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Case Series

Surgery Section

Evaluation of Surgical Outcomes of First Branchial Arch Anomalies: A Case Series

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ABSTRACT

First Branchial Arch Anomalies (FBAA) are rare congenital conditions that present as cysts, fistulas, or abscesses. Failure to suspect the nature of the presenting lesion often leads to inadequate treatment and compromises the outcomes of definitive procedures undertaken later. In the present case series, eight patients with FBAA were identified (4 males and 4 females) with an average age of 29.5 years at presentation. Six of them had undergone prior procedures, including incision and drainage or excision. All patients underwent excision through a parotidectomy approach. Four patients experienced recurrences within a median period of 17 months post-surgery. Notably, three of the recurrences occurred in patients who had prior attempts at excision rather than merely incision and drainage. A high degree of suspicion in young individuals with cystic lesions in the parotid space, imaging to delineate the extent of the lesion, and an attempt at complete excision are key factors in successfully treating these conditions. Long-term follow-up is required to ensure the adequacy of the treatment strategy.

Keywords: Branchial cleft, Parotidectomy, Recurrence, Work classification

INTRODUCTION

Anomalies of the first branchial arch are rare and constitute approximately 19-31% of all branchial arch anomalies [1,2]. They encompass various presentations, such as soft cystic swellings, sinuses, and fistulae. Incomplete involution of the branchial apparatus during embryogenesis is the primary pathology in branchial arch anomalies [2,3]. FBAA present as cysts in the region of the parotid, postauricular region, or upper neck, with sinus openings along the anterior border of the sternocleidomastoid muscle in the upper neck. These anomalies are closely related to the External Auditory Canal (EAC), which is derived from the dorsal portion of the first branchial groove [4]. Consequently, the internal openings of these fistulas can occur in the region of the EAC, middle ear cleft, or postauricular region.

Work WP et al., Olsen KD et al ., and Belenky WM, et al., have proposed various classifications, but the validity of these classifications has been questioned, as authors have struggled to categorise the cases [3-5]. Key considerations for management include early recognition of the anomaly in the clinical setting, preservation of the facial nerve during surgery, and awareness of potential fistulous communications with the EAC, middle ear, or Eustachian tube [6,7]. Despite adequate care, these conditions have a higher tendency to recur over long periods of follow-up. Recurrences have been attributed to the altered anatomy following prior surgical interventions, which constitute a major subset of FBAA cases [1].

CASE SERIES

A case series of patients with FBAA who were treated by the General Surgical unit specialising in head and neck surgery were included. Patients for whom surgery was not performed or who underwent surgery for an unrelated cause were excluded. Inpatient case records were used to identify demographic data, age at onset, location of the lesion, and any prior history of surgical intervention. Operation records were reviewed to identify the course of the cyst or sinus tract, its relationship to the facial nerve, and any identifiable internal openings. Imaging reports were evaluated for consistency with intraoperative findings. Follow-up details were gathered for any morbidities and the date of the last outpatient visit. Patients were contacted via telephone, and respondents were asked about any recurrent swelling or discharge, as well as any further interventions performed elsewhere.

For uniformity, the exact location of the cyst or sinus was recorded according to the data categories outlined by Liu H et al. [8]. The Work classification was not indicated in some records, so these details were inferred from the operative, histological, and radiological findings during the chart review.

This case series identified nine patients diagnosed with FBAA. However, upon closer examination, one patient was excluded as they did not meet the histopathological diagnostic criteria. In this case, the final biopsy report did not demonstrate any sinus tract, and it was attributed to a parotid fistula secondary to a surgery performed in childhood for an unspecified cause. It remains speculative whether the original surgery was conducted for a congenital condition, possibly an FBAA, with a subsequent complication of a parotid fistula.

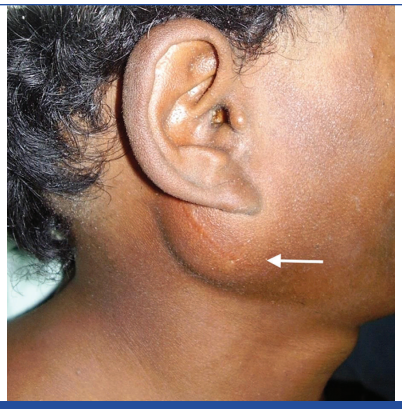
Demographics: The mean age of the study population was 29.5±10.7 years (range: 17-46 years). In the present case series, there was an equal gender distribution with 4 males and 4 females. The average age of presentation for female patients was 28.3±11.8 years, while that of male patients was 30.8±11.1 years [Table/Fig-1].

