

Case Report

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Endoscopic intervention in second branchial sinus in a 5-month old infant

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ABSTRACT

Second branchial sinus anomalies are rare congenital paediatric maladies that arise during intrauterine embryogenesis. A unique case of second branchial arch sinus in a 5-month-old infant, where a minimally invasive transcervical endoscopic intervention was undertaken, is being reported. The presentation was with recurrent discharge from a punctum at the lower lateral aspect of the neck. Global literature on second branchial arch anomalies is reviewed with clinical presentation, imaging, diagnosis, and therapeutic modalities of this rare entity.

Keywords: Congenital branchial cleft anomalies, Second branchial arch sinus, Endoscopic intervention

INTRODUCTION

Second branchial sinus anomalies are congenital conditions with a likely onset at about the 5th week of intrauterine development of the human embryo.¹ Infact incomplete fusion of the branchial clefts, is consequent to formation of a tract of sinus, localised cyst or a fistula. 95% of all branchial arch anomalies arise in the second branchial arch.² The third arch defects are less frequent compared to the first and second branchial clefts.³ Agaton-Bonilla, Gay-Escoda and Davenport, documented these anomalies to abnormal development of the branchial apparatus.^{3,4} Usual presentation is a cystic lesion instead of a sinus or a fistula. The latter infact present later, i.e. in the second decade of life. Cysts are palpable non-tender indurations deep to the sternocleidomastoid muscle. Often swelling, with

redness, warm periphery, pain, odynophagia, recurrent infections or cervical abscesses may be seen.⁵ Branchial cleft cyst infection with an increase in the size of the swelling and pain was documented by Leung.⁵

CASE REPORT

A 5-month-old baby presented with a small opening on the left side of the neck, at the anterior border of the junction of lower one third and upper two third of the sternocleidomastoid (Figure 1). There was off and on mucus discharge from the opening. Ultrasound and MRI scans revealed a sinus tract extending from the skin to the superior constrictor muscle overlying the tonsillar fossa (Figure 2 A-C). The tract was cannulated and methylene blue injected to delineate its upward and inward course. (Figure 3). With a limited skin ellipse at its proximal

origin, a circumferential iris scissors dissection was undertaken (Figure 4).



Figure 1: Punctum on left side of the neck, at the anterior border of the junction of lower one third and upper two third of the sternocleidomastoid.

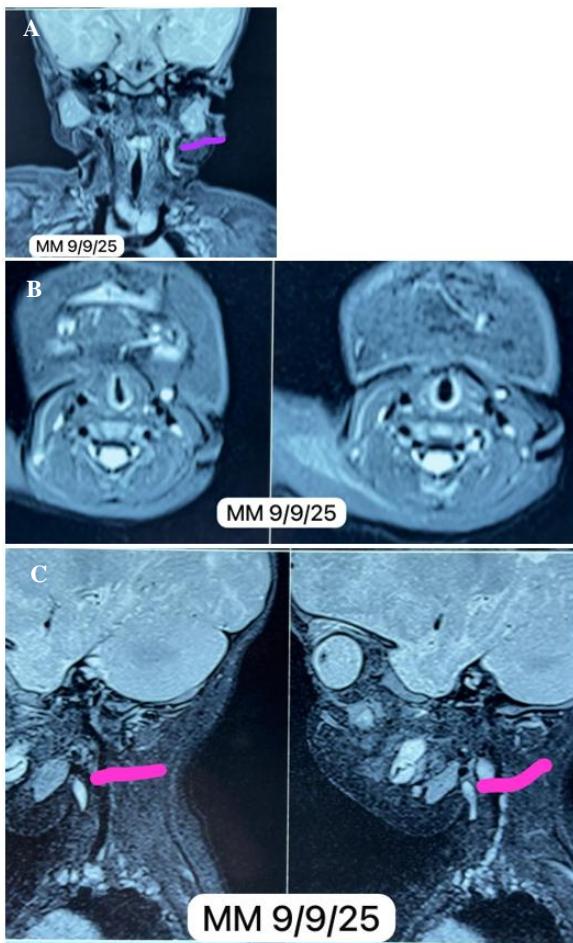


Figure 2 (A-C): MRI with contrast in coronal, axial and lateral views, delineating extend of tract from anterior border of sternomastoid to the tonsillar fossa.

