

Bilateral cervical chondrocutaneous remnants: a case report and review of the literature

Haluk Öztürk¹, Tunç Özdemir¹, Suzi Demirbağ¹, Cüneyt Atabek¹, İlhami Sürer¹
Mükerrem Safalı², Salih Çetinkuşun¹

Departments of ¹Pediatric Surgery, and ²Pathology, Gülhane Military Medical Academy, Etlik-Ankara, Turkey

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Cervical chondrocutaneous remnants are less common lesions, which are encountered at the lateral neck. They are similar in appearance to preauricular tags, which are more frequent. Bilateral appearance of this pathology is quite uncommon. The lesions always present at birth, and are located in the middle or lower third of the lateral neck with a significant prevalence of location anterior to the sternocleidomastoid muscle. The overlying skin is similar to the surrounding neck skin and the lesion is painless, lacking any inflammation or discharge. Surgically there is no connection with deep underlying structures. The therapy of choice should be complete surgical removal.

Several associated anomalies may accompany cervical chondrocutaneous remnants. Thus these patients must be evaluated carefully in order to detect any additional anomaly.

We herein report a four-year-old patient with bilateral cervical chondrocutaneous remnant located at the inferior third of the lateral neck anterior to the sternocleidomastoid muscle. We also review the literature for patients with bilateral cervical chondrocutaneous remnants and discuss embryologic and diagnostic aspects.

Key words: chondrocutaneous branchial remnants, neck, congenital, child.

Regarding congenital lesions of the cervical region, cysts and sinuses are the most frequently encountered anomalies. Cervical chondrocutaneous branchial remnants are less common congenital lesions in contrast to chondrocutaneous tags, which locate at the preauricular region. Several congenital anomalies may accompany cervical chondrocutaneous branchial remnants; this remnant has proven in many cases to be a visible marker for more serious associated anomalies¹. Therefore, careful physical examination must be carried out and certain imaging methods may be mandatory¹.

Three cases were found with bilateral appearance of cervical chondrocutaneous branchial remnants in the English-language literature¹⁻³.

Case Report

A four-year-old boy presented with bilateral pedunculated tumors in the lower third of the neck anterior to the sternocleidomastoid

muscles. The lesions had diameters of 1 cm at right side and of 1.5 cm at left side, and were painless, skin colored and smooth (Fig. 1). No inflammation or mucoid or purulent discharge was determined and no fistulous opening was encountered. The masses had a thin pedicle



Fig. 1. Bilateral cervical chondrocutaneous remnants anterior to the sternocleidomastoid muscle.

to the neck. The physical examination of the patient revealed an otherwise healthy and well-developed child.

Anterior-posterior and lateral roentgenograms of the cervical region showed no calcification of the lesions. Cervical computerized tomography did not reveal any extension of the masses or other pathological lesions. Abdominal ultrasonography (US) findings were normal.

Both tumor-like lesions were removed under general anesthesia. The masses were resected en bloc and no extension or connection was detected beyond the platysma. In macroscopic examination of both lesions, cartilaginous tissue was detected in the deep layers covered with skin (Fig. 2). Histopathologic findings of both tumors revealed similar morphology. The lesions were covered by keratinizing squamous epithelium and had skin appendages which contained mostly hair follicles beneath (Fig. 3). Deeper layers revealed connective fatty tissue and cartilage (Fig. 4).

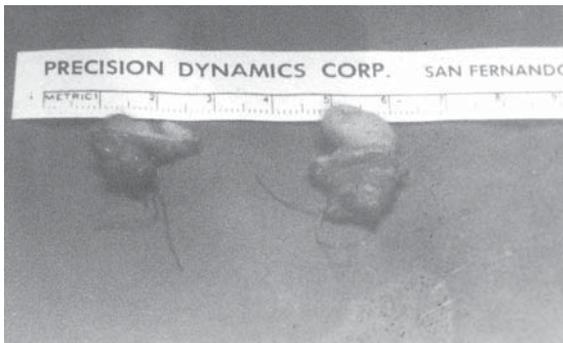


Fig. 2. Macroscopic appearance of the lesions after removal.



Fig. 3. Pedunculated lesion demonstrated squamous epithelium superficially and deeply seated cartilage islands (H&E stain, x40).