Demographic variables	Values
Gender	
Female	4
Male	4
Mean age at onset of symptoms in years±SD (Range)	18±11.5 (7-41)
Female	16±8.9
Male	20±14.8
Mean age at presentation in years±SD	29.5±10.7
Female	28.3±11.8
Male	30.8±11.1
Laterality	
Left	5
Right	3
Mean age at onset based on side	
Left	12±5

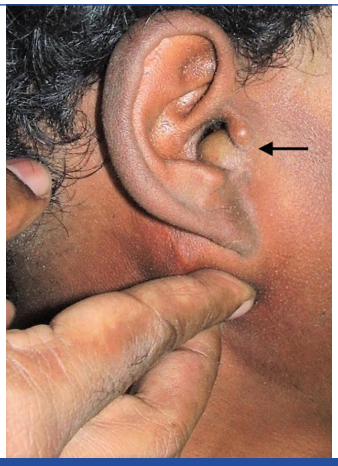
Right	21.6±13.3
Previous surgeries (%)	6 (75)
Incision and drainage	3 (37.5)
Excision of sinus tract	3 (37.5)

[Table/Fig-1]: Distribution of the FBAA (n=8).

Two of the eight patients did not have any previous interventions- one patient presented with a cyst in the parotid region, and another with a postauricular sinus. The other six patients had prior procedures: three underwent incision and drainage (single-2 patients /multiple-1 patient), while three had excisions of the sinus tract (single-2 patients/multiple-1 patient) at other centers, where the diagnosis of first branchial arch anomaly was likely missed [Table/Fig-2,3]. As part of the evaluation, imaging studies were performed on seven patients [Table/Fig-4,5].



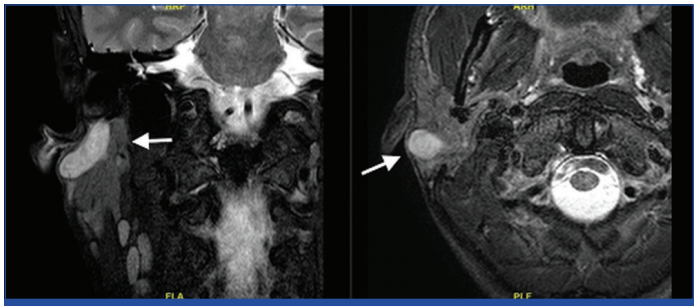
[Table/Fig-2]: Swelling in the right parotid region with the scar of previous surgery (white arrow) (Case no.1).



[Table/Fig-3]: The sign of cross-fluctuation (black arrow) (Case no.1).

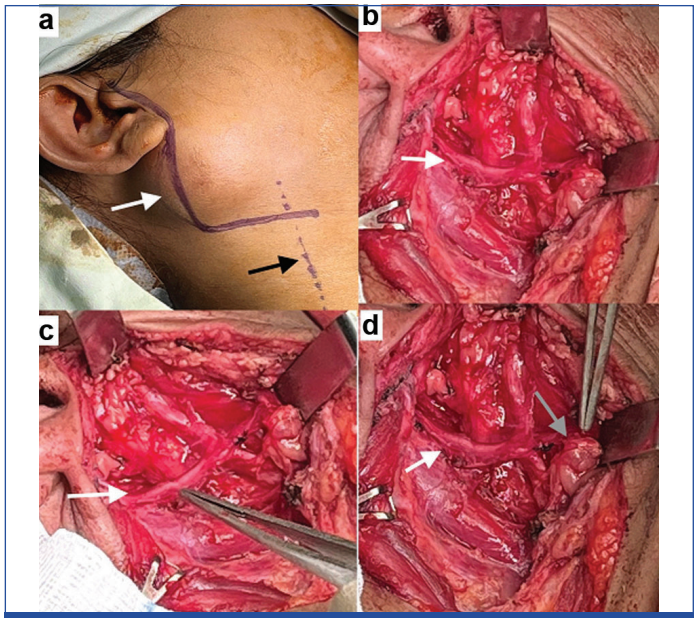
Case no.	Imaging	Brief findings	Work classification [3]	Olsen classification [4]
1	Nil		2	cyst
2	USG	Ill-defined linear hypoechoic area in the parotid gland	2	sinus
3	MRI	Well-defined cystic lesion just posterior to the left parotid gland	1	cyst
4	MRI	Thin long TR hyperintense fistulous tract seen from the inferior pole of the cyst extending down and laterally to open in the skin surface at the inferior pole of the right parotid gland	1	sinus
5	MRI	Tubular T2W hyperintense lesion in the right parotid extending towards the external meatus	2	sinus
6	CT	Heterogenous area with intermittent low-density areas; associated thickening of the lower part of the right pinna	1	fistula
7	MRI	Multiloculated lesion in the subcutaneous plane, extending till the posterior border of the external acoustic meatus.	1	sinus
8	MRI	Cyst within the parotid gland inferior to external auditory meatus	2	cyst

