

Case Report

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Cervical Chondrocutaneous Branchial Remnants: A Case Report

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ABSTRACT

Unilateral cervical chondrocutaneous branchial remnants (CCBRs) are uncommon, but when they do occur, they usually occur at the anterior margin of the sternocleidomastoid muscle on the lateral side of the neck, whereas normal skin with a cartilage core makes up the lesions. They have no deep underlying tissues; however, they frequently adhere to the sternocleidomastoid muscle fascia. Surgical excision is the preferred treatment for a CCBR. To rule out underlying significant anomalies, a comprehensive physical examination and ultrasound evaluation of the abdomen and heart are required.

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Introduction

Cervical chondrocutaneous branchial remnants (CCBRs) are uncommon embryonic lesions that can occur unilaterally or bilaterally observed. A significant preponderance of men was previously observed, however Klockars et al. discovered no gender propensity in a 2015 review [1]. The phrase “chondrocutaneous branchial remnant” refers to the presence of heterotopic cartilage under the skin in the cervical area [2]. Only 117 cases have been recorded in the medical literature to date, with 34 of these being bilateral [3]. For the past century, the real cause of these lesions has been challenged, and the lack of published instances has hindered the investigation [4]. They also have an elastic cartilage core with cutaneous appendages that is covered by keratinizing squamous epithelium [5]. CCBRs are usually benign, although their prevalence in the general population varies greatly. Although CCBRs are rarely symptomatic, they have been linked to significant congenital abnormalities in the past. As a result, finding CCBRs can and should stimulate further investigation [6].

CCBRs were frequently discovered to be apparent indicators of more serious medical conditions [6]. Cervical chondrocutaneous branchial remnants located particularly along the anterior border of the sternocleidomastoid muscle, are known as choristomas. A choristoma is a benign tumor made up of normal tissue that has grown in inappropriate position. Cysts, sinuses, fistulae, and cartilaginous remains are all possible manifestations [7]. Cal excisions of these lesions are effective; however, swellings in the anterior Surgineck along the anterior sternocleidomastoid muscle have a long list of differential diagnoses. Establishing a proper diagnosis is mandatory before treatment can be started.

Case Presentation

We report an child of 2 years old, with a skin lesion on the lower part of the left side of the neck, anterior to the sternocleidomastoid muscle, the lesion was elastic, there was no visible discharge, and the skin on top appeared normal, the swelling was about 2 cm in length x 0.5 cm in width, Palpation revealed a non-tender mass unattached to the underlying tissue and mobile in all directions in the subcutaneous plane (figure 1). No cervical lymph nodes were palpable, and the overlying skin was partially connected to the swelling, there were no apparent congenital defects such as abnormal ear implantation, auricular pits, auricular appendages, or fistulae, there were no other members of the patient's family who had similar symptoms, the results of a general examination were normal, to identify if her CCBR had any associated anomalies, an ultrasound of her neck and cardio-abdomino-pelvic organ was performed, Ultrasound of neck showed a hypoechoic (likely cartilaginous) mass measuring 7mm x 4 mm with no significant internal vascularity identified whereas cardi- abdomino- pelvic assessment was unremarkable. Ultrasound was chosen due to its least invasiveness that also provided diagnostic benefit.



Figure 1: Cervical chondrocutaneous branchial remnant (CCBR) on the lower left side of the neck.

Patient was planned for excision of lesion Under general anesthesia, after written consent by parents and pre-anesthesia clearance, A complete surgical excision was performed, the lesion extended into the neck and was attached to the fascia of anterior part sternocleidomastoid muscle's (figure2,3), The specimen was fixed in formalin and stained with hematoxylin and eosin (H&E). A histopathological examination revealed that the lesion consisted of normal skin, subcutaneous tissue, adnexa, fat lobule, no atypia or necrosis, and piece of hyaline cartilage. (Figure 4a,4b).



Figure 2: Adherence of the CCBR to the Fascia of the Sternocleidomastoid Muscle

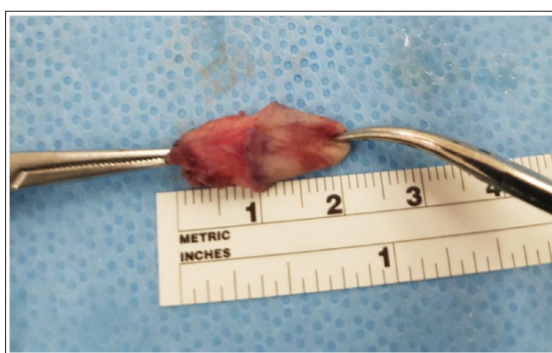


Figure 3: Adherence of the CCBR to the Fascia of the Sternocleidomastoid Muscle

Discussion

Chondrocutaneous branchial remnants are a rare entity, with only 117 cases previously reported in the English medical literature, and while they can be found in a variety of places in the human body, including the eyes, nasopharynx, middle ear, and oral cavity, the neck is the most common site of occurrence [8]. It wasn't until 1997, after research by Atlan G et al., that these rare disorders were given a name. Over the course of 13 years, 20 individuals with cervical tags were investigated, 17 of them turned out to be CCBRs, while three were ruled out as skin tags with thyroglossal ducts. This study, along with that of Begovic et al., has offered some evidence-based characteristics of diseased entity to assist medical practitioners in appropriate identification. The majority of lesions are unilateral, with bilateral lesions becoming less common, Unilateral lesions are more likely to appear on the left side of the neck than on the right side, Furthermore, one-third of patients have other defects, the most prevalent of which are in the cardiovascular and genitourinary systems, necessitating a complete physical examination [9].