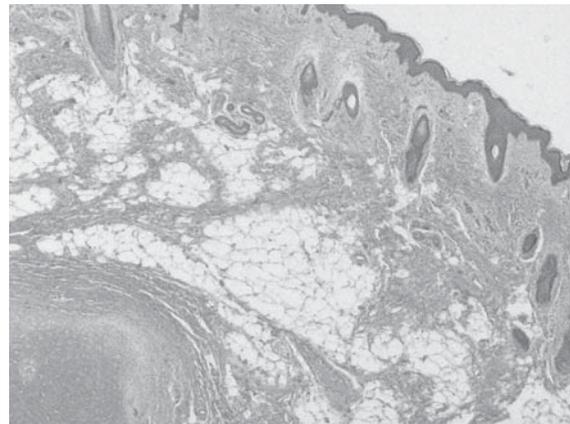


Fig. 4. Higher magnification of Figure 1 showing hair follicles, fatty tissue and cartilage (H&E stain, x100).

Discussion

Developmental tumor-like anomaly consisting of tissues foreign to the site at which they are located is defined as choristoma. Cervical chondrocutaneous branchial remnants are choristomas of the neck, which are in the group of congenital cervical masses. Most congenital anomalies of this area originate during transformation of the branchial embryonic structures that normally should disappear during maturation. Two suggestions for the embryologic source of cervical chondrocutaneous branchial remnants have been proposed. Some authors believe that they arise from ectopic auricular tissue³⁻⁵. Others have proposed that cervical chondrocutaneous branchial remnants originate from branchial tissues contributing to the formation of most cervical tissues^{6,7}. The type of the cartilage within the remnants lends a clue to the origin of the lesions. The presence of elastic cartilage would suggest an auricular (first or second branchial arch) origin or derivation from the lower neck (fourth through sixth branchial arches)⁷. Presence of hyaline cartilage would exclude an auricular origin and favor a cervical source. Histopathological examination of our specimens revealed the presence of elastic cartilage. Whether bilateral or unilateral, localizations of the lesions overall are similar to those of lesions such as branchial cysts or fistulas. The external ear forms from three hillocks derived from the first branchial arch (helical root and tragus), while the rest of the auricle originates from three more caudal hillocks of second arch origin⁸. Therefore, Atlan et al.¹ suggested that

cervical chondrocutaneous branchial remnants may be 'left behind' by the auricular hillocks (first or second branchial arch) during the migratory process of these six hillocks along the general direction of the sternocleidomastoid muscle to their final lateral facial position.

The clinical characteristics of cervical chondrocutaneous branchial remnants are: male predominance, presence at birth, and location in the middle or lower third of the neck anterior to the sternocleidomastoid muscle. The overlying skin is similar to the surrounding neck skin and the lesion is painless, lacking inflammation and discharge. Surgically there is no extension or connection with deep underlying structures of importance⁹.

Our patient is the fourth case with bilateral cervical chondrocutaneous branchial remnants (Table I)¹⁻³. While the lesions of the patients reported by Atlan et al.¹ and Braun et al.² were located at the inferior third of the neck, Kim et al.³ reported a patient with suprasternal lesions. They suggested that the lesions were accessory tragi.

Contrary to single-sided lesions, no other anomalies are associated with bilateral lesions. All patients were male and lesions were located at the inferior third of the neck in three^{1,2} and suprasternal region in one³. Consistent with the literature about the cases with bilateral involvement, there was also no associated anomaly detected in our patient. As reported by Atlan et al.¹, systemic investigation may not provide any useful additional information beyond that obtained from a complete physical examination, unless it is guided by specific clinical findings aside from certain ear malformations. Further tests and imaging techniques may be carried out if suggested by certain clinical findings.

Malignant degeneration of these lesions has not been reported. Surgical excision of these lesions should be done only for cosmetic reasons. During surgery, possible extension to any level of the neck tissues should be investigated. Although no recurrence of the lesions has been reported, complete excision is necessary.

Table I. Summary of the Reviewed Patients and Our Patient

	No of patients	Sex	Age	Location	Height	Depth	Histopathology of cartilage
Atlan et al. ¹	1	M	7 mo	Anterior to SCM	Inferior third	SCM	Elastic cartilage
Braun et al. ²	1	M	4 mo	Anterior to SCM	Inferior third	?	Elastic cartilage
Kim et al. ³	1	M	25 y	Anterior to SCM	Suprasternal	?	Elastic cartilage
Ozturk et al.*	1	M	4 y	Anterior to SCM	Inferior third	Platysma	Elastic cartilage

*Present report. SCM: Sternocleidomastoid muscle.

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