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Case Report

Branchial arch anomalies: need for early diagnosis

Jayita Poduval*

Department of ENT, Pondicherry Institute of Medical Sciences, Pondicherry, India

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***Correspondence:**
Dr. Jayita Poduval,
E-mail: drjayitapoduval@gmail.com

ABSTRACT

Branchial fistula and similar anomalies may present in childhood; the diagnosis may not be obvious at initial presentation. The treatment is by surgical exploration and excision. Misdiagnosis results in recurrence and considerable morbidity. This case report is an example of a missed left branchial fistula which was later managed successfully.

Keywords: Branchial Fistula, Diagnosis, Surgical excision

INTRODUCTION

Branchial fistulae are disorders of embryological development in which there is incomplete fusion of any of the elements of the branchial arches that make up the tissues and organs of the head, face and neck. They may present as sinus openings or cystic swellings in which case the diagnosis is rather straightforward. In a few cases, misdiagnosis occurs because the lesion is extremely rare, leading to wrong management and recurrence, often causing great morbidity. We present here the case of a young girl who was treated as a case of thyroid abscess but eventually turned out to have a branchial fistula that was subsequently treated with good results.

CASE REPORT

A 17- year-old girl presented to us with the history of purulent discharge and an ugly scar over her neck from previous infection and surgery, wanting to get it cosmetically repaired. Her previous medical records mentioned an abscess over the neck a few years earlier which was diagnosed elsewhere as a tuberculous abscess of the thyroid and treated accordingly. A surgical exploration was subsequently performed but a second infection followed after a couple of months with swelling, pain and purulent discharge from the site.

An ultrasound of the neck was done but no abscess could be clearly identified; only a lymphadenopathy was reported. Aspiration yielded a minute amount of pus which was sent for smear and culture for mycobacteria but it was inconclusive.

The patient was discharged on broad- spectrum antibiotics and asked to review a week later.

Upon review, the infection had settled but this time the patient reported that fluid was escaping from the site when she drank water or milk, and a clinical suspicion of branchial fistula was reached.

A contrast X-ray and CT sinogram revealed a tract extending from the left pyriform fossa to the ipsilateral neck and the diagnosis of left fourth branchial arch fistula was confirmed. The patient was operated, the fistulous tract identified and removed and the scar revised. To date, approximately four months later, she continues to be symptom- free.

DISCUSSION

Incomplete development of the branchial arches results in non-fusion of the elements that make up the branchial clefts and pouches. Persistence of the branchial cleft results is the formation of a sinus that opens externally on to the skin or surface, while persistence of a pouch causes an internal sinus with a mucosal opening to form. Complete breakdown of both the cleft and the pouch of a particular arch without fusion of any of the elements give rise to a fistula that connects skin to mucosa. A cyst results if the ectodermal lining of the arch persists.

Fistulae, sinuses or cysts of the second arch are more common than the others, and make up more than 90% of such anomalies.[1] First arch anomalies are rare, as are those from the third and fourth arches. The latter may, particularly, present difficulties with diagnosis and are commonly mistaken for suppurative thyroiditis,[1] though this entity itself is rather uncommon. Left- sided fistulae are more common than those on the right.[2] A careful history taking is imperative but may still be misleading, as in our case, where the patient only reported that a previous diagnosis of tuberculosis had been made and that she had taken the prescribed course of anti-tuberculosis treatment. Further confusion occurred when the documents describing the previous treatment mentioned that a histopathological diagnosis of tuberculosis had been made but the histopathology report itself was not traceable. Repeated infection continued to take place and a corrective surgery, details of which were also unavailable, had probably obstructed whatever external opening there might have been and the patient initially denied regurgitation of fluids from the site. At the follow-up visit with us and having completed a fresh course of antibiotics for the most recent infection she reported the episodes of regurgitation, at which point the diagnosis became obvious.

To save time and expense and to clearly be able to delineate a tract and its relationship to adjacent structures, a CT sinogram was done. The tract was seen to be opening into the apex of the left pyriform sinus and a fourth branchial fistula was thus diagnosed. It was, however, not running through the carotid bifurcation and confirmed at surgery. This is contrary to the theory of development of such fistulae, which classically describes them passing through the bifurcation and thus proving to be surgical challenge. Similar findings have been reported by others.[3]

The internal opening of the fistula was also cauterised with 5% silver nitrate though what has been advocated by others, even as the first line of treatment, is chemocauterization with 40% tri- chloro-acetic acid.[2,4] A novel method of treatment using sclerotherapy has also been mentioned.[5] These methods may prove successful if a correct diagnosis has been reached at initial presentation. Surgical extirpation remains the definitive treatment and may include revision of the scar whenever required.

Though uncommon in the case of fistulae, squamous cell carcinoma has been reported to arise from a branchial cleft cyst though every effort must be made to exclude a metastasis from a head and neck primary before arriving at such a diagnosis.[6]

CONCLUSION

Branchial arch anomalies, though rare, present diagnostic challenges in some cases and may hence be mismanaged, leading to considerable morbidity. They usually present in childhood and therefore pediatricians need to maintain a high index of suspicion and refer the patient appropriately.

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