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Original article

Cervical Chondrocutaneous Branchial Remnant: A Case Report highlights the Essential Role of Histopathology in Diagnosing Congenital Neck Lesions

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Cervical Chondrocutaneous Branchial Remnant (CCBR), Congenital Neck Lesion, Histopathology, Hyaline Cartilage.

ABSTRACT

Cervical Chondrocutaneous branchial remnant (CCBR) is an extremely rare, benign, congenital choristoma of the neck. Fewer than 120 cases have been reported to date, mostly as isolated case reports. Because of its clinical similarity to other congenital neck anomalies, such as branchial cleft cysts and thyroglossal duct cysts, establishing an accurate clinical diagnosis remains challenging for surgeons and pathologists. Differentiating CCBR is clinically significant because it influences surgical management and prognosis. We present the case of a five-year-old Libyan girl with a painless, unilateral congenital lesion on the left anterolateral neck, overlying the sternocleidomastoid muscle. The patient's medical history was notable for a resolved atrial septal defect (ASD) and bilateral hearing loss, which may suggest systemic associations warranting further evaluation. Following complete surgical excision, histopathological evaluation confirmed the diagnosis of CCBR. Histopathological evaluation is essential to confirm CCBR and differentiate it from other congenital cervical masses. Given the potential clinical implications, correlating these findings with systemic anomalies is vital for comprehensive patient management.

Introduction

CCBR are rare choristomas of the head and neck [1], with fewer than 150 cases reported in the literature to date [2]. It was first described in 1958 by Birkett et al [3]. Previously known as accessory tragus, wattles, or periauricular appendages [4], these represent an uncommon subset of congenital cervical anomalies characterized by the presence of ectopic cartilage [5]. These lesions typically present as painless [6], pedunculated or sessile nodules located predominantly in the lower third of the sternocleidomastoid muscle [6]. May present as unilateral or bilateral lesions [4].

From an embryological perspective, the pathogenesis of CCBR is not fully understood [7], and is widely regarded as a remnant of the second branchial arch [7], resulting from a failure of the involution of the cervical sinus [7]. Despite their benign nature, their clinical appearance often overlaps with other congenital neck pathologies [8], ranging from dermoid cysts to more complex lymphatic malformations, such as cystic hygromas [8]. CCBR has been associated with other congenital anomalies in approximately 76 % of cases [1]. Karyotyping and ultrasonography are recommended for children with CCBR [1].

CCBR can be difficult to differentiate through clinical and radiological examinations alone [8], and they may occur in association with other congenital auditory, respiratory, or cardiovascular anomalies [4]. A definitive diagnosis is essential [9]. Histopathological evaluation following surgical excision remains critical for confirming the diagnosis and excluding other differential diagnoses [3].

Case Presentation

A five-year-old Libyan girl presented to the outpatient clinic of the Burn and Plastic Surgery Center in Tripoli with a painless congenital lesion on the left anterolateral aspect of the neck (Figure 1). The patient's medical history was significant for bilateral hearing loss; however, there was no family history of congenital anomalies. Notably, she had been diagnosed with an atrial septal defect (ASD) at the age of two, which reached complete spontaneous resolution, as confirmed by follow-up echocardiography. At the time of presentation, no cardiovascular complications were observed.

Physical examination revealed a firm, painless, pedunculated mass covered with unremarkable skin. The lesion was localized to the lower third of the left sternocleidomastoid muscle. There were no clinical signs of inflammation, active discharge, or evidence of associated sinus tracts. Ultrasonography examination revealed a well-defined, hypoechoic soft-tissue mass in the neck, without extension or connection to deeper structures. To confirm the diagnosis and for aesthetic reasons, surgical excision was performed under general anesthesia. The postoperative period was uneventful, with no complications. Follow-up showed good recovery.

