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From Sinus to Fistula: The Hidden Cost of Repeated Incision and Drainage in Paediatric Branchial Cleft Anomalies, a Case Series

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Abstract: Branchial cleft anomalies commonly present as branchial cleft sinuses, which are epithelialized tracts that communicate with the skin or pharynx. While these anomalies are often asymptomatic, they can become complicated by infection, resulting in abscess formation. The standard management of an infected branchial cleft sinus involves prompt incision and drainage (I&D) to alleviate symptoms and control infection. However, repeated I&D procedures without early definitive surgical intervention to address the underlying anomaly may lead to significant preventable complications such as sinus-to-fistula transformation, chronic discharge, and substantial psychosocial morbidity, particularly in children. This case series describes two children with a history of recurrent lateral neck swellings that initially lacked any external openings. Both underwent multiple I&D procedures before presenting with fluid extrusion from newly formed cutaneous openings, which led to social distress, including peer ridicule. Definitive surgical excision was performed via an external approach, resulting in complete symptom resolution at the six-month follow-up. Early recognition, timely specialist referral and definitive management are key to minimising physical and psychosocial morbidity in branchial cleft fistulae.

Keywords: branchial sinuses, fistula, complications, needle aspiration, outcomes

Introduction

Branchial cleft anomalies (BCAs) result from incomplete obliteration of the branchial apparatus during embryonic development and may present as cysts, sinuses, or fistulas.[1] Second branchial cleft anomalies are most common, while third and fourth cleft anomalies are rare.[1–3] These BCAs are often asymptomatic and should be suspected in cases of recurrent lateral neck swellings.[1–4] If no external opening is present, the anomaly could be a cyst or internal sinus.[1,2] Typically located along the anterior sternocleidomastoid (SCM) border, second, third or fourth BCAs can be differentiated from other paediatric neck masses using ultrasound, with fistulograms or CT scans aiding surgical planning.[4–6]

Acute infections, particularly with upper respiratory tract infections (URTIs), require antibiotics and pain management.[3,4,7] Suppurative BCAs have traditionally been managed with incision and drainage (I&D), but needle aspiration is now preferred to preserve anatomy for definitive surgery.[5,8] Despite this, repeated I&D procedures remain common in resource-limited settings and may lead to chronic inflammation, fibrosis, and sinus-to-fistula transformation.[5] Fistulae pose additional surgical challenges due to scarring and require precise tract mapping before excision.[7,9]

Early definitive surgery prevents complications and recurrence.[10–13] Following infection control, preferably with needle aspiration and antibiotics,[5] surgical excision is recommended once inflammation subsides.[2,4,6] External approaches remain standard, with pharyngoscopy used to identify internal openings at sites such as the tonsillar fossa (for second BCA) or pyriform sinus (for third or fourth BCA).[4,6,7,9] Postoperative outcomes depend on infection severity and scarring. Delayed treatment increases risks of poor healing, recurrence, chronic skin irritation, and a small but notable risk of malignant transformation.[7,10,12] Importantly, the psychosocial impact of these complications, particularly

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visible neck discharge, stigma, and peer ridicule, remains under-recognised in the literature.[6,7,13,14] This case series was therefore undertaken to highlight the clinical and social consequences of repeated incision and drainage in paediatric BCAs, using consecutively collected cases. It also emphasizes the importance of early referral to Ear Nose and Throat specialists, and early definitive surgical management to prevent avoidable morbidity.

Case Report 1

A 5-year-old male presented with a 2-year history of fluid leakage from the left anterior neck whenever he drank liquids. Initially, at one year of age, he developed a painless swelling on the left side of the neck without an external opening. The swelling periodically enlarged during URTIs and later became painful, requiring multiple I&D procedures with antibiotic treatment.

Two years prior, an attempted surgical excision resulted in persistent leakage of clear fluids from the neck whenever he drank, though semi-solid and solid foods were unaffected. He was subsequently referred to our paediatric ENT clinic for further evaluation.

On examination, the child was in good health with normal growth parameters and no breathing difficulties or chronic illnesses. A fistula was noted at the lower third of the anterior border of the left sternocleidomastoid muscle, discharging clear liquid feeds (Figure 1). No tenderness, swelling, or additional neck abnormalities were observed.

