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CLINICAL RECORD

First branchial arch abnormality: diagnostic dilemma and excision with facial nerve preservation

P JOICE[1], T SUDARSHAN[2], S S M HUSSAIN[1]

Departments of [1]Otolaryngology and [2]Radiology, Ninewells Hospital and Tayside Children's Hospital, Dundee, Scotland, UK

Abstract

Objective: To report a case of first branchial arch abnormality and the problems associated with misdiagnosis. A succinct literature review is included.

Setting: Teaching hospital in Scotland.

Methods: A 10-year-old girl presented with localised erythema and swelling in the left parotid region. This was treated with antibiotics and incision and drainage. She re-presented four years later with a history of recurrent discharge. A first branchial arch abnormality was suspected and a magnetic resonance imaging scan arranged.

Results: Imaging showed a fluid-filled sinus tract originating adjacent to the anterior wall of the cartilaginous left external auditory canal. The sinus tract was seen to extend anteriorly and inferiorly through the superficial lobe of the left parotid, and to open onto the left cheek lateral to the left masseter. The tract was explored and excised under general anaesthesia, via two separate incisions, with preservation of the facial nerve.

Conclusion: The diagnosis of a first branchial arch abnormality is generally based on a high index of clinical suspicion, when a neck swelling is noted in a child. Magnetic resonance imaging is a useful modality for investigation, and helps to delineate the position of the tract and its relationship to the facial nerve.

Key words: Branchial Arch; Magnetic Resonance Imaging; Neck; Facial Nerve; Children

Introduction

First branchial cleft anomalies are rare. They are estimated to account for only 1–5 per cent of all branchial anomalies.[1–4] Due to their infrequent presentation, diagnosis can be delayed and initial management may be inappropriate.[5]

Case report

A 10-year-old girl presented with a 6-month history of localised erythema and swelling in the left parotid region (Figure 1). This was diagnosed as an isolated abscess. Aspiration was not considered appropriate in view of the patient's age. She was treated with intravenous co-amoxiclav together with incision and drainage under general anaesthesia. Culture and sensitivity testing of the drained pus demonstrated streptococci and anaerobes sensitive to co-amoxiclav. No sample was sent for histopathological analysis.

Review at three months showed some improvement in the area of inflammation (Figure 2). At nine months, it was concluded that the inflammation had settled, and the patient was discharged.

Four years later, the patient re-presented to the senior author with multiple further episodes of discharge and failure of resolution of the area of inflammation. In addition to discharge, the patient also described pain which radiated to the left ear.

A first branchial cleft abnormality was suspected, although the differential diagnosis included atypical mycobacterial and actinomyces infections.

The lesion was investigated with magnetic resonance imaging. This demonstrated a fluid-filled sinus tract originating adjacent to the anterior wall of the cartilaginous left external auditory canal. The sinus tract was seen to extend anteriorly and inferiorly through the superficial lobe of the left parotid, and to open onto the left cheek lateral to the left masseter, with inflammation of the surrounding skin (Figures 3 to 5).

The tract was explored and excised under general anaesthesia. Two separate incisions were used, one at the site of the external opening on the lower face, and the second a classical parotidectomy incision (Figure 6). The tract was found to extend medial to the facial nerve and to terminate in the medial aspect of the anterior cartilaginous external auditory canal. The tract was cartilaginous and formed a duplicate external canal (Figure 7). Following excision, the external ear canal was reconstructed. The facial nerve and its branches were preserved (Figure 8). The ear canal was packed with bismuth iodoform paraffin paste ('BIPP') and the wound closed.

The patient made an uneventful recovery.

Discussion

Branchial anomalies occur when there is disturbance in the maturation of the branchial apparatus during the fourth to eighth week of fetal development. In the normally developed fetus, the first branchial cleft gives rise to the external auditory canal and the lateral aspect of the tympanic membrane.

