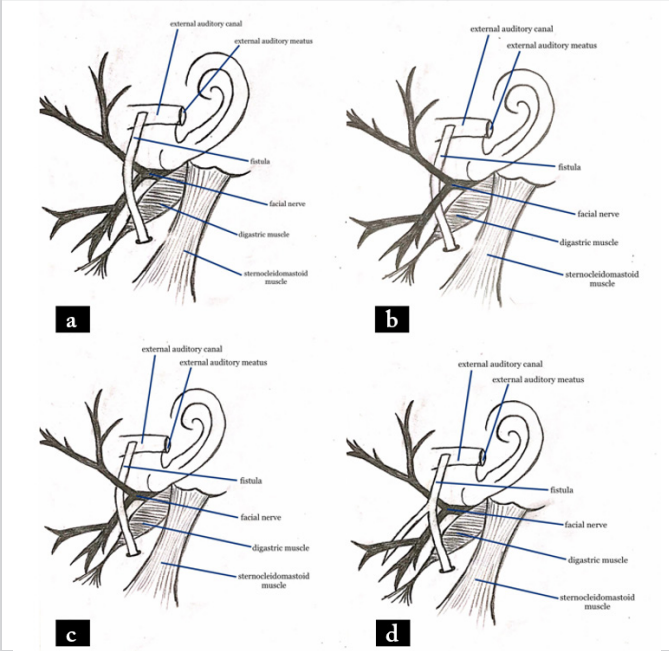

Figure 1. a) Fistula tract superficial to the facial nerve, b) Fistula tract deep to the facial nerve, c) Fistula tract between branches of the facial nerve, d) Fistula tract superficial to the facial nerve with additional deep extension

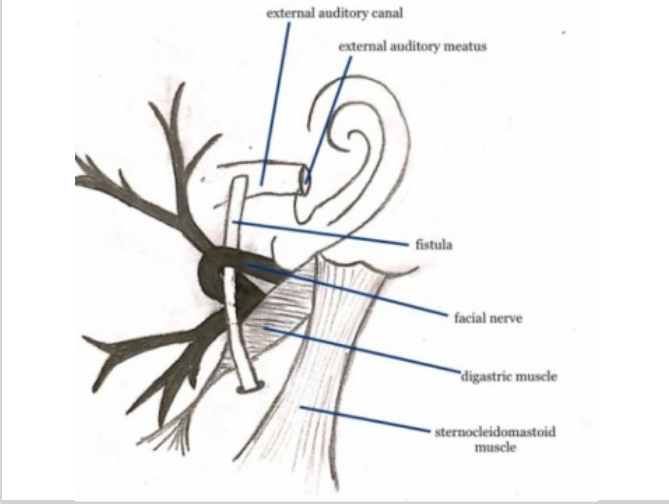

Figure 2. Facial nerve passing through the surface of the tragal pointer cartilage and rotating around the fistula tract (the situation in our patient)

Intraoral and otoscopic examinations were normal. There was no external auditory canal anomaly. Magnetic resonance imaging (MRI) of the neck revealed a 3x0.8x1 cm tubular structure extending from the left-sided skin to the parotid gland and further to the anteroinferior wall of the external auditory canal (Figure 4).

Although MRI revealed a fistula tract extending to the external auditory canal, the fistula orifice was not observed in the external auditory canal in otoscopic examination. The diagnosis of the first branchial cleft fistula, which opened under the skin of the external auditory canal on the left, was considered, and surgery was planned. Examination of the facial motor functions of the facial nerve in the preoperative period was evaluated as bilaterally normal. Superficial parotidectomy and total excision of the fistula tract were performed with intraoperative facial nerve monitoring (Figure 5). There was no need to perform a fistulogram or use methylene blue to identify the pathway during surgery.

