Case Report

Posttraumatic extraskeletal chondroma of the posterior neck: A systematic literature review on a rare finding and report of a case

ABSTRACT

Extraskeletal chondromas (EC) are uncommon, benign cartilaginous tumours. Most common locations are upper and lower extremities. Location in the neck is extremely rare and reported only within the anterior compartment. Data are limited to just four case reports in the paediatric population. The first case of EC in neck's posterior compartment is described herein. EC present peculiar features on imaging. Aetiology is unclear; however, trauma has been suggested as possible causative mechanism. Treatment of choice is surgical excision; recurrence is not uncommon, but additional removal seems to be resolutive. All cases in literature were asymptomatic, except for one presenting respiratory stridor. The present patient suffered from neck functional limitation and upper limb hypoesthesia. Symptoms improved after surgery in both cases. Imaging follow-up at 6 months in the present case showed no sign of recurrence. ECs are rare, benign lesions. However, they may be preoperatively misinterpreted as more malignant counterparts (both radiologically or histologically), so accurate diagnostic work-up and planning of the surgical procedure are essential.

Keywords: Extraskeletal chondroma, posterior neck, posttraumatic chondroma, soft-tissue chondroma

INTRODUCTION

Extraskeletal chondromas (ECs) are relatively uncommon benign cartilaginous tumors.[1] Morphologically, they are mainly composed of hyaline cartilage mixed with fibrous or myxoid stroma. They appear as isolated whitish encapsulated lesions without any connection to prospicient bone or periosteum.[2]

The most common locations are the upper and lower extremities.[1,3] Location in the neck is extremely rare and reported only within the anterior compartment. Data are limited to just four case reports in the pediatric population.

Not yet described are cases within neck's posterior compartment, in adult population, presenting with a large painful mass. The extreme variability in the pathological findings of this anatomic region makes it fundamental to consider even this apparently rare nosological entity in the differential diagnosis.

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The authors report the unique case of EC of the neck's posterior compartment ever described. A systematic literature review has been performed deepening the insight on this rare pathology.

MATERIALS AND METHODS

A review of the literature was performed: The PubMed database was searched using combinations of the following Medical Subject Headings and free-text terms: "soft tissue OR extraskeletal chondroma." Reference list of included studies was searched for further additions. Inclusion criteria were full-text availability and description of a case or of a case series comprising extraskeletal chondroma of the neck's soft tissues. Only articles in English were considered. Publications describing chondromas of the larynx were excluded due to their peculiarity and low relation to the aims of our work.

RESULTS

The search produced a total of 563 headings. After screening using inclusion and exclusion criteria, and after removal of duplicates, a total of 4 articles describing EC of the anterior compartment of the neck were retrieved [Table 1].

Illustrative case

A 37-year-old woman fell while riding her horse 9 years before presenting to our center. She suffered a distortion trauma of the cervical spine for which she did not undergo diagnostic examinations. After some months, she reported the development of an enlarging lesion in the posterior part of her neck. She finally underwent magnetic resonance imaging (MRI) examination 6 years before presentation, which showed a median-left paramedian mass (2.5 cm × 1 cm × 3 cm) in the paravertebral muscle compartment, that appeared iso-hypointense in T1-weighted images and hyperintense in T2-weighted images. The patient did not seek a specialist attention at the time.

A subsequent MRI, repeated just before referral to a neurosurgeon, showed a marked dimensional growth (4 cm × 4 cm × 5.7 cm) and a change in composition, since the lesion appeared multiloculated with adipose tissue-like components, with inhomogeneous contrast enhancement [Figure 1]. A computed tomography (CT) scan showed foci of calcification inside the lesion [Figure 1]. On clinical examination, a large, mobile, nonpulsatile, nontender mass was evident, which limited neck mobility. Neurological examination revealed a left-hand hypoesthesia without clear radicular distribution. Moreover, the patient reported paresthesia at night on her left forearm and hand.

The lesion was surgically excised. Intraoperatively, it appeared firm and well demarcated from surrounding tissue and was removed *en bloc* [Figures 2 and 3]. Postoperatively, no new neurological deficit was observed, and the patient reported an improvement in neck mobility and was discharged after a short observation time.

The histopathological analysis confirmed the suspicion of EC, showing a mesenchymal neoplasm characterized by a fibrous capsule and cartilage lobules separated by vascularized fibrous tissue. Chondrocytes were well differentiated and showed no atypia [Figure 3]. Ki67 immunohistochemical analysis result was <1%.

A follow-up MRI 6 months after the surgical procedure showed no recurrence nor residual of the lesion, and the patient is doing well at the last examination.