[Table/Fig-4]: Imaging studies and classification types in patients with FBAA (n=8).
USG: Ultrasound sonography; MRI: Magnetic resonance imaging; CT: Computed tomography



[Table/Fig-5]: MRI images showing a T2-Weighted (T2W) hyperintense lesion in the right parotid extending towards the external meatus with no definite opening into it. (Case no.5).

Surgery: All patients underwent excision using a parotidectomy approach with a modified Blair incision. The external location of the cyst or sinus was located in the parotid region in four cases and was postauricular in four cases. In one case, the tract led into the EAC, while the others had no fistulous connections. The relationship of the tract to the facial nerve was reported in six cases, all of which were superficial [Table/Fig-6].



[Table/Fig-6]: a) Marking of the Modified Blair's incision, External jugular vein marked in dotted line; b) Exposure of the facial nerve with its branches; c) Main trunk of the facial nerve; d) Relation of the branchial cyst to the facial nerve (Case no.8).

Pathology: All the tracts were submitted for histological examination. The lining epithelium was stratified squamous epithelium in five cases, respiratory epithelium in one case, and inflammatory granulation in two cases. One of the patients with stratified squamous epithelium exhibited pilosebaceous structures in the cyst wall [Table/Fig-7].

Outcomes: All patients, except one, were followed up for at least three years or until the recurrence of symptoms. The median follow-up period for the entire group was 30.5 (IQR: 5.25-99.5) months. One patient, who was non-compliant with follow-up, was symptom-

Case no.	Biopsy report	Lining epithelium
1	Retroauricular cyst with chronic inflammation	Inflammatory granulation tissue and foreign body granulomas
2	Branchial sinus with ulceration and inflammatory granulation tissue	Stratified squamous epithelium
3	Branchial cyst	Respiratory epithelium
4	Branchial cyst with a fistulous tract	Keratinised stratified squamous epithelium with pilosebaceous structures
5	Sinus tract with inflammatory granulation tissue	Inflammatory granulation tissue
6	Epithelial cyst with chronic inflammation	Stratified squamous epithelium
7	Branchial cyst	Stratified squamous epithelium
8	Branchial arch cyst	Stratified squamous epithelium

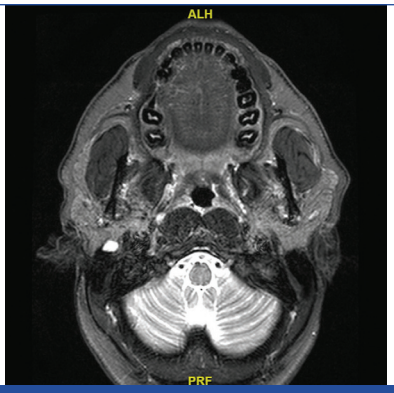
[Table/Fig-7]: Histopathological examination findings in patients with FBAA (n=8).

free at the last visit three months post-operation (Case No.2). In this series, four patients developed a recurrence of symptoms, with a median time to recurrence of 17 (IQR: 5.25-92.5) months. Three patients (75%) experienced recurrences within two years, whereas one patient developed a recurrence after nine years. The various characteristics of this group are shown in [Table/Fig-8].

