Case Report

An extremely rare case of adult with first branchial cleft fistula: Case report

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ABSTRACT

Introduction: Anomalies of the first branchial cleft are rare and represent 10% of lateral cervical malformations. Very few cases have been described in literature. In our research, we didn’t find cases of adults with first branchial cleft fistulas. First branchial cleft fistulas in adults are difficult to diagnose, and the surgery of the parotid gland to extract the fistula requires to be precautious.

Case presentation: The aim of this work is to present an extremely rare case of a 65 years-old patient with first branchial cleft fistula. Fistula excision surgery completed with superficial parotidectomy were performed. No sign of recurrence were found after 6 months surveillance.

Discussion: First branchial cleft fistula is hard to diagnose, especially with adults. Very few cases of adult male are described in the literature. It is often misdiagnosed with parotid tumours or as otitis with ear drainage. The treatment of First branchial cleft fistula is surgical. A complete resection of the tract is necessary to prevent recurrency. It usually requires a superficial parotidectomy and the fistula tract passes under the parotid gland.

Conclusion: First branchial cleft fistulas are rare, and forms with adults are extremely rare to encounter. Imagery can’t always help to diagnosis. Surgery is indicated in the management of this pathology, with preservation of the facial nerve.

1. Introduction

Anomalies of the first branchial cleft are rare and represent 10% of lateral cervical malformations with an estimated incidence of 1/100,000 [1]. During embryological development, the first branchial cleft is unique in that it is the only cleft that does not become obliterated completely by the eighth week of gestation and persists as a definitive structure in the embryo. First branchial anomalies are an uncommon group of lesions that arise from incomplete closure of the ventral portion of the first branchial cleft [2]. First branchial cleft anomalies are subdivided using the Work classification in 1972. Type I are generally cysts of ectodermal origin, considered to be a duplication of the membranous external auditory canal. Classically, these occur medial to the concha and frequently extend to the postauricular crease, running superior to facial nerve [3,4]. Type II are cysts or sinuses of ectodermal and mesodermal origin, considered to be a duplication of the membranous external auditory canal and pinna.

The purpose of our work is to report an extremely rare case of first branchial cleft fistula, diagnosed and treated successfully in the academic hospital of Casablanca.

The work has been reported in line with the SCARE 2020 criteria [5]:

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Cartilage of tragus after resection of fistula.
The postoperative check-up did not show any complications. The follow-up period was 6 months; the clinical examination did not find any sign of recurrence.

3. Discussion

Anomalies from the first branchial arch represent 8% of all branchial cleft anomalies. Of these, 16% are fistulae [1,4]. Sinus and fistula are mostly encountered with infants and children, when cyst is more common in adults [4]. Many classifications have been used to describe anomalies of the first branchial cleft. The first classification was proposed by Arnot in 1971 who described two types based on morphology.

Then, Work, in 1972, described two types of first branchial cleft anomaly. This classification is based on tissue of origin. Finally, Olsen in 1980 proposed a simpler classification system based on the clinical presentation of the lesion: Cyst, Sinus or Fistula [3]. In our case, it’s an adult with his lesion on the left side, where it’s most encountered. First branchial cleft fistulas are rare to encounter. In the literature, very few cases are described in adults and no case have been reported in third age patient.

Olsen, recognized that an infected cyst or a surgically drained cyst might form a sinus or a fistulous tract. He proposed a classification with anatomic variations on of the fistula tract and its relation with the facial nerve [1]. Most of first cleft fistulous tracts have a greater incidence of passing deep to the facial nerve [6]. At present, there is no imaging method capable of identifying a first branchial cleft anomaly with certainty [7].

In our studying case, the patient went through a surgery in his young age. Diagnosis was established on the clinical examination with an orifice at the inferior part of the left external auditory canal. Imagery with ultrasound and MRI scan were not relevant. Surgery went under difficulties due to muscular fibrosis. The dissection of fistulae tract was hard to execute. All the branches of facial nerve could not be entirely exposed. The fistulae passed under the superficial lob of parotid gland.

First branchial cleft fistula is hard to diagnosticate, especially with adults. Very few cases of adult male are described in the literature. It is often misdiagnosed with parotid tumours or as otitis with ear drainage.

The treatment of First branchial cleft fistula is surgical. A complete resection of the tract is necessary to prevent recurrence. It usually requires a superficial parotidectomy and the fistula tract passes under the parotid gland.

4. Conclusion

First branchial cleft fistula are rare, and forms with adults are extremely rare to encounter. Imagery can’t always help to diagnosis.
Surgery is indicated in the management of this pathology, with preservation of the facial nerve.

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**Ethical approval**

This type of study does not require any ethical approval by our institution.

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**Author contribution**

Wilfrid LENDOYE: Drafting and writing the article. Mouna LYOUBI: writing the article. Hafed RADHI: study concept. Youssef OUKESSOU: revising the article. Mohamed Mahtar: final approval.

**Consent**

Written informed consent was obtained from the patient for publication of this case report. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

**Registration of research studies**

Not applicable.

**Guarantor**

Dr. Wilfrid LENDOYE.

**Provenance and peer review**

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**Declaration of competing interest**

The authors declare no conflict of interest.

**Appendix A. Supplementary data**

Supplementary data related to this article can be found at https://doi.org/10.1016/j.amsu.2021.102807.

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