Case report

Left sinus pyriform fistula treated by minimally invasive approach: A case report

Hicham Lyoubi, Hicham Ngham *, Youssef Oukessou, Sami Rouadi, Reda Allah Abada, Mohamed Roubal, Mohamed Mahtar

ENT and Head and Neck Surgery, Hospital August 20, 1953, Ibn Rochd Teaching Hospital, Casablanca, Morocco

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ABSTRACT

Introduction: Abnormalities of the fourth branchial arch are less common than those of the second arch and usually present with inflammation of the left thyroid lobe.

Case presentation: We report the case of a 10 years old girl who presented to our department with recurrent cervical cellulitis, and who was diagnosed, upon endoscopic exploration, with a left sinus pyriform fistula. The patient was treated using mini-invasive surgery by electrocoagulation, with good clinical outcome.

Conclusion: Branchial arch malformations are rare congenital malformations. The diagnosis is mainly based on clinical examination, imaging and, endoscopic investigations. The conservative attitude may be the treatment of choice, especially if the cervical mass is not well individualized.

1. Introduction

The most common developmental abnormalities of the branchial arch are the second cleft, which constitutes almost 95% of all abnormalities [1]. Abnormalities of the third and fourth branchial arch, although rare, usually present as incomplete sinuses or pyriform sinus fistulas (PSF) [2]. This congenital anomaly manifests as recurrent cervical abscesses or acute suppurative thyroiditis. A cervical mass is usually present during the first decade of life, more frequently on the left than on the right. Therefore, a diagnosis of pyriform sinus fistula should be suspected in all pediatric cases of recurrent left neck abscess or acute suppurative thyroiditis [3].

As a result, there are no consistent recommendations on diagnosis and management, and reports of recurrence and complication rates vary widely.

The remains of the fourth arch are extremely rare reported here is a case for its rarity and late presentation. This work is reported by following the surgical case report (SCARE) guidelines [4].

2. Case presentation

A 10 years old girl with no drug history or similar case in the family, including any relevant genetic information, and psychosocial history, presented with a history of sore throat for 2 months with difficulty swallowing associated with fever. Clinical examination revealed a progressively increasing left cervical swelling in the lower part of the sternocleidomastoid muscle, which was fluctuating and tender (Fig. 1), the temperature was 38.5°C.

Ultrasound examination, made on admission, shows a thyroid gland increased in size of the left lobe measuring $37 \times 31 \times 22$ mm with blurred outlines; thickening of the SCM muscle and the presence of multiple jugulocarotid lymphadenopathies, initially suggesting a thyroid abscess or unilateral suppurative thyroiditis.

The child received an appropriate oral antibiotic treatment, amoxicillin-based 80 mg/kg/day intravenously for 10 days, with good clinic evolution, then reappearance of the mass a month later with marked general signs.

Computed tomography (CT) showed a thyroid gland of normal size, with the presence in its left lobe of an inferior polar lesion with irregular and hypodense contours, enhanced after injection of PDC with infiltration of adjacent fat associated with a mangle of lymphadenopathy of territories III and IV probably of infectious origin (Fig. 2).

The patient was treated conservatively with antibiotics and pain relievers, and she was admitted to the operating room for hypopharyngoscopy under general anaesthesia, which showed a fistula from the bottom of the left pyriform sinus (Fig. 3).

Coagulation of the orifice of the fistula was performed...
endoscopically using bipolar forceps. The procedure was performed by an ENT professor. Patient perspective was good, compliance and tolerability of the intervention were favorable.

The postoperative follow-up at 12 months is unremarkable with good clinical outcome.

3. Discussion

Branchial apparatus anomalies include branchial cysts, fistulae, and sinuses. The branchial sinuses can open externally or internally. Second arch anomalies are the most common, followed by first and third. The fourth arch anomaly is very rare, and few reports are available in the literature [5].

Pyriform sinus fistula represents only 3–10% of all branchial malformations [6].

Correct diagnosis is essential to avoid inadequate operation for newborns and children.

In children, the diagnosis requires a high index of clinical suspicion in the face of left-sided neck abscess [7].

A CT scan may be performed after the barium swallow (in cases of suppurative thyroiditis); however, a direct contrast injection (in cases of neck fistula) through the cannulated external opening will be more useful in delineating the anatomical course of the entire tract than a plain CT scan [8].

Computed tomography is also necessary to exclude other lateral cervical pathology and to guide the plane and direction of fistulectomy [2].

Endoscopy is useful for diagnosis, and should be performed under general anesthesia [2].

There are no reports regarding the carcinogenic risk in PSF, but the coexistence of thyroid cancer in an adult with PSF has been reported [9].

In cases of suppurative thyroiditis, incision and drainage must be performed on an emergency basis; but, this makes the subsequent surgery more difficult. However, complete excision of the fistulous tract with a partial or hemithyroidectomy, during a quiescent period, is the treatment of choice [2].

Many reports have described recurrences and complications in open-neck surgery include recurrence, wound infection, vocal cord motion impairment/vocal cord paralysis, salivary fistula, Horner syndrome, injury to the recurrent or superior laryngeal nerves, and facial nerve paralysis [10].

Recently, as a minimally invasive surgery for PSF, endoscopic surgery for the opening of the fistula has been reported [11].

This approach is difficult for the treatment of PSFs in children because the working spaces is small and narrow [10].

In chemocauterization of the fistula tract, trichloroacetic acid or silver nitrate has been used; however, albuminoid degeneration in tissues causes scar formation [12].

Sealing materials, such as Glubran 2 or a combination of Tissucol and Deflux, have also been used during endoscopic surgery [13].

Electrocauterization or laser cauterization have been performed [14], but these methods carry the risk of pyriform sinus mucosa penetration [11].

In our case a Coagulation of the fistula orifice was performed endoscopically without any complications over a follow-up period of 3 months.

4. Conclusion

The fourth branchial pouch is rare and is found along the left thyrotracheal axis. The diagnosis of fourth branchial pouch fistulas can be confirmed by endoscopic visualization of an opening in the bottom of the pyriform sinus. The relationship between the abnormality and the surrounding structures can be verified by ultrasound and computed tomography.

The introduction of the technique of endoscopic diathermy at the opening of the pyriform fossa sinus in children with anomalies of the fourth gill pocket has revolutionized their management by avoiding open and potentially morbid surgery.

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Ethical approval

I certify that this kind of manuscript does not require ethical
approval by the Ethical Committee of our institution.

Consent

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

CRediT authorship contribution statement

XX: conception and design of the study.
XX: conception and design of the study.
XX: acquisition of data.
XX: drafting the article.
XX: drafting the article.
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Registration of research studies

This is a Case report that does not require a research registry.

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Declaration of competing interest

The authors declare that they have no competing interests.

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