



## Fourth branchial cleft anomaly: Management strategy in acute presentation



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### ARTICLE INFO

#### Article history:

Received 3 March 2014

Received in revised form 16 May 2014

Accepted 11 June 2014

Available online 20 June 2014

#### Keywords:

Fourth branchial sinus

Acute management

### ABSTRACT

**Objectives:** Branchial malformations are common congenital head and neck lesions usually diagnosed in childhood during the first decade of life. Acute presentation is usually managed with conservative protocols before a definitive surgical procedure although the risk of life-threatening septic complications may influence the physician's decision. Surgery is the treatment of choice with the removal of the lesion alone, nevertheless more aggressive approaches must be considered in complicated cases. Selective neck dissection including the removal of part of the thyroid lobe with the congenital lesion should be considered as the "ultima ratio" treatment to avoid recurrence.

**Methods:** We reviewed literature and report our experience concerning two patients with fourth branchial cleft sinus.

**Results:** A three-year-old child with a clinical history of recurrent neck abscess was referred to our department after several drainages performed in another centre. A three-year-old child referred to our department for a left side lower primary neck abscess. In both cases the diagnosis of a complicated fourth cleft remnant was confirmed by rigid endoscopic visualization of the mucosal orifice of the sinus in the pyriform fossa. Surgical management during acute presentation was challenging; in one patient the early fasciitis required an emergency procedure to remove the infected sinus that were strictly adherent to the deep vascular-nervous axis.

**Conclusion:** Surgery was the definitive treatment in both cases and at 12 and 25 months follow-up respectively no recurrences were observed.

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### 1. Introduction

Branchial malformations are common congenital head and neck lesions, usually diagnosed in childhood, during the first decade of life. Ascherson [1] linked the development of lateral cervical cysts with the presence of branchial remnants (*am vero disquiramus utrum proprietates fistularum colli . . . expositae conjectura illa, fistulas fissurarum branchialium residua esse facile explicari possint*). Branchial remnants are classified as cysts, sinuses and fistulas of first, second, third and fourth cleft, according to the arch of origin [2] and they are usually lined by squamous epithelium [3]. First and second cleft anomalies are more frequent and comprise more than 96% of all branchial lesions [4], while anomalies of third and

fourth cleft are rare (1–4%) with the largest single-centre series of 18 patients was reported by Madana et al. [5]. The anatomic difference between malformations of third and fourth cleft is based on the origin from the pyriform fossa: third branchial arch anomalies originate from the base (cranial end) of the pyriform fossa and pass above the superior laryngeal nerve, while fourth branchial sinuses or fistulas originates from the apex (caudal end), passes through the cricothyroid membrane beneath the superior laryngeal nerve [6] and result in the formation of sinus tracts or cysts which occur in close proximity to the thyroid gland [7]. The sinus or fistula's tract is normally short and it opens in the hypopharynx, behind the thyroid ala, caudal to the superior laryngeal nerve and cranial to the recurrent laryngeal nerve, in proximity to the posterior wall of the thyroid gland, ending at the thyroid gland or in para-tracheal position [3]. Its cervical position explains the possible complications in case of infection or surgical procedures. The clinical presentation is often acute [3,4], and differential diagnosis with infected neck nodes, hemangiomas, thyroid cysts, ectopic thyroid and cervical thymic cysts is mandatory. Imaging is useful in characterizing these anomalies

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**Fig. 1.** Patient 1: swelling of the lower left cervical area with evidence of scar due to previous skin incisions.

