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Case Report

A rare bone tumor of the mandible: Chondromyxoid Fibroma

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Abstract

Chondromyxoid fibroma is a rare benign chondrogenic bone neoplasm characterized by chondroid and myxoid stromal components, multinucleated giant cells, and distinctive mineralized areas. It predominantly affects the long bones of young adults, while involvement of the maxillofacial region is exceedingly uncommon, with fewer than 40 cases reported in the English-language literature. Radiographically, the lesion may simulate cemento-ossifying fibroma, benign odontogenic tumors exhibiting mixed radiolucent-radiopaque features, osteoid osteoma and chondrosarcoma. We report a case of mandibular chondromyxoid fibroma in a 29-year-old woman. Although the diagnosis is primarily established by routine hematoxylin and eosin (H&E)-stained sections, immunohistochemical results for α-smooth muscle actin (α-SMA), vimentin, S-100 protein, CD10, Ki-67, and p53 are presented. Six-year clinical and radiographic follow-up is also reported.

Keywords: Chondromyxoid fibroma; mandible; immunohistochemistry; chondrogenic neoplasms; jaw neoplasms.

Statement of Clinical Significance

Because chondromyxoid fibroma is extremely rare in the jaws and may clinically and radiographically mimic other benign maxillofacial tumors, accurate diagnosis requires careful correlation of clinical, radiographic, and histopathological findings. Awareness of this entity helps prevent misdiagnosis and inappropriate management.

1. Introduction

Chondromyxoid fibroma (CMF) is a rare benign chondrogenic neoplasm of bone, first described by Jaffe and Lichtenstein in 1948, accounting for less than 1% of all primary bone tumors.[1,2] It predominantly arises in the metaphysis of long bones, particularly the proximal tibia, distal femur, and fibula, and is most frequently diagnosed during the second and third decades of life without a definitive sex predilection.[1,22]

Although CMF is more common in young adults, reported cases range from infancy to 88 years of age.[14,15] According to the current WHO Classification of Soft Tissue and Bone Tumours (5th edition, 2020), CMF is a benign chondrogenic neoplasm characterized by a distinctive

lobulated architecture composed of chondroid, myxoid, and fibrous tissue containing spindle-shaped cells and scattered multinucleated giant cells.[22,23]

Craniofacial involvement is exceedingly uncommon and represents a diagnostically challenging subset of CMF. Reported sites include the mandible, maxilla, zygomatic bone, nasal septum, maxillary sinus, and temporomandibular joint.[3,7,15,18] The posterior mandible appears to be the most frequently affected jaw site.[2,8,17] A review of the literature published between 2010 and 2025 identified only 19 well-documented cases involving the jaws, highlighting the rarity of this entity and the limited evidence available to guide diagnosis and treatment.[3–21]

Clinically, CMF usually presents as a slowly enlarging intraosseous lesion that may remain asymptomatic for long periods, often resulting in delayed diagnosis.[6,9,16] When symptoms occur, pain, facial asymmetry, cortical expansion, tooth displacement, and paresthesia may be observed, particularly in larger mandibular lesions.[12,14]

Radiographically, CMF typically presents as a well-defined unilocular or multilocular radiolucent lesion containing internal radiopaque masses and exhibiting varying degrees of cortical thinning. Computed tomography (CT) and cone-beam computed tomography (CBCT) are valuable for assessing the extent of the lesion and the integrity of the cortical bone.[9,13] However, the imaging features are nonspecific and may resemble those of fibro-osseous lesions, particularly cemento-ossifying fibroma. Odontogenic myxoma, ameloblastoma, and other benign mixed odontogenic tumors should also be considered in the differential diagnosis.[24–26]

Histopathologically, CMF exhibits a characteristic lobulated architecture composed of chondroid tissue containing stellate cells, myxoid areas with spindle-shaped cells, and multinucleated giant cells located at the interface between these two components.[1,2,28] Owing to its cellular heterogeneity and chondroid differentiation, CMF may be misdiagnosed as chondrosarcoma or other cartilaginous neoplasms of the jaws.[13,28] Immunohistochemistry plays an ancillary role in the diagnosis, whereas recurrent GRM1 gene rearrangements have recently emerged as a characteristic molecular alteration that supports its neoplastic nature.[13,14,22]

The present study describes a rare case of mandibular CMF in a 29-year-old woman and details its clinical, radiographic, histopathological, immunohistochemical, and therapeutic features in the context of the contemporary literature.