During surgery it was found that the facial nerve trunk was more superficial than normal and had an atypical location compared to the fistula tract. After the facial nerve trunk passed through the surface of the tragal pointer cartilage, it was observed that it rotated around the fistula tract and

Figure 3. Hyperemic lesion appearance on clinical examination

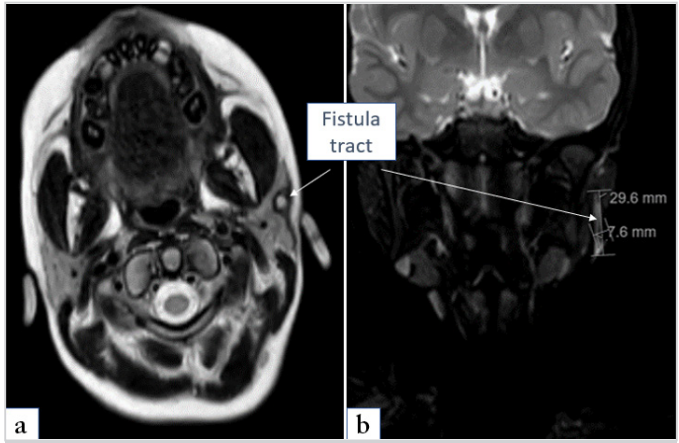

Figure 4. a, b) MRI T2 axial and coronal sections showing the fistula tract on the left side

divided into branches in the medial (deep) part of the tract (Figures 2, 6, 7).

After dissecting and removing the fistula tract with superficial parotidectomy, the facial nerve was identified with its main branches (Figure 8).

When the fistula tract was dissected, it observed that the fistula tract opened subdermally to the anteroinferior of the external auditory canal. The entire tract was excised.

Figure 5. Planning the surgical incision

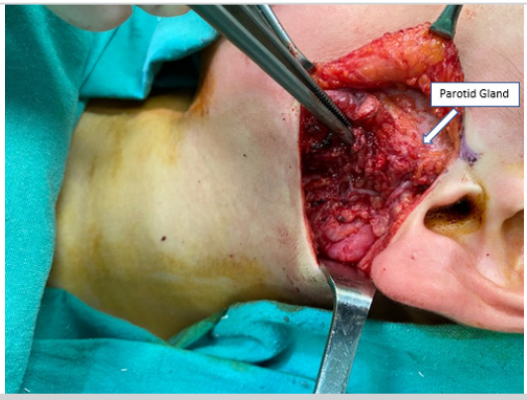

Figure 6. Exposure of the facial nerve

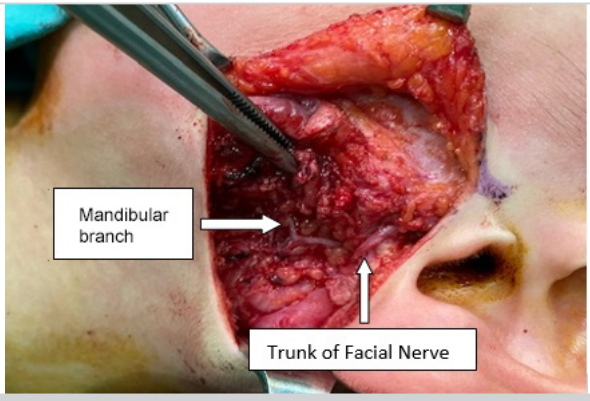

Figure 7. The facial nerve rotates around the fistula tract and divides into its branches

Electromyography responses were obtained by intraoperative facial nerve monitoring. Facial nerve examination was normal in the postoperative period. No signs of recurrence was observed during the six months follow-up of the patient.

Written informed consent was obtained from the parents of the patient for publication of this case report.

Discussion

Branchial clefts are the embryological precursors of the face, neck, and pharynx. In children, branchial cleft anomalies are the second most common congenital lesion in the head and neck region. They are more common in females. Among the branchial cleft anomalies, second branchial cleft anomalies are the most common type. First branchial cleft anomalies are the second most common and constitute 1–8% of the four cleft types (5).

First branchial cleft anomalies are often misdiagnosed and managed inadequately before surgical excision. They are typically located as cysts/sinuses/fistulas between the external auditory canal and the submandibular area. They can occur in the form of a flowing pit in the angle of the mandible, an inflammatory mass in the parotid gland region, or a discharge in the external ear canal (6).