Case no.	Surgery done	Location of cyst	Relation of the anomaly to the Facial Nerve	Internal opening	Morbidity	Recurrence
1	Excision	Parotid region	Superficial	No	SSI	Yes
2	Excision of sinus with superficial parotidectomy	Post auricular	Superficial	No	Temporary facial nerve palsy	No
3	Excision	Post auricular	Superficial	No	Nil	Yes (Second)
4	Fistula tract excision	Parotid		No	Nil	No
5	Excision of sinus with superficial parotidectomy	Parotid region		No	Nil	No
6	Excision of sinus	Post auricular	Superficial	EAC	Nil	Yes
7	Excision of sinus with underlying cyst	Post auricular	Superficial	No	Nil	Yes
8	Cyst excision with adequate parotidectomy	Parotid region	Superficial	No	Seroma	No

[Table/Fig-8]: Surgical approach, intra operative findings and outcomes in patients with FBAA (n=8).

The radiology team reviewed the MRI images of the patients with recurrences. Cases 1 and 3 did not have any postoperative imaging. Case No. 7 had a pre-auricular component that was incompletely excised. Case No. 6 underwent an incision and drainage before presenting at our center, and the follow-up MRI following surgery here showed a residual deeper parotid cystic lesion that was not fully excised, which communicated to an external opening in the postauricular region [Table/Fig-9].



[Table/Fig-9]: Postoperative MRI imaging of Case no.6 shows an 8 mm x 7mm T2W hyperintense lesion posterior to the deep lobe of the right parotid gland. Postoperative fibrotic changes in the skin in the post-auricular region (Case no.6).

DISCUSSION

The FBAA are rare and arise from buried cell nests in the ventral portion of the first branchial groove. They are commonly misdiagnosed at the initial presentation, leading to inadequate excision or drainage, which in turn complicates the surgical field, results in postoperative complications, and increases the likelihood of recurrence [6]. While Olsen KD et al., reported the prevalence of first arch anomalies among all branchial arch anomalies as 8%, recent series indicate this rate to be between 19% and 31% [1,2]. It remains unclear whether this discrepancy constitutes a referral bias.

There is a paucity of large single-institution studies on FBAAs. Liu H et al. described the features of first arch anomalies in a review of 277 cases [8]. The male-to-female ratio was 1:1.4, with a similar left-to-right distribution ratio of 1.14:1. The mean age in this series was 29.5 years, which is higher than in other reported series where children were treated [8,9]. However, this aligns with the mean age of 29 years in the series by Kumar R et al. [10]. Several factors may contribute to this, such as lack of access to a medical center, initial diagnostic delays, and the primary treatment being attempted by a non-specialist surgeon, in addition to the fact that this department typically treats patients older than 16 years.

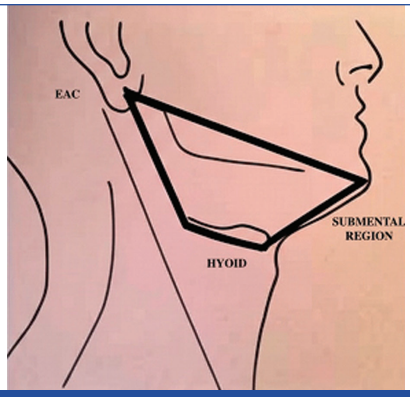
Patients with FBAA often undergo incision and drainage as their first procedure, especially when the cyst is inflamed. This is due to the uncertainty surrounding the diagnosis and the varied locations of the lesions. According to Poncet, Work Type 2 cysts present in a triangular region externally, bounded cranially by the EAC, anteriorly by the mental region, and inferiorly by the hyoid bone [11]. In the present series, most cases with prior attempts at excision developed recurrences; among those with recurrences, only one patient had undergone a prior incision and drainage.

