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Case Report

Chondromyxoid Fibroma of the Clavicle: A Case Report of a Rare Clinical Entity

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Keywords
Chondromyxoid fibroma · Clavicle · Benign cartilage-forming tumor · Diagnosis · Treatment · En bloc resection

Abstract
Introduction: Chondromyxoid fibroma is a rare benign cartilaginous tumor that accounts for less than 1% of all bone tumors and involves long bones of the lower extremities more frequently. Therefore, the clavicle is a rare location of involvement for this entity. ***Case Presentation:*** The authors report a case of a diaphyseal chondromyxoid fibroma of the right clavicle in a 30-year-old male that was submitted to en bloc resection of the lesion and reconstruction with an autologous tricortical graft from the iliac crest. The post-operative period was uneventful, and the patient had regained excellent function of this right shoulder. At 2 years of follow-up, there was no evidence of a recurrence of the disease. ***Conclusion:*** Chondromyxoid fibroma of the clavicle should be included in the differential diagnosis of an indolent growing mass in this anatomic location.

Introduction

Chondromyxoid fibroma (CMF) is a rare benign tumor of cartilaginous origin that accounts for less than 1% of all primary bone tumors [1, 2]. The incidence of CMF displays a bimodal appearance, with a peak between the second and third decades and a later peak between the fifth and seventh decades of life [1]. CMF seems to affect both males and females in the same ratio [1, 3, 4]. The most common location for the occurrence of CMF is the proximal tibial metaphysis, followed by the distal femur and pelvis. The upper limb is an uncommon location of involvement. Therefore, the presentation of CMF in the clavicle is infrequent [1].

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The histological features of CMF englobe a lobulated architecture composed of spindle- or stellate-shaped cells rich in myxoid or chondroid intercellular material [5]. The frequent presence of cellular atypia with pleomorphic hyperchromatic cellular nuclei and a high index of proliferation can lead to a misdiagnosis of myxoid chondrosarcoma or chondroblastoma [1, 2, 6–8]. Although CMF is considered a benign tumor because of metastasis absence, it can present local aggressive behavior with an estimated recurrence rate of 25% [2, 9].

A search in *PubMed* using the Mesh terms "fibroma" and "clavicle" revealed 8 case reports of clavicle CMF described in the literature, which supports the rarity of this entity. The CARE Checklist has been completed by the authors for this case report, and it is attached as online supplementary material (for all online suppl. material, see https://doi.org/10.1159/000536138).

Case Report

The authors report a case of a 30-year-old Caucasian male, bus driver, right-hand dominant, with a history of diaphyseal fracture of his right clavicle during his childhood treated conservatively in another hospital. He had no other medical history of note. The patient presented to our department with sudden pain and tenderness after an indirect trauma to his right shoulder. He denied previous right shoulder pain before the referred trauma. Clinical examination revealed notable edema and a palpable painful mass of hard consistency of 6 × 3 cm dimension, not adherent to the skin, localized over the middle third of the right clavicle. There was an evident limitation in the general shoulder range of motion due to pain. The patient's general physical status was good; there were no systemic symptoms referred, such as fever, night sweats, or weight loss, and there was no evidence of lymphadenopathy. There were no neurological or vascular alterations. The remaining physical examination was unremarkable. The patient was immobilized with a shoulder sling.

The plain radiographs showed an incomplete pathological diaphyseal fracture within an expanded osteolytic lesion of the middle third of the right clavicle. The lesion was well-circumscribed with areas of peripheric cortical thinning, central erosion, and septal appearance (shown in Fig. 1). There was no evident periosteal reaction. Computed tomography revealed an osteolytic expansive lesion of 6 × 3.5 cm dimension with chondroid matrix core and internal septa, with an antero-superior incomplete pathologic fracture. There was no soft-tissue involvement noted or signs of calcification (shown in Fig. 2). Magnetic resonance imaging revealed an isointense appearance lesion in T1-weighted imaging and hyperintense appearance in T2 with lobulated heterogeneous caption pattern after contrast enhancement (shown in Fig. 3). There was no involvement of the brachial neurovascular bundle which was confirmed with angio-CT imaging.

