CASE REPORT

Spontaneous closure of branchial sinus of the pyriform fossa

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SUMMARY

Management of third and fourth branchial cleft anomalies are similar. These anomalies should be suspected in a child with recurrent low-anterior neck abscess. Investigations in the form of cross-sectional studies and examination of the pharynx under anaesthesia will facilitate diagnosis and resolution of abscess. Spontaneous closure of the pyriform sinus can occur following conservative management with antibiotic treatment and abscess drainage. This emphasises the role of second-look prior to implementing endoscopic cauterisation or surgery.

BACKGROUND

Branchial cleft anomalies are an important differential diagnosis for paediatric recurrent anterior and lateral neck abscesses. They account for approximate 17% of all paediatric neck masses.¹ Third or fourth branchial cleft anomalies are a rare occurrence.^{2,3} They are associated with the classical finding of pyriform sinus fistula, which can be appreciated from cross-sectional studies or by direct visualisation of the opening at pyriform sinus during pharyngoscopy.^{4,5} Image modalities used to diagnose this condition include ultrasonography, CT with or without fistulography and MRI.^{2,6} Third and Fourth cleft anomalies can only be differentiated definitively from each other by their relationship with the Superior Laryngeal Nerve (SLN) during neck dissection.⁵ However, their management remains similar.²

Currently, there is limited evidence reporting spontaneous closure of these anomalies.^{7,8} The physiology and timeframe of closure are also not well understood. Here, we describe a third or fourth branchial cleft anomaly, which was diagnosed based on the CT (figure 1), pharyngoscopy (figure 2) and clinical findings which underwent spontaneous resolution from a 16-month-old girl who presented with low-anterior neck mass and restricted neck movement.

CASE PRESENTATION

A 16-month-old girl presented to the Accident and Emergency department when her parents noticed she has a restricted neck movement and reduced oral intake, following a 1 week history of upper respiratory tract infection. There was no associated swallowing difficulty, choking episodes or noisy breathing. The baby had no other significant past medical history and had achieved all her developmental milestones.

On examination, she had a firm indurated low-anterior neck mass left to the midline, in the

region of her thyroid gland with no signs of cellulitis or external fistula opening. She was admitted and treated with intravenous co-amoxiclav.

INVESTIGATIONS

Ultrasound of her neck showed a cystic collection anterior to her thyroid gland. Contrast-enhanced CT Neck showed a fluid collection with surrounding soft tissue enhancement anterior to the left thyroid gland lobe, consistent with abscess formation (figure 1). No other abnormal contrast enhancement of the soft tissue was noted at the level of the thoracic inlet. Thus further thoracic window assessment was not performed.

DIFFERENTIAL DIAGNOSIS

The main differential diagnoses for this patient include third or fourth branchial sinus or branchial cleft fistulae. It was unlikely for this case to be a branchial cleft fistula as a fistula tract was not visible on cross sectional imaging and there was an absence of an external fistula opening on examination. Only a branchial sinus opening was visualised on pharyngoscopy.

Both third and fourth branchial cleft anomalies can present as pyriform sinus fistulae, more commonly on the left side.^{2,5,8} While third branchial cleft fistula tends to originate from the upper lateral wall of the pyriform sinus, the fourth branchial cleft fistula tends to originate from the caudal end of the pyriform sinus.^{5,6} They can also be distinguished by the location of their fistula tract. The third branchial cleft fistula runs superficial to both superior and recurrent laryngeal nerves while the fourth branchial cleft fistula runs deep to the superior but superficial to the recurrent laryngeal nerve.^{6,9} Both can present with recurrent neck abscesses.⁸ While cysts from the third branchial cleft frequently arise posterior to the sternocleidomastoid muscle in the posterior triangle, the fourth branchial cleft anomaly tends to present with left-sided neck abscess and acute suppurative thyroiditis, as the track inclines to end at the upper part of the left thyroid.^{5,8} In this case, it was difficult to distinguish between third and fourth branchial sinus based on clinical findings. However, this does not have clinical significance, as the management of both third and fourth branchial anomalies is the same.

