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Case Report

Bilateral branchial arch anomaly: a rare case report

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ABSTRACT

Anomalies of branchial arches are uncommon anomalies of embryonic development and may present as cysts, sinus tracts, fistulae or cartilaginous remnants. They represent the embryological precursors of face, neck and pharynx and are the second most common congenital lesions of head and neck in children. Although pharyngeal apparatus (branchial) anomalies are frequently seen, bilateral cases are rare (only 2% to 3%). Our patient was a 3 months old child with a swelling on right side of neck and discharging sinus from left side since birth. Surgical excision of the right branchial cyst with its complete tract was done along with excision of the left branchial fistulous tract upto the tonsillar fossa. Post-operative course was uneventful and patient was discharged the next day.

Keywords: Branchial apparatus, Cyst, Fistula, Sinus

INTRODUCTION

The branchial arches develop in the fourth week of gestation and are the embryological precursors of ear and muscles, blood vessels, bones, cartilage and mucosal lining of the face, neck and pharynx. Depending on the anatomic location, branchial anomalies have been classified into first, second, third and fourth arch anomalies.[2] Second branchial arch anomalies are the most common accounting for approximately 95% of cases. Our case had bilateral branchial anomaly.

CASE REPORT

A three months old male child brought by mother with complains of swelling over anterior right side of neck and discharge from an opening on the left side, since birth.

There was no history of fever, respiratory distress, stridor, birth trauma or weight loss. He had been delivered full term, vaginally. General and systemic examinations were normal.

Local examination revealed a cystic swelling on the right side in lower one-third of anterior triangle of neck, non-tender, measuring 2.5x2 cm (Figure 1).

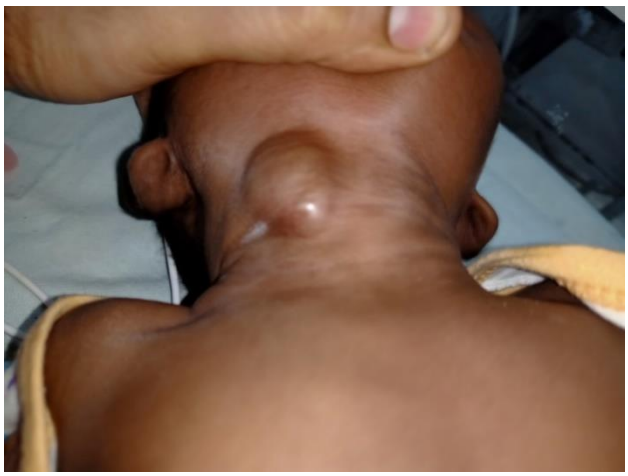

Figure 1: Cystic swelling at lower aspect of right-side neck.

Discharging sinus was present on left side of upper third of anterior triangle of neck. Under general anaesthesia

with neck extended the fistula on left side was cannulated with 3-0 proline suture for delineation. A transverse elliptical incision was made around the external opening. With needle tip cautery the tract was dissected from the surrounding tissues, traced superiorly upto the tonsillar fossa, excised completely from the base, and transfixed (Figure 2). On the right side the cyst (Figure 3) was completely excised and its stalk was transfixed. Post-operative course was uneventful and patient was discharged the next day.

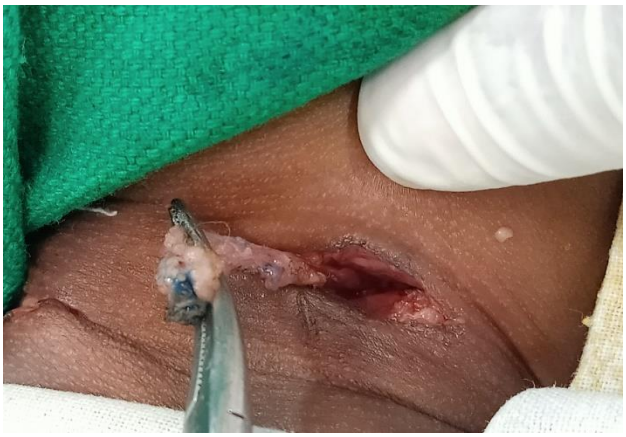

Figure 2: Excision of tract.

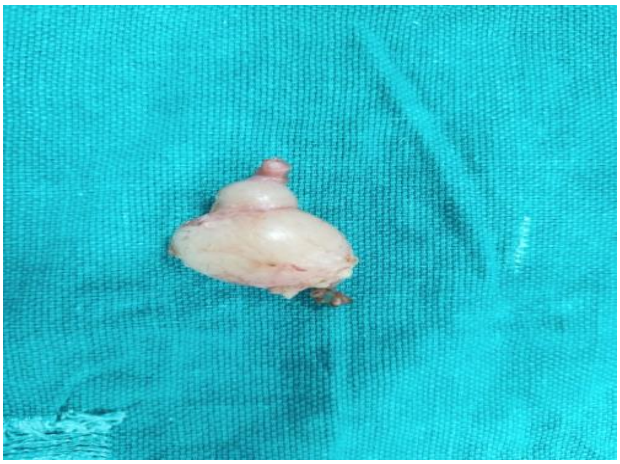

Figure 3: Intact excised cyst with its tract.

Histopathology showed presence of squamous epithelium consistent with branchial cyst on right side and fistula on left side.

DISCUSSION

Several theories have been proposed for the development of branchial anomalies. These include branchial apparatus theory, cervical sinus theory, thyropharyngeal theory and inclusion theory. Amongst these, the widely accepted theory is that branchial anomalies develop as a result of incomplete involution of the branchial apparatus.[3] Pharyngeal apparatus development begins at about the second week of gestation and is completed by the sixth to seventh week. It is made up of pouches (endoderm), arches (mesoderm) and grooves (ectoderm). Most branchial arch anomalies arise from the second branchial cleft 92.45%.[4] They can present as cysts, fistulas, sinuses or cartilaginous remnants. Branchial cysts usually present in older children/young adults, whereas fistulas typically present in infants/young children. A sinus is a blind ending tract which may connect with either the skin (branchial cleft sinus) or with the pharynx (branchial pouch sinus). A fistula is a communication between two epithelialized surfaces, i.e., between a persistent pouch and cleft. If there is no communication with the inner mucosa or outer skin, then the trapped branchial arch remnant will form a cyst. 2nd cleft anomalies can occur anywhere from skin to the supraclavicular fossa, between the internal and external carotid arteries, to pharynx at the level of the tonsillar fossa. Few cases of bilateral pharyngeal (branchial) cleft anomalies have been reported in literature.[5] Huangetal reported 3 bilateral cases in their series of 37 patients.[6] However, only 6 cases of bilateral branchial cleft fistulas have been reported in the English language literature.[7] Bilaterality has a familial predisposition. Gatti and Zimm reported 2 cases.[8] Our institute has previously reported bilateral branchial sinuses in identical twins, a first of its kind.[9] Recurrence rates are 14% and 22% with previous infection and surgery respectively, whereas recurrence rate for primary lesion is 3%. No radiological investigations are usually required. There is lack of spontaneous regression and a high rate of recurrent infection, hence complete surgical excision is necessary.

CONCLUSION

Bilateral anomalies arising from second pharyngeal arch are rare. Diagnosis is mostly clinical and complete excision provides definitive cure. This report describes rare bilateral presentation managed surgically in same setting.

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