The laboratory tests, including a hemogram with complete blood count, electrolytes, erythrocyte sedimentation rate, and C-reactive protein, revealed no alterations. CT-guided biopsy was performed, and the sample was sent for histopathological examination that revealed moderate cellularity composed of fusiform cells with chondromyxoid stroma and without atypia. There was no evidence of mitosis, areas of necrosis, or hemorrhage. These findings were considered consistent with fibrous-predominant CMF.

After 2 weeks of immobilization, the patient presented regression of the edema but still displayed tenderness to palpation of the middle third of the right clavicle. There was also an explicit limitation of active range of movement, being capable of 100 degrees of anterior elevation.

The patient underwent en bloc resection of the lesion with subtotal cleidectomy, preserving the clavicle insertion of the trapezoid and conoid ligaments that were not

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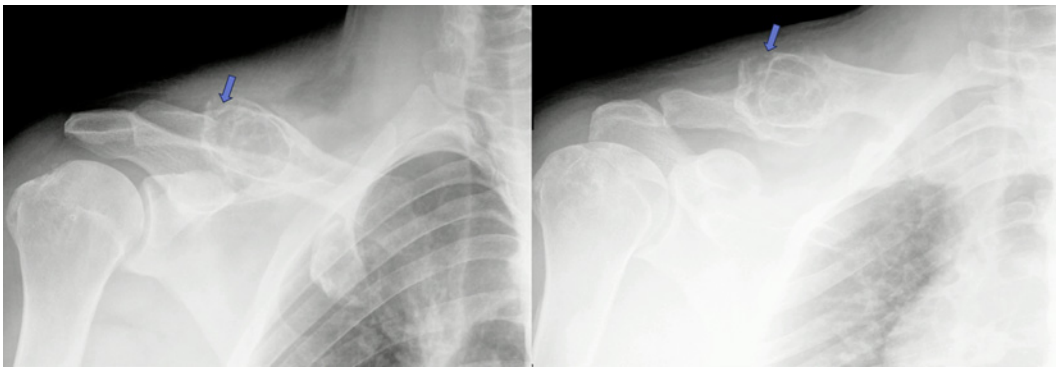

Fig. 1. Plain radiographs showing an incomplete pathological diaphyseal fracture within an expanded osteolytic lesion of the middle third of the right clavicle (blue arrow pointing to the fracture).