TREATMENT

Subsequently, the patient underwent incision and drainage of the neck abscess with

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Figure 1 CT Neck with contrast.

pharyngolaryngoscopy under general anaesthesia. During visualisation of the pharynx, an opening was noted at the left pyriform sinus ([figure 2](#)). A third or fourth branchial cleft anomaly was diagnosed based on the physical examination, pharyngolaryngoscopy and imaging characteristics.

OUTCOME AND FOLLOW-UP

The patient was discharged on a weeklong course of oral co-amoxiclav and was re-admitted for planned endoscopic cauterisation of pyriform sinus 4 weeks later. Before the surgery, the risks and benefits of endoscopic cauterisation and open-neck surgery including left hemithyroidectomy were discussed with

Figure 2 Initial pharyngolaryngoscopy view of the left pyriform sinus. *Red Circle highlighting the opening of branchial sinus.

Figure 3 Follow-up pharyngolaryngoscopy view of the spontaneously closed left pyriform sinus probed with a 5-Ch ureteric catheter.

the parents. On the day of the definitive procedure, the pyriform sinus was visualised again, but the opening at the pyriform sinus had scarred and closed over ([figure 3](#)). Thus no further interventions were required. The patient also had her 6 month follow-up. During this period, she did not have any readmissions, signs or symptoms that suggested the re-accumulation of her neck abscess. She was subsequently discharged from the specialty follow-up clinic.

DISCUSSION

Third and fourth branchial cleft anomalies occur following an incomplete involution of the branchial apparatus.¹ This can result in the formation of cyst, sinus or fistula.¹ Traditional treatments for congenital third or fourth branchial cleft anomalies are similar. These include treating the acute infective phase with

antibiotics and incision and drainage of the abscess, followed by open-neck surgery involving removal of the track or endoscopic cauterisation.² One systematic review on the management of third branchial cleft anomalies demonstrated that failure rate for endoscopic cauterization is comparable to traditional open-neck surgery (15% and 18% respectively; median age of diagnosis: 10-year-old).⁸ However, A case series of 20 patients with fourth branchial cleft anomalies (median age of diagnosis: 4-year-old) noted that there is a higher rate of failure of endoscopic treatment in neonatal presentations as these lesions are naturally secreting and re-opening of the tract can occur.² They recommended open-neck surgery for neonates with large cyst or early recurrence following incision of the cyst, while endoscopic approach was recommended for older children due to their lower relapse rate.² It is the author's opinion that endoscopic cauterisation should be first line in managing branchial cleft anomalies for older children if sinus were to persist during second-look as was our practice in this case study.

Spontaneous resolution of pyriform sinus fistula has previously been reported in children aged between 3 to 11.5 years old.⁷ A systematic review of third branchial cleft anomalies reported 10 cases of resolution following conservative treatment across all age groups with the majority of these cases belonging to those aged four and above.⁸ This case report provide additional evidence that spontaneous resolution of branchial cleft sinus occurs in an infant of 16 months' old.

It is possible that cases of spontaneous resolution might be higher than the number reported in the literature as branchial cleft anomalies could often be misdiagnosed in children presenting with a single or recurrent neck abscesses. Therefore, we advocate high vigilance for branchial cleft anomalies in

children presenting with recurrent low-neck abscesses, especially for those anterior to the thyroid gland or presenting with suppurative thyroiditis. Cross-sectional studies and examination under anaesthesia should be performed in this group of patients to clinch the diagnosis. Acute phase management will involve antibiotic treatment followed by incision and drainage of the abscess as incision and drainage as a single treatment modality alone is associated with high recurrence rate.⁸ We also propose that a planned second-look should be considered in all cases. Endoscopic cauterization should only be considered if sinus were to persist at that stage. Out-patient follow-up should be offered for monitoring if the definitive intervention is reserved during the second look, at least in 6 months' time.

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Learning points

- High index of suspicion for branchial cleft anomalies in children presenting with recurrent abscess of low anterior thyroid region.
- Diagnosis of branchial cleft anomalies should be made with cross-sectional imaging and pharyngoscopy under general anaesthesia.
- Antibiotics and abscess drainage should precede definitive management third/fourth branchial cleft anomalies.
- A planned second-look with and without endoscopic cauterization should be considered in older children prior to definitive management of branchial cleft anomalies.

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