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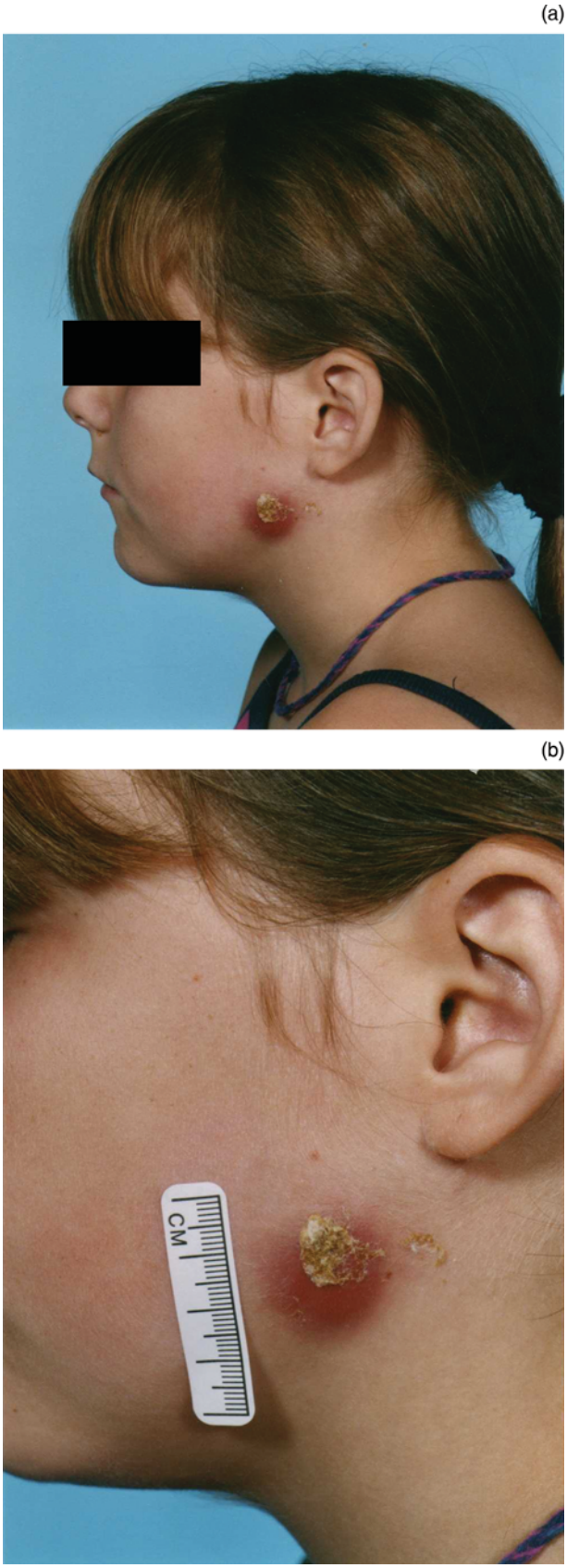

FIG. 1
Clinical photographs showing the skin inflammation at presentation.