0-degree endoscopic visualization delineated a 4 x 16 cm cylindrical tract, which was traced between the fork of the carotids (Figure 5A and B). It was transfixed at the level of the superior constrictor muscle (Figure 6). Haemostasis was achieved and the incision was sutured over an underlying drain that was removed on the second day. Post op period was uneventful and sutures were removed on the seventh day. Histopathology confirmed the diagnosis of a branchial sinus.



Figure 3: Sinus cannulated and methylene blue injected.

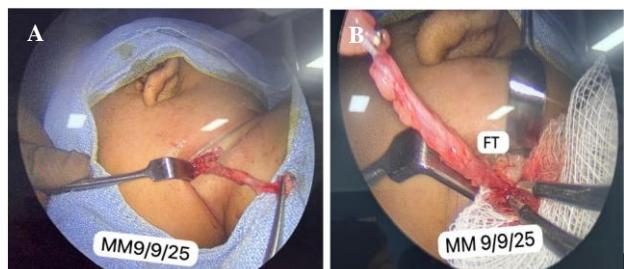


Figure 4 (A and B): Creating a proximal skin ellipse, a circumferential iris scissors dissection of the tract.

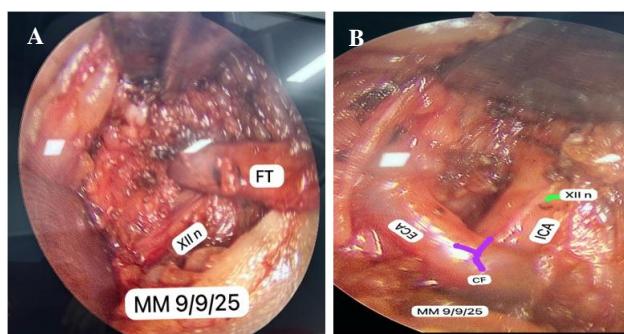


Figure 5 (A and B): A cylindrical tract ft, which was traced between the fork of the carotids. Carotid fork (bifurcation / bulb).

ICA - internal carotid artery; ECA - external carotid artery; XII n - hypoglossal nerve.

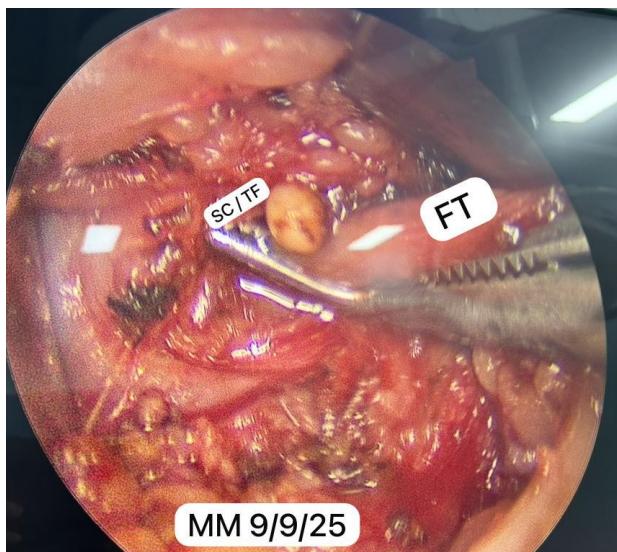


Figure 6: FT - fibrous tract being transfixed just above the SC /TF - superior constrictor / tonsillar fossa.



Figure 7: 5 cm long Branchial sinus dissected out 8.



Figure 8: Post operative period was uneventful.

DISCUSSION

Anomalies of the second branchial cleft have been categorised utilizing the conventional 1929 Bailey system. Our lesion fulfilled the type 3 criteria; that is the least common, where the tract ascends from the anterior neck, traversing between the internal and the external carotids, ultimately terminating into the lateral pharyngeal wall. It was superficial to the IX and the XII lower cranial nerves prior to termination in the superior constrictor. In this tract sinuses or cysts of the second branchial cleft are likely.^{6,7}

The tract descended from the palatine tonsil down to the anterior neck, passing between the internal and external carotid arteries and superficial to cranial nerves IX and XII. Second branchial cleft anomaly has a typical presentation with a skin pit or punctum juxtaposed to the anterior border of the sternocleidomastoid, in its lower third. Distally it connects to the tonsillar fossa of the pharynx. Secondary infection manifests with localised redness, maceration, induration and tenderness. Airway compromise is likely in advanced cases. Mucoid or purulent discharge on the skin or in the pharynx is suggestive of a second branchial cleft anomaly associated with a sinus tract.^{8,9}

Diagnosis is typically made through a combination of clinical examination, imaging studies, and histopathology. Pereira et al emphasised on, a complete excision of the sinus tract being crucial to avoid a recurrence.¹⁰ Surgical excision is the treatment of choice, and endoscopic or open approaches can be utilized depending on the extent of the anomaly.