Preoperative full blood count was normal. Preoperative imaging was not pursued due to the high financial cost to the patient. Endoscopic evaluation under general anaesthesia confirmed a connection between the sinus tract and the left pyriform sinus, diagnosing a third branchial cleft fistula (Video S1). Excision was performed via a single transverse neck incision at the mid-jugular level. The fistula tract was followed medial to sternocleidomastoid, lateral to common carotid artery with challenging dissection due to fibrosis. The immediate postoperative period was uneventful. A drain was left in situ and removed after 24 hours. The patient was discharged on postoperative day two, reviewed at one month, and demonstrated complete symptom resolution at six months.

Case Report 2

An 8-year-old male presented with a four-year history of extrusion of porridge and fluid from the left anterior neck. His mother first noticed a small, painless swelling when he was 3-years old, which had no external opening. Over time, the swelling periodically enlarged during infections and resolved with over-the-counter antibiotics. Eventually, it became suppurative, requiring repeated I&D procedures at a local health facility. Four years prior, the wound from an I&D failed to heal, and the child began experiencing semisolid fluid leakage. He expressed distress over being ridiculed by peers

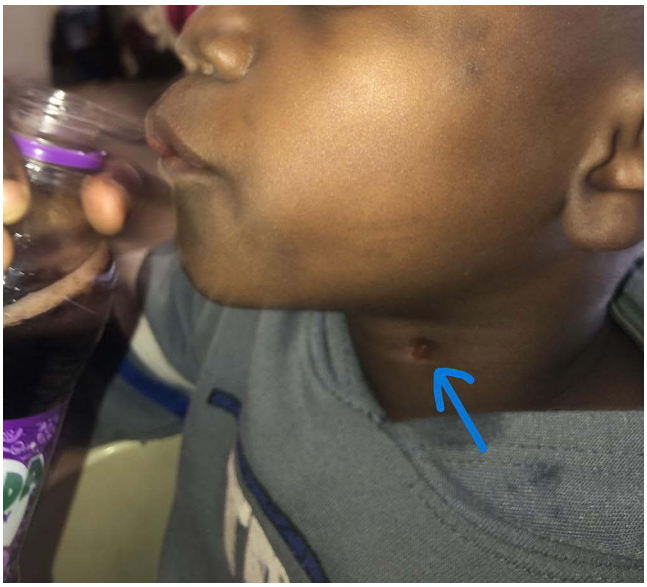

Figure 1 Extravasation of colored fluid being drunk by patient from the external opening (*arrow*) at the anterior border of the sternocleidomastoid muscle.

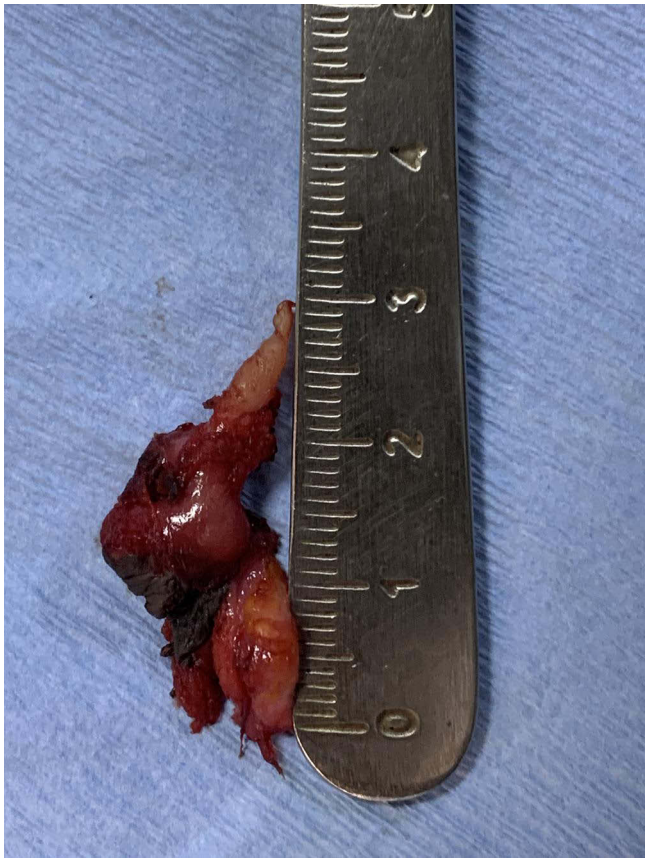

Figure 2 The 3cm-long fistula tract after excision.

during school mealtimes. Although referred to a hospital with ENT expertise, financial constraints delayed intervention. Occasionally, the wound ceased leaking, but recurrent URTIs triggered new episodes. He had no known chronic illnesses and was ultimately referred to our unit via a community outreach program.