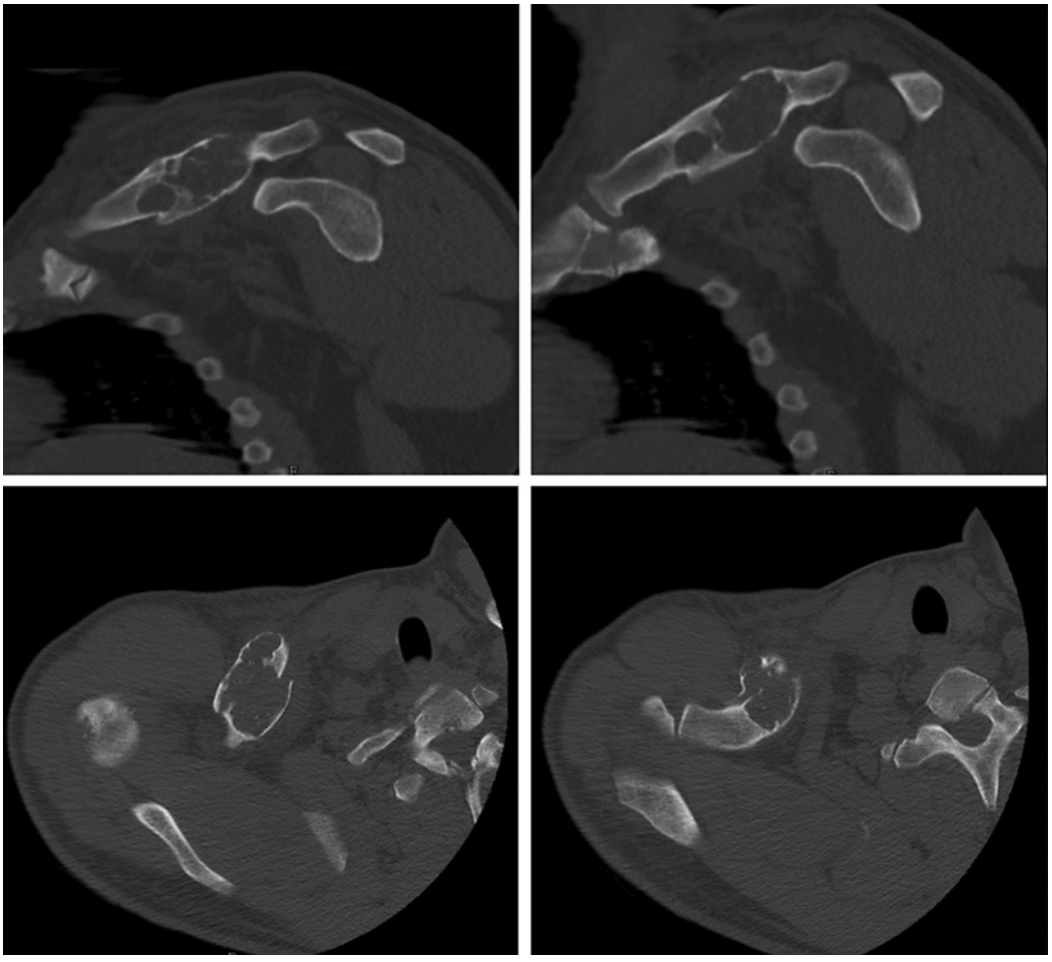

Fig. 2. Computed tomography revealing an osteolytic expansive lesion of 6 × 3.5 cm dimension with chondroid matrix core and internal septa, with an antero-superior incomplete pathologic fracture.

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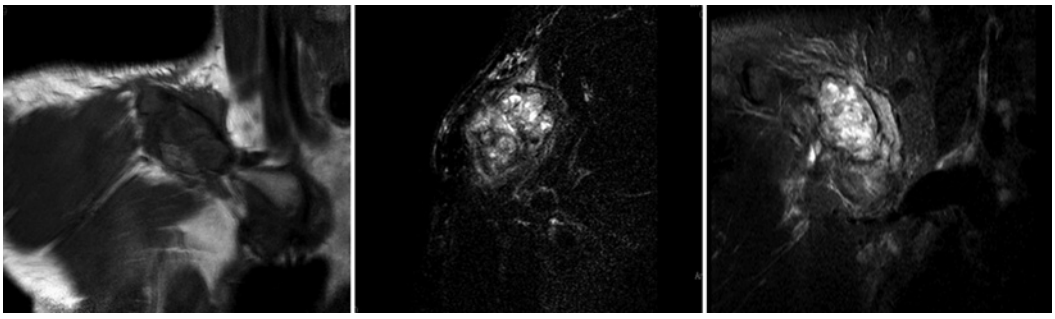

Fig. 3. Magnetic resonance imaging eliciting an isointense appearance lesion in T1-weighted imaging and a hyperintense appearance in T2 with a lobulated heterogeneous caption pattern after contrast enhancement.

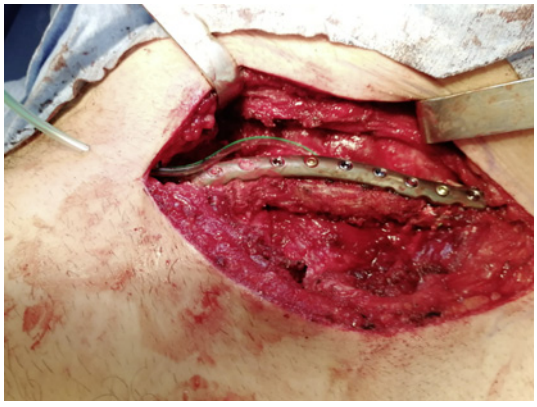

Fig. 4. Intra-operative image after en bloc resection of the lesion with subtotal cleidectomy and reconstruction with an autologous tricortical graft donated from the ipsilateral iliac crest and fixation with locking plate.