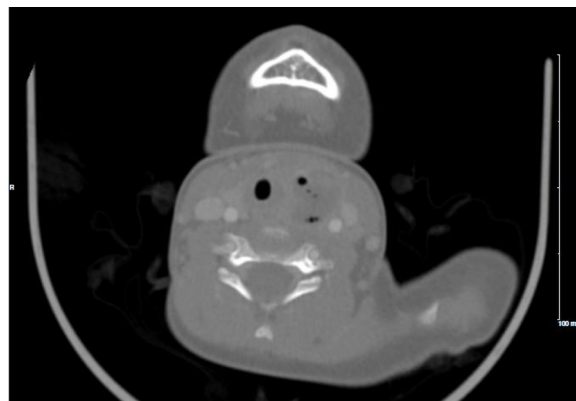
and different modalities have been described: contrast-enhanced computed tomography (CT) scan or magnetic resonance imaging (MRI) are mostly accepted to define the mass and to exclude a thyroiditis or a suppurative lymphadenitis. The identification of the exact position of the tract of the sinus or fistula can be complex or even impossible by imaging due to its small diameter associated to inflammatory changes of surrounding tissues and, often, his presence is only assumed. Treatment consists in antibiotic therapy followed by surgery. We report our experience on two cases which presented neck abscess due to IV branchial cleft sinus, with a review of the pertinent literature, aiming to contribute in defining the adequate treatment and definitive strategy, especially during the acute presentation, when physicians face to a high risk of septic complications.

## 2. Methods

Retrospective review of the clinical charts of two patients operated for a fourth branchial left cleft sinus between 2010 and 2012 at the ENT department of the University of Cagliari (Italy). Data are presented according to age, associated signs and symptoms, perinatal diagnosis and management, preoperative evaluation, surgical treatment, and outcome. We also performed a review of the literature concerning the fourth branchial cleft sinuses and fistulas.

### 2.1. Case report 1

A 3-year-old male was referred for evaluation of a chronic inflamed neck mass (Fig. 1). Three previous skin incisions for drainage of a recurrent left side neck abscess were performed in another hospital. Contrast-enhanced CT scan revealed an abscess on the left side of the neck, lateral to the laryngo-tracheal axis, postero-superior to the thyroid gland involving the superior pole of the left thyroid lobe. A fourth branchial cleft sinus was suspected due to the recurrent characteristic of the mass and the patient underwent panendoscopy under general anaesthesia. The diagnosis was confirmed by the identification of the orifice of the sinus in the left pyriform fossa. The sinus was removed along with the cervical fascia in form of a selective neck dissection of levels III and IV, including the scar tissue resulting from the previous neck incisions. The superior pole of the left thyroid lobe was spared, although it was strictly adherent to the lesion. The orifice of the sinus was sutured with two planes of 3/0 absorbable suture. Histological examination confirmed the diagnosis showing the presence of malpighian epithelium lining the tract along its path as



**Fig. 2.** Patient 2: axial contrast-enhanced CT scan image of a left cervical hypodense lesion, suggestive of an abscess, with the presence of air, adjacent to the thyroid gland.

well as the pouch. The postoperative course was uneventful and the patient was discharged after 4 days. At 2 years follow-up no recurrence was observed.

### 2.2. Case report 2

A 3-year-old girl was referred to our department. The patient was previously hospitalized for 7 days in the Department of Paediatrics after a long history of enlarging neck mass associated to fever, dysphagia, pain and hyperaemia of the overlying skin. Contrast-enhanced CT scan revealed an abscess on the left side of the neck, lateral to the trachea, extensively involving the left lobe of the thyroid gland, below the strap muscles (Figs. 2–4). Diagnosis was hypothesised in view of the history of persistent neck mass and it was confirmed through rigid endoscopy (0°) under general anaesthesia, that allowed direct visualization of an orifice on the antero-medial wall of the left pyriform fossa (Fig. 5). The patient underwent antibiotic therapy with ceftriaxone 1 g every 24 h and metronidazole 200 mg every 8 h, switched after admission in our department to tazocilline 500 mg every 8 h, in order to reduce the infection before surgical treatment. Despite seven days of i.v. full-dose antibiotic therapy and two needle drainages, the swelling was still recurring, fever did not decrease and the general status of the patient was deteriorating; therefore, after counselling with patient's parents, the surgical management was considered necessary and not to be delayed. A trans-cervical removal of the



**Fig. 3.** Patient 2: sagittal contrast-enhanced CT scan image illustrating a low cervical abscess.

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