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Case Report

A rare case report: bilateral cervical chondrocutaneous branchial remnants

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ABSTRACT

Cervical chondrocutaneous branchial remnants (CCBR) are rare congenital benign neck masses. They are usually present in males and can be both unilateral and bilateral. In this article, we present a case of bilateral CCBR in a child which is one of the very few bilateral cases that have been reported till date in the literature. The masses were excised completely and diagnosis was confirmed on histopathology. It is important to be aware of this disease entity as it is associated with a number of congenital anomalies and syndromes.

Keywords: Cervical chondrocutaneous branchial remnant, Choristoma, Bilateral congenital neck mass, Cervical tags

INTRODUCTION

Cervical chondrocutaneous branchial remnants (CCBR) are rare congenital benign neck masses.[1] They are usually present in males and can be both unilateral or bilateral. CCBR are usually associated with other congenital anomalies of the cardiovascular system or genitourinary system.[2,3] They have been previously known by different names like cervical tags, skin appendages, hamartomata, vestiges, fibromata, accessory auricles or tragai and wattles.[4-6] The name 'cervical chondrocutaneous branchial remnants' was suggested by Atlan et al.[2]

CASE REPORT

A 3-year-old boy presented to the outpatient department with bilateral painless neck swellings since birth. The swellings were gradually increasing in size with age. On examination, (Figure 1) two smooth skin covered pedunculated swellings were noticed in the lateral aspect of upper neck measuring 1.5×0.5 cm bilaterally. On palpation both swellings were firm with cartilaginous feel to them and were non tender. There was no significant history of a similar swelling in any other family member. Ultrasonography was done which demonstrated ill marginated hypoechoic lesions measuring 8×3 mm and 7.6×2 mm in the skin and upper subcutaneous plane of upper lateral neck on left and right sides respectively (Figure 2). There was no evidence of extension into the myofascial planes with no obvious calcification or significant vascularity within. Systemic examination was normal with no other abnormality detected. Provisional diagnosis of branchial remnants was made and surgical excision was planned.

The child was operated and both the masses were excised completely with and without the overlying skin on the right and the left sides respectively under general anesthesia. The swellings were made up of cartilage without any deeper extensions into the muscular plane (Figure 3). The excised tissue was sent for histopathological examination which confirmed the

contents to be elastic cartilage remnants bilaterally (Figure 4).

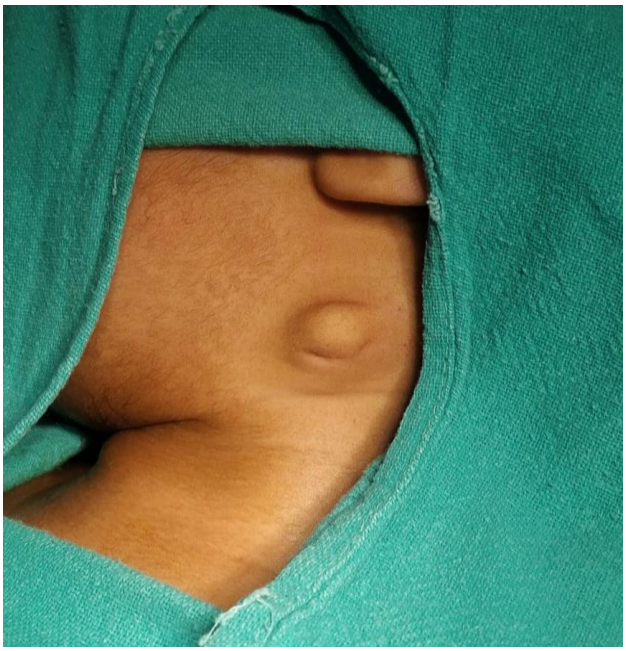

Figure 1: Left sided swelling.

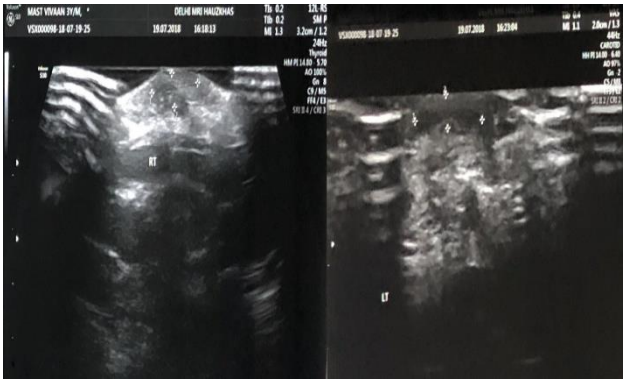

Figure 2: Ultrasonography images which demonstrated ill marginated hypoechoic lesions measuring 8×3 mm and 7.6×2 mm in the skin and upper subcutaneous plane on left and right sides respectively.