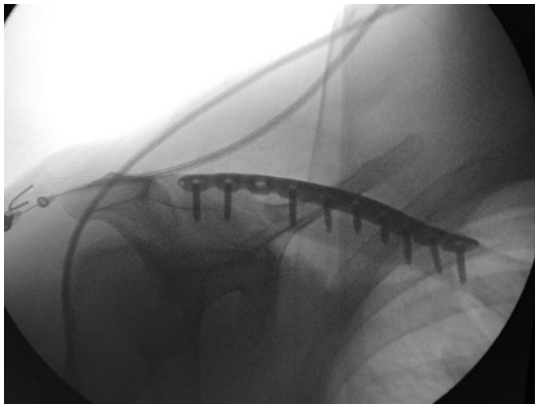

Fig. 5. Intra-operative fluoroscopy.

involved by the lesion, reconstruction with autologous tricortical graft donated from the ipsilateral iliac crest and fixation with locking plate (shown in Fig. 4, 5). The resected lesion had 8.5 × 4.4 × 3.8 cm of dimension (shown in Fig. 6). The histopathological examination of the resected specimen revealed areas of high cellular index of fusiform and stellate cells with chondromyxoid content on its nuclei with establishing the diagnosis of clavicle CMF (shown in Fig. 7). The immunohistochemical study showed low expression of SMA and S100 protein. There was no lesion detected in the extremities of the resected specimen.

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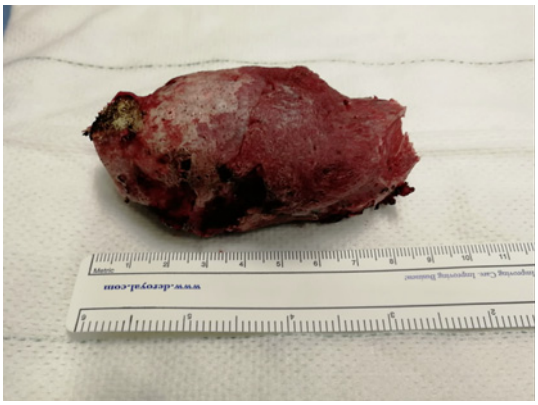

Fig. 6. Image of resected lesion.

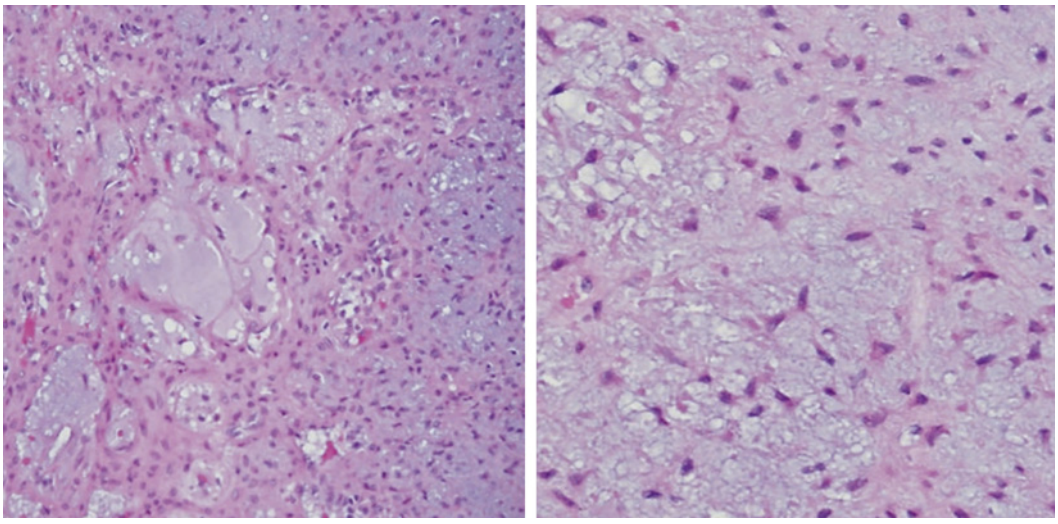

Fig. 7. Magnified image of histopathological examination of the central portion of the resected specimen showing spindle and stellate cells with myxoid stroma.

