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## Atypical first branchial cleft fistula: A case report

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## ABSTRACT

**INTRODUCTION:** First branchial cleft anomalies (FBCA) are rare. They have an estimated incidence of 1 in 100,000. Type I are those that embryologically duplicate the membrane (cutaneous) external auditory canal.

The aim of this case is to describe an unusual path of a type II first branchial cleft fistula tract in a 3 years old child and its surgical management in the academic hospital of Casablanca.

**CASE PRESENTATION:** This case is about a 3 year old girl who presented to the Ear Nose Throat (ENT) consultation for recurrent right lateral cervical infection. Clinical examination found an unsightly scar attached to an orifice giving pus located near the right mandibular angle suggesting type II first branchial cleft anomaly.

Surgical excision was performed under general anesthesia by the superficial parotidectomy approach, the facial nerve was identified and preserved. The fistula cord was dissected and followed, it went under the facial nerve and the parotid gland to end under the digastric muscle where we tied it up. The post-operative check-up did not show any complications. The follow-up period was 12 months; the clinical examination did not find any sign of recurrence.

**CONCLUSION:** First branchial cleft fistula are rare and can be in form of cyst or fistula. Its management is surgical excision keeping the tract cyst of the fistula intact with facial nerve preservation.

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## 1. Introduction

First branchial cleft anomalies (FBCA) are rare. They have an estimated incidence of 1 in 100,000 [1,2]. In the embryonic period at seven weeks, the arches fuse and the clefts obliterate. Duplication or failure of obliteration of the embryologic tract is the likely etiology of these lesions [3]. During development, the closure time of the cleft is concurrent with the migration of the facial nerve and emergence of the developing parotid gland, which originates from the second branchial arch; thus, FBCA have a close relationship between these structures [4].

Despite their heterogeneity, several classification systems have been proposed to categorize these anomalies. First branchial cleft anomalies are subdivided using the Work classification in 1972 [5]: type I are those that embryologically duplicate the membrane (cutaneous) external auditory canal. They are located medial, inferior or posterior to the concha and pinna. The fistula tract or sinus stay superficial to the facial nerve and may parallel the external auditory canal and ends in a blind cul de sac at the level of the mesotympanum. Type II deformities are composed of both ectodermal and mesodermal elements and therefore contain, in addition

to skin, cutaneous appendages and cartilage. The external opening is near the angle of the mandible, while the tract courses superiorly and can be closely associated to the facial nerve.

The aim of this case is to describe an unusual path of a type II first branchial cleft fistula tract in a 3 years old child and its surgical management in the academic hospital of Casablanca.

This work has been reported in accordance with the SCARE criteria [6].

## 2. Case presentation

This case is about a 3 years-old girl with no relevant medical history who presented to the Ear Nose Throat (ENT) consultation for recurrent right lateral cervical infection. At the age of one year, the first infectious episode occurred in the form of an abscess of the soft tissues under angulo-mandibular without notion of otorrhea. The patient received local treatment with incision and drainage of the abscess and general antibiotic therapy without improvement. The course was characterized by fistulization of the skin and resulting from pus (Fig. 1). Clinical examination found an unsightly scar attached to an orifice giving pus located near the right mandibular angle suggesting type II first branchial cleft anomaly, no orifice at the level of the external auditory canal and the tympanic membrane was normal in appearance. The patient underwent ultrasound to obtain a diagnosis.

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Fig. 1. Picture of the child showing the right laterocervical cleft fistula.

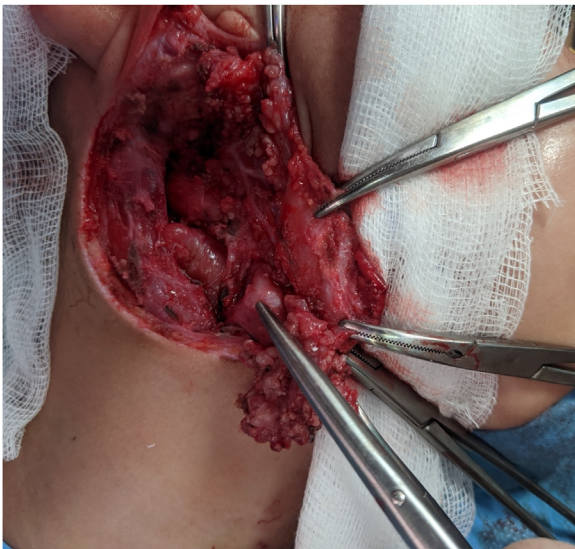


Fig. 2. Per-operative view of the right cleft fistula course with dissected facial nerve.

Surgical excision was performed under general anesthesia by a senior neck surgeon by an antegrade superficial parotidectomy approach, the facial nerve was identified and preserved. The fistula cord was dissected and followed, it went under the facial nerve and the parotid gland to end under the digastric muscle where we tied it up (Fig. 2). The postoperative check-up did not show any complications. The follow-up period was 12 months; the clinical examination did not find any sign of recurrence.