Table 1: Literature review

Author	Age	Sex	Congenital	Location	Symptoms	US	CT	MRI	Treatment	Recurrence
Kamysz *et al*., 1996[4]	5	Female	No	Anterior neck (from thyroid cartilage to sternal notch) with trachea displacement	Inspiratory and expiratory stridor	Solid heterogeneous	Several punctate calcifications; heterogeneous contrast enhancement	Not performed	Excision	Not reported
Divino *et al*., 1996[5]	3	Male	No	Midline submental	Asymptomatic	Solid heterogeneous; no calcifications	Not performed	Not performed	Excision	Not reported
Ikeda *et al*., 2015[6]	5	Male	Yes	Midline anterior	Asymptomatic	Solid	No calcifications	Not performed	Excision	No (6 months)
Temsamani *et al*., 2016[7]	12	NR	Yes	Bilateral over the sternoclei domastoid muscle	Asymptomatic	Not performed	Not reported	Not performed	Excision	Not reported

CT - Computed tomography; MRI - Magnetic resonance imaging; US - Ultrasonography; NR - Not reported

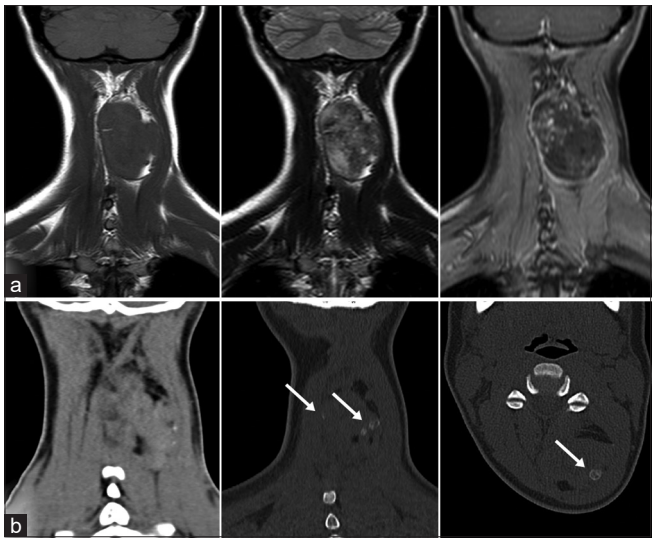

Figure 1: Neuroradiological findings. (a) Magnetic resonance imaging (from left to right): T1-weighted coronal view showing a hypo-isointense lesion of the posterior neck compartment; T2-weighted coronal view with the hyperintensity typical of these lesions; post-gadolinium coronal scan in which inhomogeneous contrast enhancement is evident. (b) Computed tomography scan (from left to right): Coronal view on soft-tissue sequences, coronal and axial view on bone sequences showing an isodense mass in the posterior compartment of the neck, containing calcifications (white arrows)

DISCUSSION

ECs are relatively infrequent, benign tumors, composed by cartilaginous tissue. They are mostly diagnosed between 30 and 60 years of age,[1] representing 1.5% of all soft-tissue tumors.[8] The most frequent locations are the upper and lower extremities, with more than 80% found in hands and feet.[1,3,8]

They usually present as slowly growing lesions, occasionally causing pain, well defined and demarcated from surrounding structures, firm to the touch. They are reported to be seldom larger than 3 cm.[3]

ECs present peculiar features on imaging; however, differential diagnosis must include malignant lesions such as chondrosarcoma and extraosseous osteosarcoma. On CT imaging, they appear as iso- or hyperdense well-circumscribed masses, and they show various degrees of mineralization in 33%–70% of cases, going from ring calcifications to ossification.[9-11]

At MRI, ECs show hyperintense signal on T2-weighted sequences and low-to-intermediate signal intensity on T1-weighted sequences.[9,12] Patterns of contrast-enhancement may vary:[13] it is usually septal or peripheral, as opposed to malignant cartilaginous tumors in which contrast enhancement is often intense and homogeneous.[12]

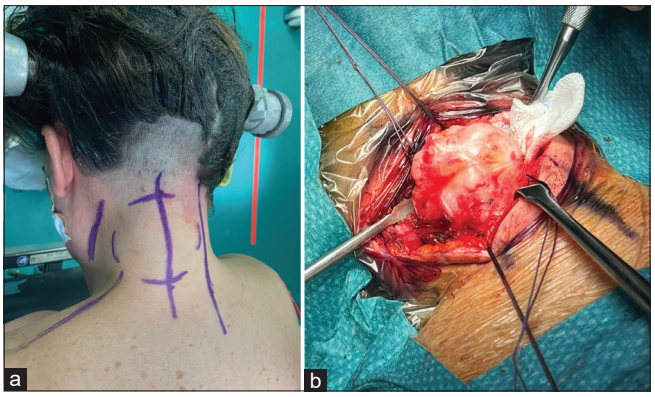

Figure 2: Intraoperative view. (a) Patient's neck before incision; (b) surgical excision

Treatment of choice is surgical excision; recurrence is common, being reported between 15% and 25%, but an ulterior surgical removal seems to be usually resolutive.[10,11] A 9-year mean time to recurrence has been previously reported.[14]