The post-operative period was uneventful. The patient underwent a rehabilitation program starting with passive and active assisted mobilization of the shoulder 4 weeks after surgery and regained excellent function of this right shoulder (shown in Fig. 8). At 1 year of follow-up, the patient presented an active range of motion of 160° of anterior elevation, 160° of abduction, 60° of external rotation, and 80° degrees of internal rotation. At 2 years of follow-up, the patient presented an assessed constant score of 80, and the last radiographs and CT scan showed consolidation of bone graft and no evidence of recurrence of the disease (shown in Fig. 9).

Discussion

To the author's knowledge, there are 8 cases of CMF of the clavicle reported in the literature until this date, which reflects the rarity of this entity. The differential diagnosis of CMF should include myxoid chondrosarcoma, chondroblastic osteosarcomas, fibrous dysplasia, chondroblastomas, and osteomyelitis [9]. In order to achieve a correct diagnosis, a thorough clinical history, a meticulous physical examination, and a careful radiologic correlation are needed [10]. The most common clinical presentation of CMF in the clavicle is the progressive onset of pain and

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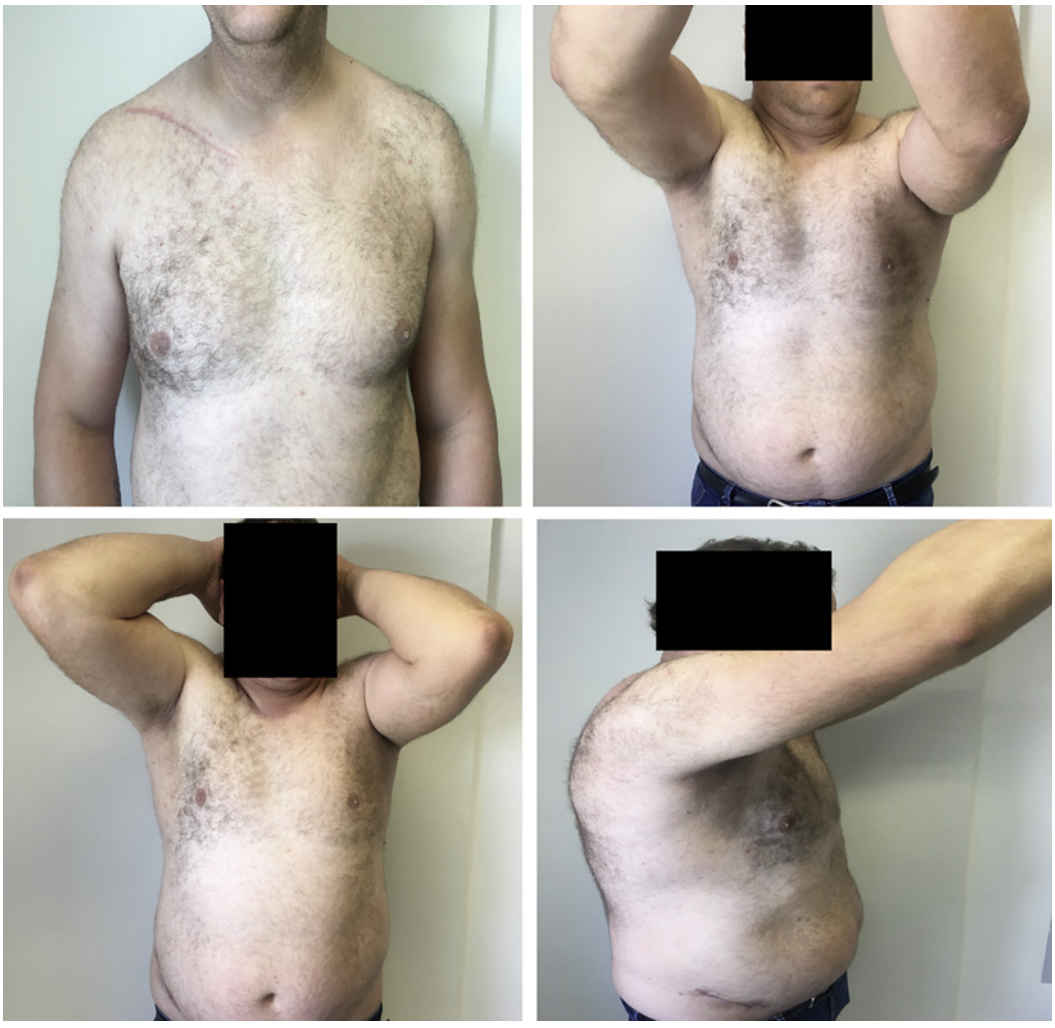