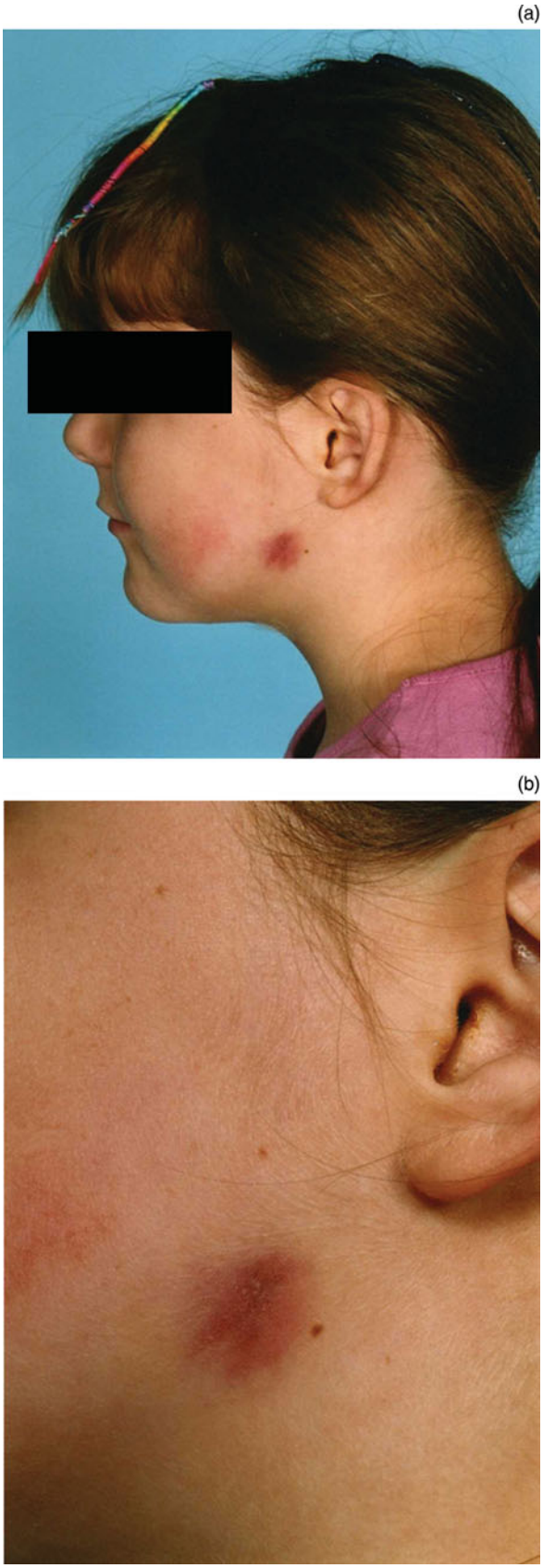

FIG. 2
Clinical photographs showing the skin inflammation at three-month follow up, after intravenous antibiotics, incision and drainage.

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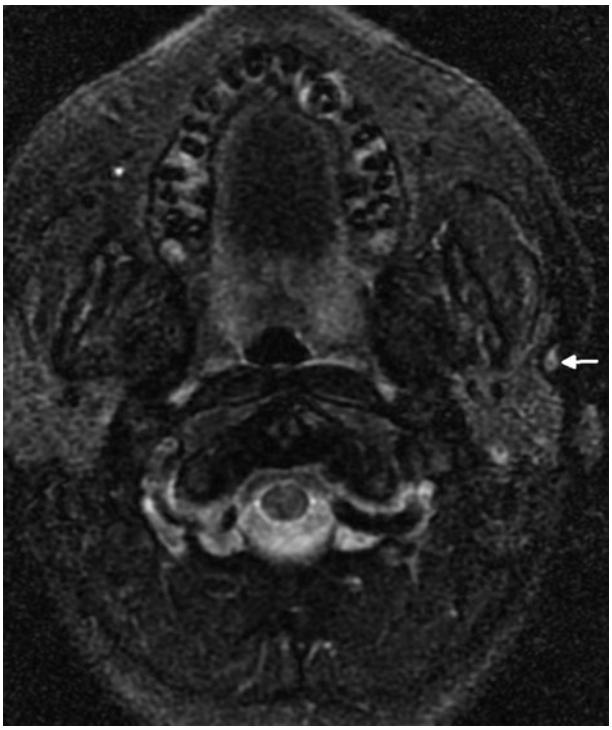

FIG. 3
Fat-saturated, T2-weighted, axial magnetic resonance imaging scan showing a fluid collection (arrow) anterior to the cartilaginous left external auditory canal.