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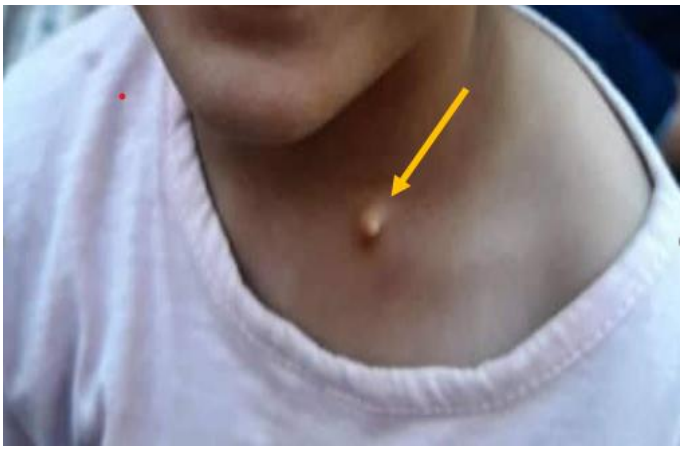

Figure 1 Clinical image showing a well-defined, fleshy nodule covered with intact skin (yellow arrow), located at the lower third of the left sternocleidomastoid muscle, measuring 1.5 × 1 cm.

Histopathological examination grossly revealed that the excised specimen consisted of a skin-covered, pedunculated mass of firm consistency, measuring approximately 1.5 × 1 cm. The specimens were fixed in 10% neutral buffered formalin, dehydrated, and embedded in paraffin blocks. Serial sections were cut at a thickness of 4 µm and stained with Hematoxylin and Eosin (H&E) for light microscopic evaluation. Histological analysis displayed a central core of mature hyaline cartilage surrounded by fibro-adipose tissue and dense connective tissue stroma (Figure 2 A and B). The overlying skin demonstrated a normal epidermis with associated dermal adnexal structures, including hair follicles and sebaceous glands. No evidence of epithelial-lined cystic spaces, lymphoid follicles, or malignancy was found. These findings are pathognomonic of a cervical Chondrocutaneous branchial remnant.

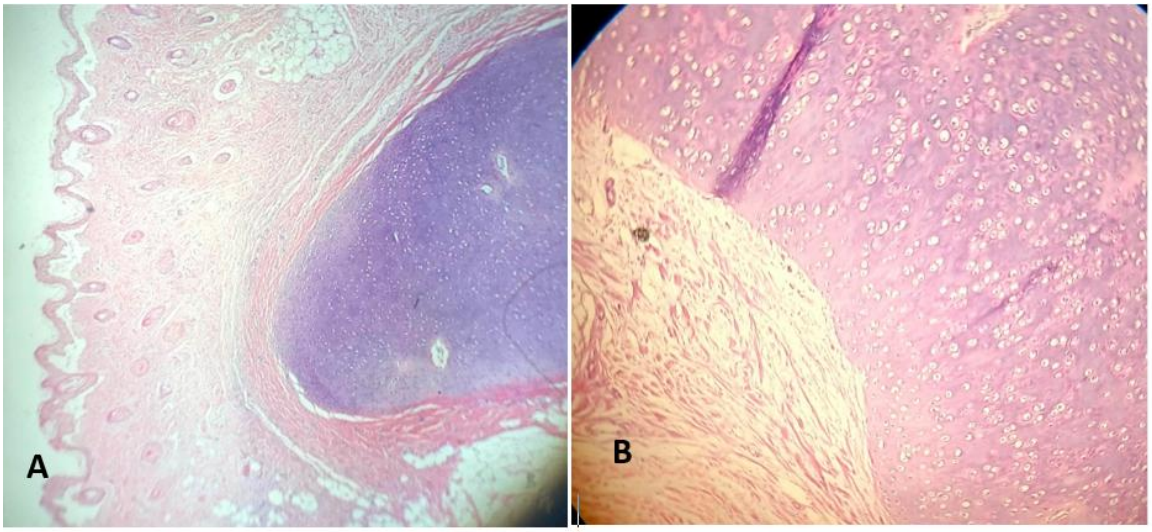

Figure 2. (A) Low-power scanning view of lesion, showing a central core of hyaline cartilage surrounded by fibrofatty tissue and hair follicles, with superficial lining of keratinized stratified squamous epithelium (H and E stains ×40). (B) Higher magnification (×200) reveals the hyaline cartilage formed of chondrocytes within lacunae embedded in a homogeneous glassy matrix.