First cleft anomalies are classified as type I or type II lesions (7). Type I lesions are the continuation of the membranous part of the external auditory canal and contain only ectodermal tissues. Type II lesions consist of ectoderm and mesoderm and may contain cartilage. Type I lesions are located lateral to the facial nerve, while type II lesions are located medial to the nerve. Type II lesions may appear as preauricular, infraauricular, or postauricular swellings or cysts below the angle of the mandible.

In the literature, different localizations of the facial nerve and fistula tract compared to each other were previously stated in a case series study (4).

In this report, we discuss a case with an incompletely formed first branchial cleft fistula located atypically with the facial nerve. No such case is mentioned in the literature.

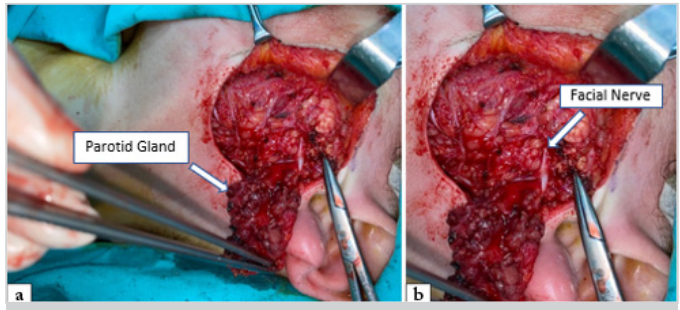

Figure 8. a) Appearance after superficial parotidectomy and removing the fistula tract, b) Exposure of the facial nerve branches

In the literature, it is stated that brachial cleft fistulas are more common on the left. When the position of the facial nerve and fistula tract are compared to each other, it is stated that the deeply located tract is more common at an earlier age than the facial nerve (4). In the presented case, the brachial cleft fistula was on the left, and the fistula tract was deeply located.

Radiological examinations provide important preoperative information for branchial anomalies. Ultrasonographic examination determines the structure and the size of the lesion. Computerized tomography and MRI help confirm the diagnosis and provide accurate anatomical information about the location. In MRI, axial and coronal T1 postcontrast imaging may show a cyst tract (8). In our case, a thick-walled fistula tract extending to the left external auditory canal was observed in the T1 and T2 MRI series (Figure 4).

First branchial cleft anomalies are at high risk for iatrogenic facial paralysis due to the proximity of the lesion to the facial nerve. Neither physical examination nor imaging techniques can clearly define the relationship between the lesion and the facial nerve (9). Several studies have been conducted on estimating the location of the facial nerve and its branches with MRI (10). However, no study has objectively shown the location of the facial nerve. Given the risk of injury to the nerve or its branches, it is critical to monitor the facial nerve during surgery.

First branchial cleft fistulas are rare. The fistula tract can cause anatomical variations in the extracranial location of the facial nerve, particularly in cysts from birth. Treatment includes surgical planning accompanied by facial nerve monitoring and total excision of the fistula tract, considering that it may be an atypically located facial nerve. Recognizing the facial nerve truncus at the early stage of dissection is critical. In young patients with fistula tract opening into the external auditory canal, variations in the atypical location of the fistula tract and facial nerve should be kept in mind, and attention should be given.

Informed Consent: Written informed consent was obtained from the parents of the patient for publication of this case report.

Authorship Contributions

Surgical and Medical Practices: S.H., E.D., Concept: S.H., E.D., Design: S.H., E.D., Data Collection and/or Processing: S.H., E.D., Analysis and/or Interpretation: S.H., E.D., Literature Search: S.H., E.D., Writing: S.H., E.D.

Conflict of Interest: There is no conflict of interest to disclose.

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Main Points

- Different variations in the relationship between the fistula tract and the facial nerve trunk and its branches are mentioned in the literature.
- Different variations are seen especially in patients presenting with their first branchial cleft fistula at a young age.
- In this study, we present an atypically located facial nerve and the relationship between the fistula tract and the facial nerve in a nine-year-old pediatric patient with a first branchial cleft fistula.

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