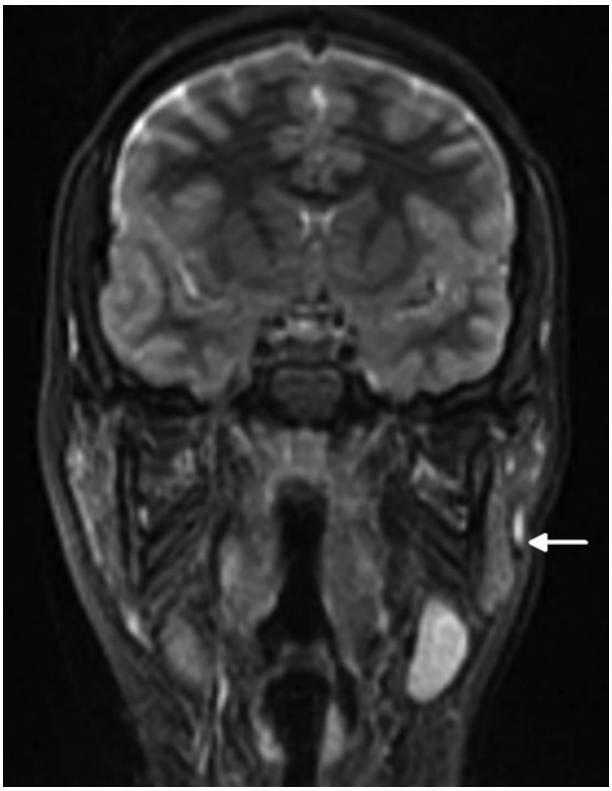

FIG. 4
Coronal, short T1 inversion recovery magnetic resonance imaging scan showing the sinus tract (arrow) extending along the superficial lobe of the left parotid.

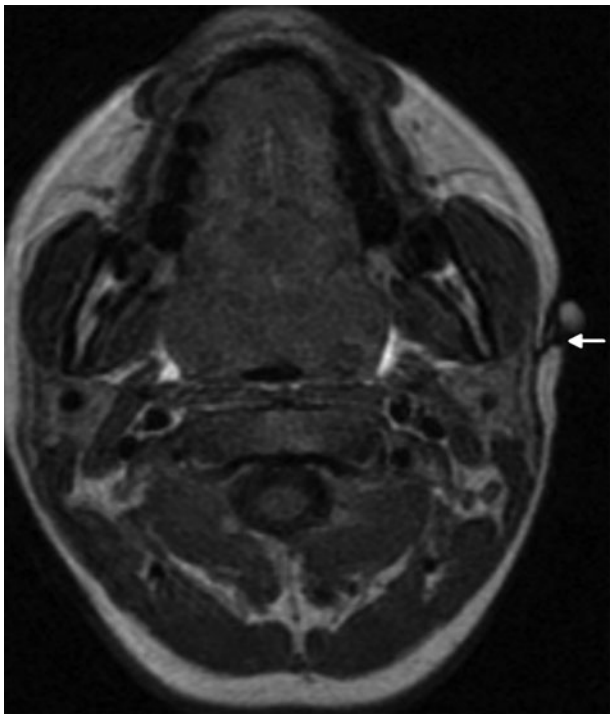

FIG. 5
Axial, T1-weighted magnetic resonance imaging scan showing the opening of the sinus tract in the skin (arrow).