Various classifications have been proposed to categorise FBAAs, but they offer limited guidance for clinicians in decision-making or benefit for patients [3,4]. The widely used Work classification categorises FBAAs into Type 1 and Type 2 lesions, which are distinct entities both anatomically and embryologically. Type 1 lesions arise from duplication of the EAC and occur externally to the postauricular crease, while Type 2 lesions represent a fistula from the upper neck region below the angle of the mandible down to the floor of the EAC [3]. Although useful, lesions can often be properly classified by Work's classification only after incorporating all details from clinical features, imaging, intraoperative findings, and histological reporting.

The clinical examination will reveal either a cystic swelling or a fistulous opening in the postauricular region (Type 1) or within Poncet's triangle (Type 2). Poncet's triangle is defined with the EAC at the apex and the base formed by an imaginary line between the chin and the midpoint of the hyoid bone, with the sides formed by lines connecting the EAC to the chin and the EAC to the greater cornua of the hyoid bone [Table/Fig-10] [12]. In recurrent cases, scars from previous operations in the region are evident.

A previously undescribed clinical finding was noted in Case No. 1. In this case, there was cystic swelling in the retromandibular region accompanied by an overlying scar from previous surgery. The swelling displayed cross-fluctuation in the posteroinferior cartilaginous EAC, consistent with Type 2 FBAA.

An otological evaluation is required when ear complaints are present. Patients typically show symptoms in the presence of fistulous communications with the EAC or middle ear. A myringeal web is



[Table/Fig-10]: Poncet's triangle bounded by the EAC posteriorly, mental region anteriorly and hyoid bone inferiorly. EAC at the apex, base formed by an imaginary line between the chin and midpoint of the hyoid bone, with the sides formed by a line joining from the EAC to the chin and from the EAC to the greater cornua of the hyoid bone.

an asymptomatic finding characterised by a fibrous band of tissue extending from the floor of the EAC to the tympanic membrane [5]. Sichel JY et al. have suggested that this finding, when present, is pathognomonic of Work Type 2 cysts, underscoring the importance of evaluating the ear for this diagnostic clue [11]. As none of the patients in the present series had any ear complaints, a detailed otological examination was not deemed necessary.

Cross-sectional imaging is highly recommended to delineate the track and fistulous openings when present. However, the crucial course of the facial nerve in relation to the cyst or fistula tract can, at best, be approximately predicted by mapping the tract/cyst location and its relationship to the stylomastoid foramen and Patey's neurovascular plane [13]. Most cases in our series underwent MRI, as it offers better delineation of the relationship between the facial nerve and surrounding soft tissue in patients with previous surgery [14]. This aids in surgical planning.

Nevertheless, it is prudent to recognise the basic principles of management, including the exposure and preservation of the facial nerve and a diligent search for otological fistulae. Surgically, the tract is approached via a parotidectomy incision, ensuring early exposure of the at-risk facial nerve trunk and its branches. The tract is completely excised, often requiring a parotidectomy and excision of any fistulous communication with the ear. The use of magnification (operating microscope or loupes) during dissection is recommended, especially for the exposure of the facial nerve [8,15].

A review of patients with post-excision recurrences highlights that they may have Work Type 2 lesions, which are deeper in the parotid gland and posterior to the facial nerve, or both preauricular and postauricular cystic lesions that need to be identified and addressed to prevent recurrence [3]. In patients with prior excision attempts, complications may arise due to distortion of natural tissue planes and scarring in the operative field. A careful review of preoperative imaging remains crucial to identify all components and achieve complete excision. The recurrence rate following surgical excision for FBAAs ranges from 6% to 70% in the available literature [8,13]. In the present series, recurrence of symptoms was noted in 50% of the cases over a variable duration, with some recurrences as late as nine and a half years post-surgery, although most cases recurred within the first two years. The symptoms of recurrence may be mild enough to deter patients from seeking further treatment. None of the patients with recurrences underwent any additional procedures at our center. Therefore, the need for close and adequate follow-up cannot be overstated.

CONCLUSION(S)

FBAA are rare and require a high index of clinical suspicion for early diagnosis and appropriate management. The best outcomes are achieved in first-time cases when the tissue planes have not been altered, as opposed to recurrent cases, which suffer from distortion of tissue planes and residual remnants of track components. Most recurrences occur within the first few years, but recurrent cases seldom return to the physician for further management. Long-term follow-up is recommended for late recurrences to draw meaningful conclusions regarding the efficacy of treatment strategies.

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