Histopathologically, they appear as well definite masses with intralesional lobulations and areas of hyaline cartilage, where sometimes areas of ossification or calcification may be present. The matrix may be hyaline, fibrous, or fibrohyaline. Cellular atypia and chondroblasts can sometimes be found; however, there is no invasion of surrounding tissue.[1,3,8] Myxoid changes and atypia may be interpreted as a hallmark of chondrosarcoma for intraosseous lesions; however, ECs do not show aggressive behavior or metastasis.[8,10,15,16]

ECs in the neck represent a minority of all cases, with most of them described in the larynx.[2] We present a rare case of an EC of the neck's posterior compartment, which according to the targeted literature review is the first ever reported. Previous reports, describing neck EC outside the larynx, illustrated lesions of the anterior neck[4-6] and bilateral lesions over sternocleidomastoid muscles[7] [Table 1].

The patients were all way younger than the mean age at presentation usually reported for EC (3–12 years old vs. 30–60 years old), while our patient fell within that range (37 years old).

All cases were asymptomatic at presentation, except for a 5-year-old girl seeking medical attention for respiratory stridor, presenting with a lesion dislocating the trachea.[4] Our patient suffered from a limitation of neck's range of motion and from hypoesthesia in her upper left limb. It is well described that ECs are usually asymptomatic, seldom tender.[1,8] However, they may cause disturbances related to the mass effect they exert on surrounding structures; in our

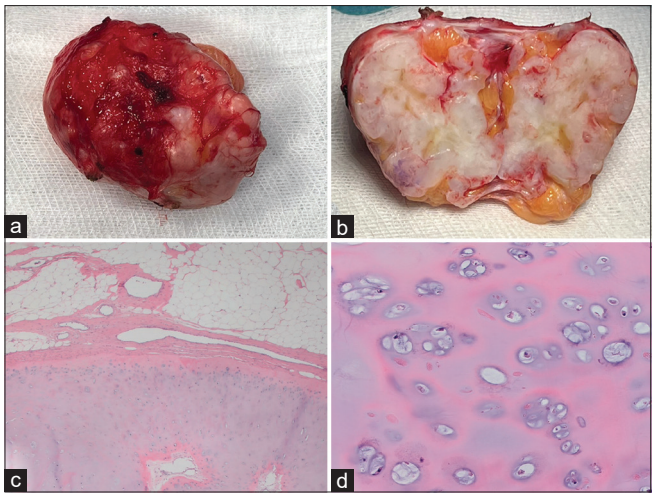

Figure 3: Pathological findings. (a) Surgical sample (dorsal surface); (b) Surgical sample; (c) microscopic view: hyaline capsulated cartilaginous tumor in adipose tissue (H and E, ×25); (d) Microscopic view: multilobulated lightly cellular cartilaginous tissue with well-differentiated chondrocytes (H and E, ×100)

case, the lesion was limiting the function of cervical posterior muscles, causing an obstacle to neck movement, as well as a compression on emerging cervical roots.

Excision was performed in all cases, and histologically, the lesions did not show any sign of malignancy, mitosis, or atypia. Similarly, our case did not show any atypia and Ki67 was <1%.

Follow-up was available just for one patient, a 5-year-old boy who did not show any sign of recurrence at the last visit, 6 months after surgery.[6] Our patient appears to be free of recurrence at the last MRI, performed 6 months after surgery. This may be too short of a time to declare a patient free of disease, since the median time for recurrence has been reported as 3.6 years, with a case of a recurrent chondroma presenting 38 years after initial surgery.[11] Chiu and Rasgon reported a mean time to recurrence of 9 years.[14]

Even though the etiology is unclear, it has been hypothesized that extra-articular remnants of synovial tissue may face a metaplastic transformation into cartilage: this idea is supported by the observation that synovial and soft-tissue chondromas display similar karyotypic abnormalities, which are not seen in bone chondromas (such as periosteal chondromas and enchondromas).[17] The metaplastic differentiation may be triggered by irritating stimuli, since a history of trauma has been reported by patients with extremity soft-tissue chondromas.[18,19] Our patient suffered a distraction trauma to her neck a few months before the appearance of the mass. Significant trauma of the neck may happen less frequently than in the hands, feet, or knees, hence the higher frequency of EC in these latter districts.

CONCLUSION

ECs are relatively rare, benign lesions. Treatment of choice is surgical excision, and recurrences are not frequent. However, they may be preoperatively misinterpreted as more malignant counterparts (both radiologically and histologically), so accurate diagnostic work-up and planning of the surgical procedure are key for these masses.

Neck location has been seldom described, while cervical paravertebral lesions were not known until today. Given the extreme variability in the pathological findings of this anatomic region, it is fundamental to consider even this apparently rare nosological entity in the differential diagnosis.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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