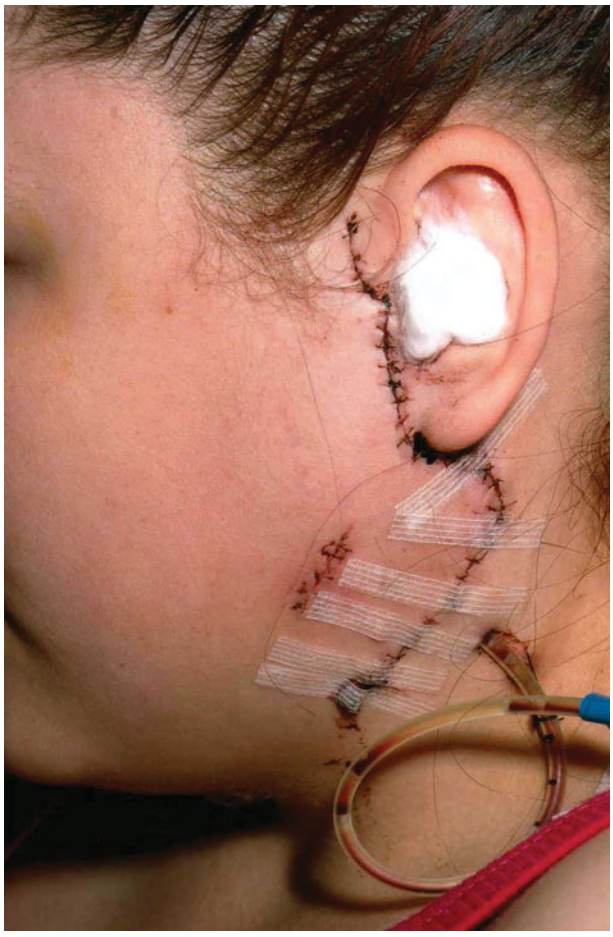

FIG. 6
Post-operative clinical photograph showing the two separate skin incisions.

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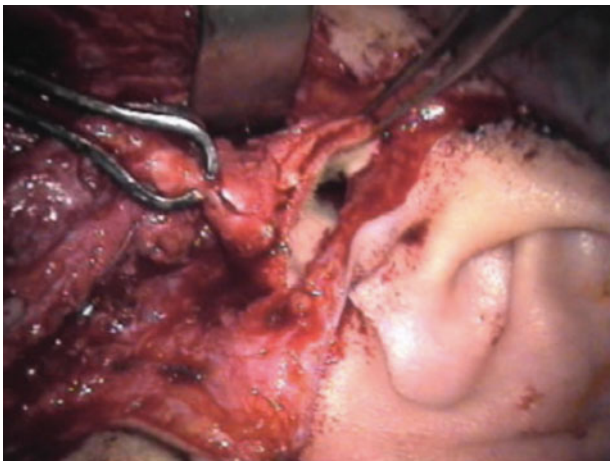

FIG. 7
Peri-operative photograph showing the tract and the cartilaginous duplicate ear canal.

Anomalies of the first branchial cleft have previously been classified as anomalies of aplasia, atresia, stenosis and duplication of the external auditory canal.[6] The tract excised in this case was a duplication anomaly.

Typically, duplication anomalies demonstrate a normally developed external auditory canal along with a sinus tract extending from the external auditory canal to the skin of the face or neck. The position of the skin opening of the tract characteristically lies on a virtual line joining the tragus to the hyoid, whilst the upper end of the tract runs inferior or anterior to the external auditory canal and terminates at the osteo-cartilaginous junction.[2] The course of the sinus tract is unpredictable and can run medial or lateral to the facial nerve; in addition, the course of the facial nerve and relevant landmarks may differ in children compared with adults.[7]

Duplication anomalies have been subclassified by Work into types one and two.[8] This categorisation differentiates between tracts which arise from purely ectodermal material (type one) and tracts which in addition contain mesoderm-derived material in the form of a cartilaginous component. Case reports of duplicate tracts that do not fit within this classification (i.e. containing duplicate ossicles) have also been published.[9] The tract in our case was of the second type, which is also the more common type.

Magnetic resonance imaging, with its superior soft tissue resolution, is the investigation of choice for accurate delineation of branchial sinus tracts, before contemplating surgery. Fluid-filled sinus tracts show high signal intensity on T2-weighted images and short T1 inversion recovery ('STIR') images. The presence of an associated cyst is usually indicated by a well defined, rounded area of fluid signal.