Fig. 8. Images of active right shoulder range of motion at 6-month follow-up.

swelling due to the slow growth of the tumor [1]. Patients may also report restricted shoulder range of motion caused by mechanical compression. In our case, the patient was asymptomatic before the reported trauma. Radiologically, CMF may show locally aggressive behavior with cortical thinning, bone erosion, and destruction. On magnetic resonance imaging, CMF presents isointense on T1-weighted images and hyperintense on T2-weighted images [1]. The final diagnosis is established by the histopathological findings, either by needle biopsy or open biopsy, when it is not possible to obtain adequate samples with needle biopsy.

Regarding treatment modalities, CMF can be treated with curettage complemented by the use of phenol or with en bloc resection with or without reconstructive procedures. The recurrence rate established in the literature following curettage treatment is 25%, although there is some evidence that the use of phenol as an adjunct to curettage can reduce local recurrence [1, 10]. En bloc resection is considered the gold-standard treatment with a negligible risk of recurrence but can produce more morbidity. There is no current consensus regarding indications to perform reconstructive procedures after clavicle excision [1, 10]. Wood [11] reported good functional outcomes after clavicular excision without reconstruction. Kalbermatten et al. [12] described a clavicular reconstruction with a vascularized anatomic osteotomized fibula in order to provide stability to the shoulder girdle.

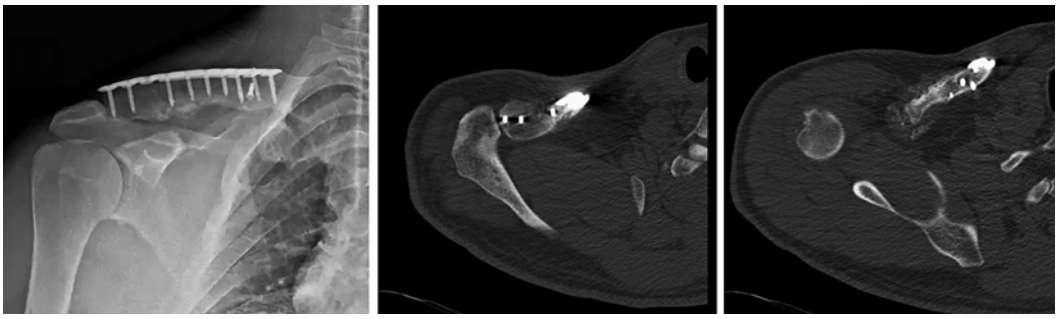

Fig. 9. Radiographs and CT scan at 2-year follow-up.