On assessment, the child was well-nourished with an ongoing URTI but no respiratory distress. Vital signs, ear, nose and throat examinations were normal. Neck examination revealed a scar at the lower third of the left anterior sternocleidomastoid border with a punctum that oozed thick, odourless pus upon pressure. No additional neck abnormalities were noted.

After treating the URTI, definitive surgery was scheduled. A prior fistulogram provided limited diagnostic information and was therefore not repeated. Further imaging was not pursued due to the high financial cost to the patient. Under general anaesthesia, endoscopic examination identified a punctum in the pharynx, 1cm below the left tonsil. The 3cm long tract (Figure 2) was excised via an external approach, via a transverse mid-jugular incision. The tract coursed medial to the sternocleidomastoid and lateral to the common carotid, with fibrosis complicating dissection. Recovery was uneventful; the drain was removed at 24 hours. The patient was discharged on day two. Postoperatively, minor wound dehiscence occurred but resolved with dressing. At six weeks, the child was asymptomatic and remained symptom-free at the 6- months follow-up visit.

The children's parents provided informed consent for the case details and any accompanying images to be published, available upon request.

Discussion

These two cases highlight the diagnostic and management challenges associated with third and fourth branchial cleft anomalies, which, unlike the more common second cleft anomalies, are rare and often complicated by recurrent infections.[1–4]

In our series, cases presented before the age of 15 years with symptoms of left lateral neck masses first noticed in early childhood, but diagnosis was delayed by over two years. Several case series similarly report the onset of BCA symptoms between infancy and adolescence,[2,4,7,11] although missed diagnoses have also been described in middle-aged

patients.[3] A left-sided predominance of neck presentations, as observed in our series, is frequently noted in the literature. For the non- specialist clinician, a high index of suspicion should be maintained for any chronic or recurrent lesions in the infra-auricular or upper neck (particularly along the anterior border of the sternocleidomastoid muscle) region, mostly in children and young adults.[13]

Beyond neck masses and their suppuration, additional clinical manifestations of third and fourth BCAs reported in the literature include thyroiditis and stridor with obstructive symptoms.[2–4,7] In our cases, the predominant feature was recurrent lateral neck swellings, often managed with I&D for infection control. However, this approach eventually led to the development of persistent fistulous tracts with external openings, resulting in chronic discharge of fluids, including food particles. These complications underscore the risks of repeated I&D, which does not address the underlying congenital anomaly and instead promotes fibrosis, scarring, and epithelialization of new tracts, ultimately predisposing to fistula formation. Similarly, Thomas et al in a review of 20 cases of third and fourth BCAs, found that none represented true congenital branchial cleft fistulas; rather, they appeared to be acquired, secondary to recurrent infections or multiple I&D procedures.[4] A high index of suspicion is therefore essential when evaluating recurrent lateral neck swellings in children. In the absence of an external opening, the lesion may represent a cyst or an internal sinus.[1,2,4] Nevertheless, diagnostic delays are common with Sheng et al reporting a mean delay of approximately two years from symptom onset to diagnosis,[7] a finding that parallels our experience.

Imaging with ultrasound is a useful first-line modality particularly when thyroiditis is suspected, while contrast-enhanced CT scans remain the modality of choice for surgical planning[4,7.] However, several retrospective series have questioned their utility in demonstrating internal openings[3,7,11] Barium studies and fistulograms also have variable sensitivities.[3,7] Endoscopic evaluation, assisted by dye or methylene blue, has therefore been emphasized as a reliable method for identifying pyriform sinus or tonsillar fossa openings.[2,4,7,11] In our cases, endoscopic visualization with dye was sufficient to confirm third branchial cleft fistulas, obviating the need for additional out-of-pocket radiological investigations and reducing the financial burden on patients. In resource-constrained settings like ours, access to advanced imaging is often limited, and their cost is borne directly by patients and their families. Therefore, relying on careful clinical assessment, basic imaging where available, and intraoperative endoscopic confirmation can enable timely definitive treatment while minimizing delays, unnecessary investigations, and financial hardship. This clinically guided approach significantly influences management decisions for pragmatic reasons, thereby improving access to care and overall outcomes in low-resource environments.