### 3. Discussion

Anomalies from the first branchial arch accounted for only 8% of all branchial cleft anomalies at the Mayo Clinic. Of these, 68% were cysts, 16% sinuses and 16% fistulae [7]. In general, sinuses and fistulae tend to develop in infants and children, whereas cysts are more common in older groups. FBCA occur more frequently in females (69%) compared with males (31%). The lesions are more likely to occur on the left side [8]. In our case, it was a little girl and the lesion was on the right side.

Liston proposed that the type II first branchial cleft anomaly may lie superficial to, deep to, or between the branches of the facial

nerve. The embryologic basis of this finding depends on the variable relationship between the migrating facial musculature, as it passes forward from the second branchial arch to the face, and the epithelial elements which persist in the region of fusion between the inferior portions of the mandibular and hyoid arches [9]. Which one develops first determines the relationship of the facial nerve with the FBCA. If the facial musculature migrated inferior to the first cleft, the facial nerve would be deep to the lesion. The case of our patient demonstrated during the surgery a fistulous path under the facial nerve and which ends under the digastric muscle.

Clinically, they may masquerade as parotid tumours or as otitis with ear drainage. First branchial cleft anomalies can be a diagnostic challenge and are often misdiagnosed. The typical clinical presentation of FBCA is purulent drainage from the external auditory canal (EAC) or swelling in the parotid area or postauricular area. A complete history and physical examination is the initial step in arriving at a diagnosis. Otoscopy should be performed to assess for any communication with the external auditory canal or attachment to the tympanic membrane. Facial nerve function should be recorded. Cervical skin should be closely examined for signs of a pit or sinus tract. Almost all FBCA occur in the Pochet's triangle area, which consists of the EAC, the hyoid body and the mandibular angle, particularly in the retroauricular groove or parotid region.

The treatment of first branchial cleft anomalies is surgical, and complete excision is necessary to prevent recurrence. In the majority of cases, definitive surgery requires a superficial parotidectomy approach with identification and preservation of the facial nerve. The surgery for type II is generally more difficult than that for type I because of his close relationship with the facial nerve. A previous study showed a high (22%) incidence of facial nerve palsy even with prior identification of the facial nerve [10].

### 4. Conclusion

First branchial cleft fistula are rare and can be in form of cyst or fistula. Its management is surgical excision keeping the tract cyst of the fistula intact with facial nerve preservation.

### Declaration of competing interest

The authors declare no conflict of interest.

### Funding

This study did not receive any sources of funding.

### Ethical approval

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

### 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.

### Author contribution

A. Chaouki : drafting the article  
 M. Lyoubi : acquisition of data  
 M. Lahjaouj : study concept  
 S. Rouadi : revising the article  
 M. Mahtar : final approval

**Registration of research studies**

Not applicable.

**Guarantor**

A. Chaouki.

**Provenance and peer review**

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**References**

- [1] K.D. Olsen, N.E. Maragos, L.H. Weiland, First branchial cleft anomalies, *Laryngoscope* 90 (1980) 423–435.
- [2] H. Arndal, P. Bonding, First branchial cleft anomaly, *Clin. Otolaryngol. Allied Sci.* 12 (1996) 203–207.
- [3] Jonathan A. MacLean, Steven E. Sobol, Congenital malformations of the neck, in: *Congenital Malformations of the Head and Neck*, Springer, 2014 (p. 162).
- [4] Wanpeng Li, Liming Zhao, Hongming Xu, Xiaoyan Li, First branchial cleft anomalies in children: experience with 30 cases, *Exp. Ther. Med.* 14 (July (1)) (2017) 333–337.
- [5] W.P. Work, Newer concepts of first branchial cleft defects, *Laryngoscope* 82 (1972) 1581–1593.
- [6] R.A. Agha, M.R. Borrelli, R. Farwana, K. Koshy, A. Fowler, D.P. Orgill, For the SCARE Group, The SCARE 2018 statement: updating consensus surgical CAse REport (SCARE) guidelines, *Int. J. Surg.* 60 (2018) 132–136.
- [7] K.D. Olsen, N.E. Maragos, L.H. Weiland, First branchial cleft anomalies, *Laryngoscope* 90 (1980) 423–436.
- [8] A.R. D'Souza, H.S. Uppal, R. De, H. Zeitoun, Updating concepts of first branchial cleft defects: a literature review, *Int. J. Pediatr. Otorhinolaryngol.* 62 (2002) 103–109, [http://dx.doi.org/10.1016/S0165-5876\(01\)00612-7](http://dx.doi.org/10.1016/S0165-5876(01)00612-7).
- [9] Stephen L. Liston, The relationship of the facial nerve and first branchial cleft anomalies—embryologic considerations, *Laryngoscope* 92 (November (11)) (1982) 1308–1310.
- [10] S.L. Liton, The relationship of the facial nerve and first branchial cleft anomalies—embryologic considerations, *Laryngoscope* 92 (1982) 1308–1310.

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