In our case, due to the young age of the patient and the dimensions of the lesion, the authors opted to perform en bloc resection and reconstruction with an autologous tricortical iliac crest bone graft and stabilization with a locking plate. The primary goal of this procedure was to treat the lesion, avoid recurrence, and regain shoulder girdle stability, preventing weakness or plexus irritation. The patient obtained an excellent functional outcome with a pain-free shoulder range of motion. After 2 years of follow-up, radiographs and CT scan show consolidation of the bone graft and no evidence of recurrence.

Conclusion

The authors report a rare CMF of the clavicle treated with en bloc resection and reconstruction with an autologous tricortical iliac crest bone graft. The diagnosis of this entity requires a high index of suspicion and a correlation of physical examination findings, radiologic imaging modalities, and histopathologic features. CMF of the clavicle should be included in the differential diagnosis of an indolent growing mass in this anatomic location.

Statement of Ethics

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editor of this journal upon request. This study protocol was reviewed and approved by the Ethical Commission of Centro Hospitalar do Tâmega e Sousa on the July 4, 2023.

Conflict of Interest Statement

The authors have no competing interests to declare.

Funding Sources

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Author Contributions

Diogo Soares: investigation, manuscript original drafting, and editing; Francisco Bernardes: investigation and manuscript review; Nuno Vieira Silva: manuscript review; Marta Cerqueira Silva: manuscript review and editing; and Daniel Lopes: manuscript review and supervision.

Data Availability Statement

All data generated or analyzed during this study are included in this article and its online supplementary material. Further inquiries can be directed to the corresponding author.

References

1 Aggarwal A, Bachhal V, Soni A, Rangdal S. Chondromyxoid fibroma of the clavicle extending to the adjacent joint: a case report. J Orthop Surg. 2012;20(3):402–5.
2 Pattamapaspong N, Peh WC, Tan MH, Hwang JS, Tan PH. Chondromyxoid fibroma of the distal clavicle. Pathology. 2006;38(5):464–6.
3 Nakazora S, Kusuzaki K, Matsumine A, Seto M, Fukutome K, Uchida A. Case report: chondromyxoid fibroma arising at the clavicular diaphysis. Anticancer Res. 2003;23(4):3517–22.
4 Sakamoto A, Tanaka K, Matsuda S, Hosokawa A, Harimaya K, Yoshida T, et al. Chondromyxoid fibroma of the clavicle. J Orthop Sci. 2006;11(5):533–6.
5 Fletcher CD, Unni KK, Mertens F, editors. World Health Organization classification of tumors, pathology, and genetics. Tumors of soft tissues and bone. Lyon: IARC Press; 2002. p. 83–4. ISBN: 9-2832-2413-2.
6 Bhamra JS, Al-Khateeb H, Dhinsa BS, Gikas PD, Tirabosco R, Pollock RC, et al. Chondromyxoid fibroma management: a single institution experience of 22 cases. World J Surg Onc. 2014;12:283.
7 Slotcavage RL, Dickson BC, Ogilvie CM. Chondromyxoid fibroma involving the metacarpophalangeal joint. Orthopedics. 2009;32(4).
8 Unni KK, Inwards CY. Dahlin's bone tumors: general aspects and data on 10,165 cases. 6th ed.50–9. Philadelphia: Lippincott Williams & Wilkins; 2010. ISBN: 978-0-7817-6242-7.
9 Vianney Hope JM, Sane JC, Souleymane D, Mugabo F, Bitega JP, Nikiema AN. Chondromyxoid fibroma of the distal clavicle: report of an additional case at very unusual anatomic location. JOJ Orthoped Ortho Surg. 2018;2(2):555581.
10 Arora S, Kashyap A, Maini L, Prakash A, Saran RK. Chondromyxoid fibroma of clavicle presenting as radiological disappearance of bone. Ann Natl Acad Med Sci. 2023;59(2):109–14.
11 Wood VE. The results of total claviculectomy. Clin Orthop Relat Res. 1986;207:186–90.
12 Kalbermatten DF, Haug M, Schaefer DJ, Wolfinger E, Schumacher R, Messmer P, et al. Computer aided designed neo-clavicle out of osteotomized free fibula: case report. Br J Plast Surg. 2004;57(7):668–72.