Management of infected BCAs requires balancing acute symptom control with prevention of long-term complications.[6,11] Although antibiotics are essential, traditional incision and drainage should be avoided, as repeated procedures promote fibrosis and recurrence.[2,4,7] Needle aspiration is increasingly recommended to preserve anatomy and facilitate definitive surgery.[5,8] Once infection resolves, early surgical excision is advised to prevent sinus-to-fistula progression. External approaches remain the standard, supported by endoscopic identification of internal openings, while endoscopic electrocautery and chemocautery have shown promise in selected series[9–11,] though these techniques are not yet widely available in our setting. A tailored algorithm has been proposed that considers lesion type, infection history, thyroid involvement, and recurrence.[6]

Postoperative outcomes vary with the extent of infection and scarring.[10,12] In one series, 5 of 14 patients who underwent excision of third or fourth BCAs experienced recurrence.[10] Another study reported an overall good outcome in 94% of 48 patients, with the two recurrences occurring in cases without endoscopic assistance during open surgery.[7] Following endoscopic electrocautery, 2/7 patients had recurrence within six months,[11] while a larger series reported a 3.6% recurrence rate in 421 patients at four years postoperatively.[12] Prior I&D was associated with a 3.4-fold increased risk of recurrence and a 2% complication rate.[12] Rare malignant transformation has been documented in older patients (>45 years).[12] In our setting, one case developed wound dehiscence but achieved resolution within six months, and both cases will be followed annually for at least four years.

Delayed intervention increases the risk of persistent discharge, poor healing, and recurrence, while also imposing psychosocial burdens such as skin excoriation and social stigma.[6,7,13] Psychosocial consequences like peer ridicule and social distress are often underemphasised to clinical outcomes of branchial cleft fistulae. No known studies quantify psychosocial outcomes specifically for branchial cleft fistulae, although several features of this condition suggest

a substantial burden.[6,7] The conspicuous location of fistulous openings on the neck with a chronic visible discharge, occasional malodour and recurrent soiling of clothing may expose affected children to embarrassment, stigma and social exclusion especially within school environments, like what was shared by our cases. It may be extrapolated from broader surgical literature, that complications and recovery are strongly associated with adverse psychosocial outcomes, including anxiety, reduced self-esteem, social withdrawal, and diminished quality of life, even when complications are not life-threatening, for at least 12 months after surgery.[14] It is probable that preventable complications such as sinus-to-fistula transformation from repeated I&D may carry psychosocial consequences that are inconsistent to their clinical severity. Sinus-to-fistula transformations may amplify psychosocial distress and have lasting impressions, particularly in children, where peer interaction and body image are still developing. This case series therefore draws attention to the psychosocial domain and implores clinicians to consider its morbidity alongside physical complications when managing branchial cleft anomalies, particularly in settings where delayed definitive care is common.

This case series involves only two patients and is innately limited in scope to be generalised to all children with BCAs. Meaningful comparisons between different management strategies or outcomes such as recurrence rates and long-term complications cannot be constructed on this small sample size. Currently, no standard diagnostic or management protocol is available in our setting, and resource and financial constraints influenced the management pathways for our cases. These practices may differ from those in higher-resource settings. Despite these limitations, the cases provide valuable clinical insights into preventable complications of repeated I&D and highlight an often-overlooked psychosocial burden, underscoring the relevance of early recognition and definitive management in similar contexts.

Conclusion

The management of recurrent infections in presumed branchial cleft anomalies may benefit from needle aspiration instead of repeated I&D to alleviate acute symptoms and prevent the progression from a sinus to a fistula. Based on the observation from our limited case series, early recognition in primary care, avoidance of repeated I&D, and timely referral for definitive excision to specialists are the main strategies to prevent complications and their psychosocial consequences.

Ethics Approval

The children's parents provided written informed consent for the case details and any accompanying images to be published, available upon request. Institutional approval was not required to publish the case details.

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Author Contributions

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