Atlan and coauthors (1997) and Begovic et al. (2014), both of whom reported 17 patients, have the most patient data on

CCBRs, we were able to gather 104 individuals from the medical literature, however, there does not appear to be any clear gender predisposition based on these findings [9-10].

A recent review by Ishigaki et al. in 2020 that summarized these findings, reporting 136 patients in the literature, 85 (62.5%) of whom were male and 119 (87.5%) of whom had unilateral lesions that were primarily left sided (74/119, 62.2%) with a median age of 38.6 months at the time of resection [11].

Some studies, such as those by Dang N et al. and Klockars T et al., also reported cases of familial presentations, indicating that this anomaly has a hereditary tendency [6]. Although the exact origin of CCBRs has yet to be identified, it has been confirmed that it is an embryological remnant rather than an inflammatory or malignant origin, Previous research has had difficulty determining the actual origin of these lesions, with some believing they are auricular tissue derivatives and others hypothesizing they are branchial tissue derivatives, Six auricular hillocks appear on the first and second pharyngeal arches during the sixth week of development, which later develop into the auricle, these hillocks enlarge, differentiate, and fuse to form the primordial auricle when the 7th week begins ,It begins to move from a ventral position on the lower side of the lateral neck to the lateral cranium, where it will end up, It is attached to anterior border the sternocleidomastoid's during this migration, those who believe CCBRs are auricular in origin believe that any interruption in this migratory process can result in a remnant along this course, those who believe CCBRs are of branchial origin believe they are formed from primordial laryngeal remains of the second and third branchial arches [12].

Atlan et al. tried to delineate a pattern by looking at two things: the location of the remnants (upper versus middle and lower neck) and the type of cartilage detected (elastic versus hyaline), Auricular origin would be suggested by remnants with elastic cartilage, while hyaline cartilage would suggest a cervical source from the second or third branchial arches [8]. We were able to confirm our diagnosis of a CCBR based on the fact that our case fulfilled most of the symptoms described by Atlan et al, and a thorough physical examination, we conclude that the lesion most likely comes from the second or third branchial arch, based on the location of the remnant (lower neck) and the histology (hyaline) described before and Our case adds to the additional instance and confirm that simple surgical excision is the best treatment for this rare anomaly.

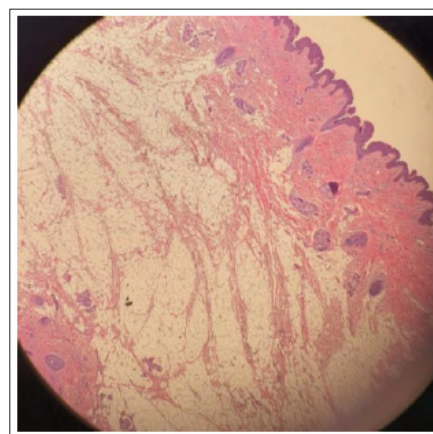


Figure 4a: 4x magnification section of low power shows skin with underlying mature adipose tissue and adnexal tissue and hyaline core which suggests a cervical origin (of the second or lower branchial arches)

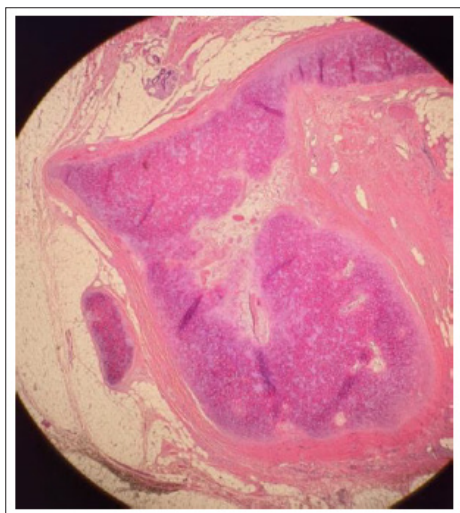


Figure 4b: 10x magnification with high power showing lobules of hyaline cartilage

Conclusion

Chondrocutaneous branchial remnants are rare, benign congenital lesions that develop over the sternocleidomastoid muscle in the middle or lower part of the neck. Knowing the embryology as well as the most prevalent presenting features can help physicians make an appropriate diagnosis in the clinic, preventing unnecessary malignant misdiagnoses, the real cause of these lesions is yet unknown, any patient suspected of having a CCBR should undergo a complete physical examination to rule out any other life threatening anomalies, and Simple surgical excision is used to treat the condition, providing a definitive diagnosis of CCBRs is important since they are frequently a visible marker of serious underlying congenital abnormalities.

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