- **Diagnosis of first branchial arch anomalies requires heightened clinical suspicion of childhood neck swellings**
- **Magnetic resonance imaging delineates tract position and facial nerve relationship**

The presentations of published first branchial cleft anomaly cases vary. They include otalgia, aural discharge, and as an

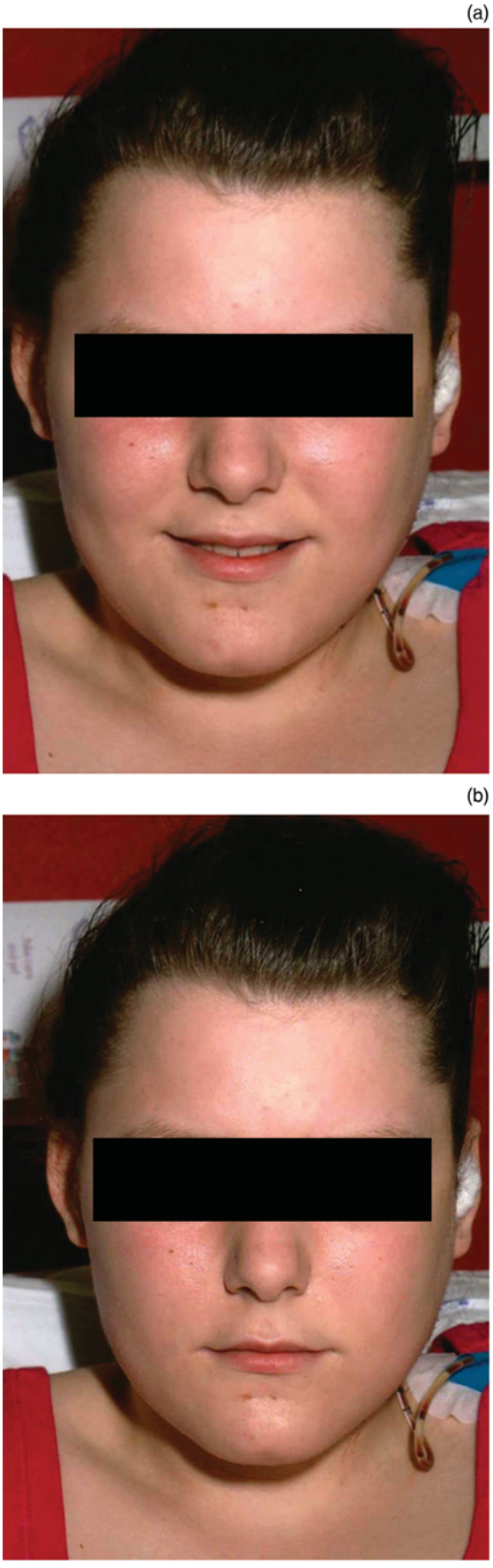

FIG. 8
Clinical photographs showing post-operative facial nerve function.

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incidental finding on imaging or otoscopic examination. However, the most common presentation is recurrent swelling, inflammation and discharge from a skin lesion. The differential diagnosis includes atypical mycobacterial and actinomyces infections. The former usually presents as unilateral cervical lymphadenopathy with altered skin coloration.[10] The latter is characterised by the formation of abscesses, fibrosis and woody induration of the tissues, and by draining sinuses that discharge 'sulphur granules'.[11]

This case demonstrates such a presentation. It also emphasises the fact that clinicians should maintain a high index of clinical suspicion for the possibility of a sinus tract when dealing with a neck swelling in a child, in order to avoid inappropriate initial management.

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Address for correspondence:
Mr S S Musheer Hussain,
Consultant Otologist & Neurotologist,
Ward 26,
Ninewells Hospital and Tayside Children's Hospital,
Dundee DD1 9SY, Scotland, UK

Fax: +44 (0)1382 632 816
E-mail: musheer.hussain@nhs.net

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