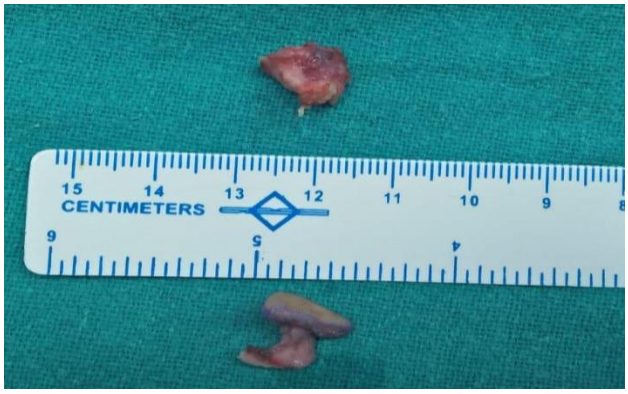

Figure 3: Right (above) and left (below) sided swellings surgical excision specimens

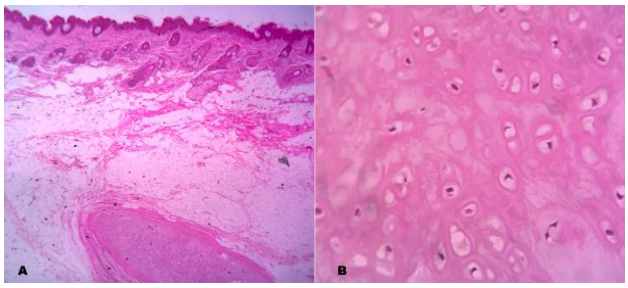

Figure 4: (A) Scanner view of skin tissue showing normal epidermis and dermis with adnexal structures; the subcutaneous fat showing a well-defined island of elastic cartilage [H & E, 40X], (B) the high power view of elastic cartilage [H & E, 400X].

DISCUSSION

CCBR are uncommon congenital lesions of the lateral neck. The most common lesion presenting as congenital cystic lesion in the neck originates from second branchial arch anomaly.[7] Clinically, CCBR is a painless, non-discharging and non-erythematous mass. It has a male predominance and is usually found in the middle and lower third of the neck, however in present case it was seen in upper neck.[8] Although diagnosis of CCBR is usually clinical, detailed history and systemic examination is essential because it is associated with various congenital anomalies and syndromes as given in Table 1.

Table 1: Congenital anomalies and syndromes associated with CCBR.

Category	Anomaly	Reference
Auditory system	Serous otitis media	2
	Sensorineural deafness	2
	Microtia	3, 9
	EAC stenosis	2, 3
Cardiovascular system	Atrial septal defect	2, 10
	Ventricular septal defect	2, 10
	Tricuspid regurgitation	10
	Patent ductus arteriosus	2
	Mitral regurgitation	10

Continued.

Category	Anomaly	Reference
Gastrointestinal system	Umblical hernia	2
	Inguinal hernia	2
Genitourinary system	Hydronephrosis	2
	Hydrocele	2
	Hypospadias	2
	Cryptorchidism	2
Head and neck	Preauricualar sinus	2, 11
	Branchial-oto-renal syndrome	11
	Tongue tie	2
	Retroauricular dermoid cyst	2
	Cleft palate	2, 12
	Celft lip	2, 12
	Treacher collins syndrome	13
Respiratory system	Pulmonary atelectasis	2
	Tracheomalacia	2
Visual system	Strabismus	2
	Lacrimal duct stenosis	2

The origin of CCBR is controversial and not clear even today. The most accepted theory is incomplete obliteration of branchial apparatus during embryogenesis. However, its origin from epithelial cell rest and auricular tissue have also been reported.[2] Histopathological examination is diagnostic for CCBR is which reveals elastic or hyaline cartilage. The presence of elastic cartilage suggests an auricular origin while hyaline cartilage indicates cervical origin.[14,15] Complete surgical excision is the only permanent cure of CCBR.

CONCLUSION

CCBR is a rare anomaly in medical literature. Our case is one of the very few bilateral cases which have been reported till date. It requires a thorough history and systemic examination as it is associated with a number of congenital anomalies and syndromes.

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