Discussion

The pathogenesis of CCBR is linked to a developmental anomaly involving the second branchial arch [2], specifically a failure of involution of the cervical sinus [10]. Histological evidence of cartilage within the lesion supports this second branchial arch origin [10]. The largest series was reported by Atlan et al. [3], who described 17 cases, predominantly male [3]; however, our patient was female. These lesions are extremely rare in our region. Clinical suspicion often arises from the lesion's typical location along the anterior border of the sternocleidomastoid muscle [5], but physical examination and imaging modalities such as ultrasonography are insufficient alone for definitive diagnosis [8]. Therefore, microscopic examination remains crucial [9].

In the present case, although ultrasonography was essential for confirming the superficial nature of the lesion and for excluding deep fistulous tracts, it could not definitively characterize the internal cartilaginous core. Therefore, microscopic identification of a central core of mature hyaline cartilage remains an essential diagnostic criterion for

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distinguishing CCBR from other congenital lesions [8]. On a genetic basis, some studies have reported over three generations of a five-patient family with familial Chondrocutaneous branchial remnant [4], suggesting that this condition could be hereditary [4].

Histopathologically, CCBR differs from branchial cleft cysts [8], which generally have a lining of squamous or respiratory epithelium without cartilage [8]. In contrast, CCBR exhibits a skin lining with skin appendages surrounding a core of elastic or hyaline cartilage, a finding that is pathognomonic and confirms the diagnosis [8]. The differential diagnosis for congenital neck masses is broad [8]. As summarized in (Table 1), the presence of mature cartilage is the key histological feature distinguishing CCBR from other cystic malformations [10].

Table 1. Differential diagnosis of congenital neck lesions based on clinical and histopathological features

Feature	Dermoid cyst	Branchial cleft cyst	CCBR	Thyroglossal cyst
Typical location	Midline	Mid and lateral neck	Lower third of the sternocleidomastoid muscle	Midline near the hyoid bone
Consistency	Fluctuant	Fluctuant	Firm and pedunculated	Fluctuant
Associated anomalies	Rare	Rare	Cardiac anomaly ASD) and renal	Rare
Histopathology	Lined by stratified squamous epithelium with skin appendages	Epithelial lining with Lymphoid follicles	Elastic or hyaline cartilage with skin appendages	Epithelial lining with thyroid tissue
References	[8]	[8]	[8]	[8]

A critical aspect of our case was the patient's clinical history of bilateral hearing loss and a resolved atrial septal defect. The literature suggests that CCBR is associated with other systemic anomalies [10], including cardiovascular, genitourinary, visual, musculoskeletal, gastrointestinal, and respiratory abnormalities [4]. The presence of a branchial remnant alongside auditory and cardiovascular anomalies necessitates the exclusion of branchio-oto-renal (BOR) syndrome [11]. In this patient, normal renal function tests and ultrasonography results were reassuring. The association with hearing loss highlights the role of a multidisciplinary team, including pediatricians, cardiologists, and otolaryngologists [12]. The treatment for this lesion involves complete surgical excision, followed by histopathological examination [12].

Conclusion

CCBR is a unique benign lesion of the cervical region resulting from a developmental anomaly of the branchial arches during embryogenesis. To accurately differentiate CCBR from other congenital neck masses, histopathological evaluation is crucial. Due to the clinical significance of this condition, a comprehensive evaluation of the patient is recommended for proper patient management.

Informed consent statement

Written informed consent was obtained from the patient's parent /guardian for publication of this case report and any accompanying images. A copy of the retain consent is available for review by the editor-in-chief on request.

Conflict of interest: There is no conflict of interest

Conflict of interest. Nil

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