Non-invasive and invasive modalities can diagnose a branchial sinus. Radiological features are a cystic lesion or sinus tract on ultrasonography, computed tomography and magnetic resonance imaging. These non-invasive procedures with precision demarcate the location and extent of the sinus and its ramifications towards the viscera and major neuro-vascular structures of the neck.¹¹ Contrast sinogram clinches the diagnosis. Biopsy of the excised specimen is undertaken to exclude neoplasia.

Therapeutic modalities are surgical excision, to remove the entire sinus tract and prevent recurrence.¹⁰ Conventional naked eye or microscope assisted, "step ladder" open surgical or the recent minimally invasive endoscopic approach can be employed to demarcate and dissect the entire tract till its distal origin from the constrictor muscles. The innovative endoscopic retro auricular, access has too been adopted, thereby hiding the scar: but with an increase in the operative time.^{12,13} Surgical intervention can be deferred to 3 to 6 months of age when there is no compression of the airway nor any cervical suppuration. Moreover where one can wait for resolution of acute infections.⁵ On the contrary surgery is necessitated in sudden airway compromise consequent to uncontrolled cervical suppuration.^{14,15} Some prefer

systemic third to fourth generation antibiotics and repeated aspirations in localised collections in lieu of incision and drainage. In these patients there is distortion and scarring of anatomical planes of the neck, that effects a later excision.¹⁶ Histologically branchial cleft cysts have a lining of stratified squamous epithelium with keratinous debris inside. Often the cyst wall is lined by ciliated columnar epithelium, which leads to more mucoid collection. Lymphoid tissue exists all around the epithelial lining. In the event of cyst infection or rupture, inflammatory cells are visualised in the cavity or stroma.¹⁷

The recurrence rate is hardly 3% post excision and 20% in revision surgery or where there were recurrent skin infections due to subcutaneous plane fibrosis and distortion; and thereby difficulty in complete surgical resection.¹⁷

Sclerotherapy is an alternative second line non-surgical treatment.¹⁸ Ethanol, Picibanil, doxycycline, tetracycline and bleomycin are the agents that can be utilized.¹⁹⁻²¹ In the event that a patient cannot undergo surgery or refuses a neck incision, ethanol ablation has been used as an alternative. However, this approach is not usually recommended as a primary treatment.¹⁸ Other agents that used, with or without ultrasound guidance, include OK-432 (Picibanil), doxycycline, tetracycline, and bleomycin.¹⁹⁻²¹

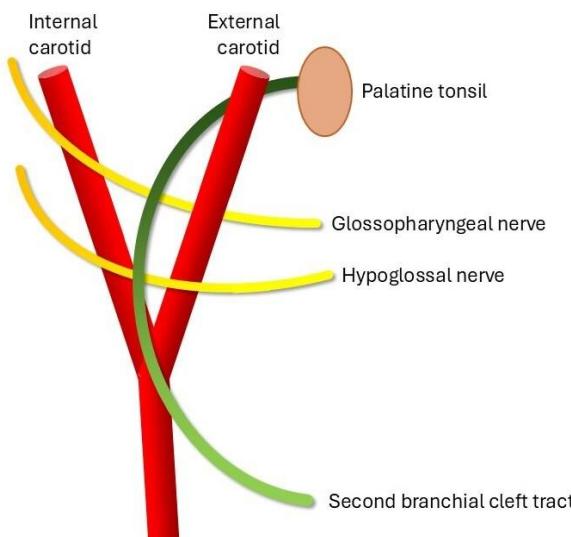


Figure 9: Schematic diagram showing the tract of the second branchial cleft.²²

CONCLUSION

Second branchial arch sinuses are rare congenital anomalies that require prompt diagnosis and surgical management. Imaging studies are useful in evaluating the extent of the anomaly, and surgical excision is the definitive treatment. We report a rare case of a second branchial arch sinus in a 5-month-old infant, highlighting

the importance of early diagnosis and treatment complete excision to prevent recurrence.

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