2. Case Report

A 29-year-old woman was referred to the Oral and Maxillofacial Medicine and Pathology Service at the Faculty of Stomatology, Universidad Peruana Cayetano Heredia, with a complaint of a slowly enlarging swelling involving the basal cortical bone adjacent to tooth 46, associated with

mild tenderness on palpation. Clinical examination revealed obliteration of the buccal vestibular sulcus and slight expansion of the inferior border of the mandible in the affected area. No spontaneous pain or paresthesia was reported, and there was no history of trauma or odontogenic infection.

Panoramic radiography and cone-beam computed tomography (CBCT) revealed a well-defined, corticated, oval-shaped lesion of mixed radiodensity. A hypodense area surrounded a centrally located irregular hyperdense mass. Thinning and mild expansion of both the buccal and lingual cortical plates were observed. The mandibular canal was displaced superiorly; however, its cortical boundaries remained intact (Fig. 1A–D).

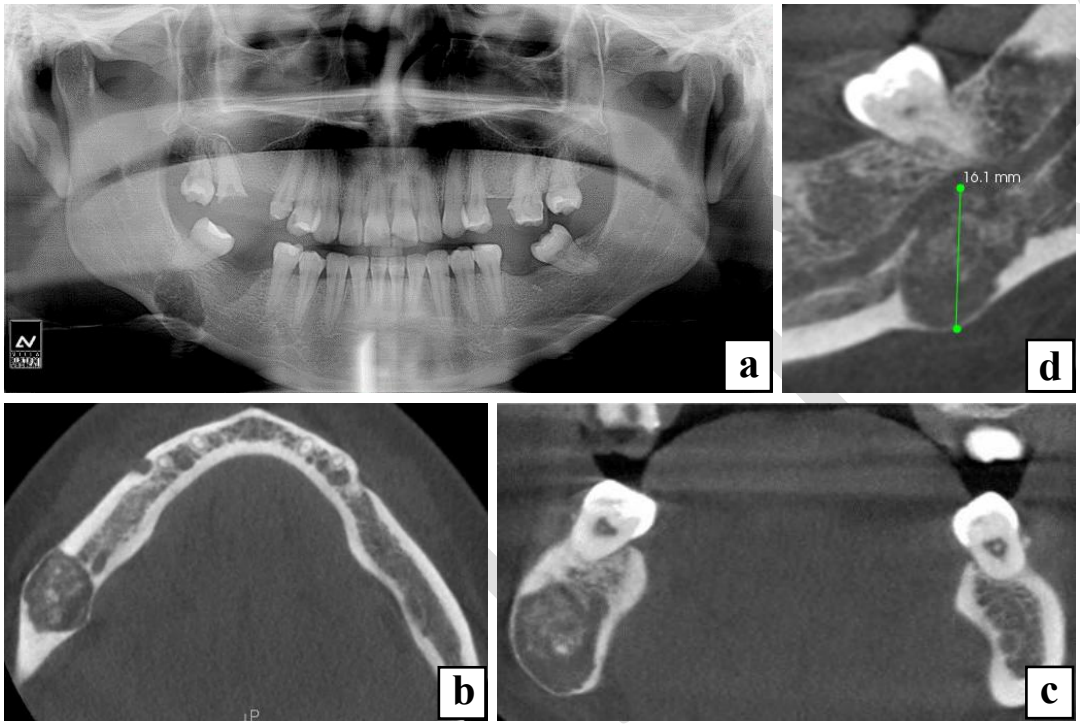
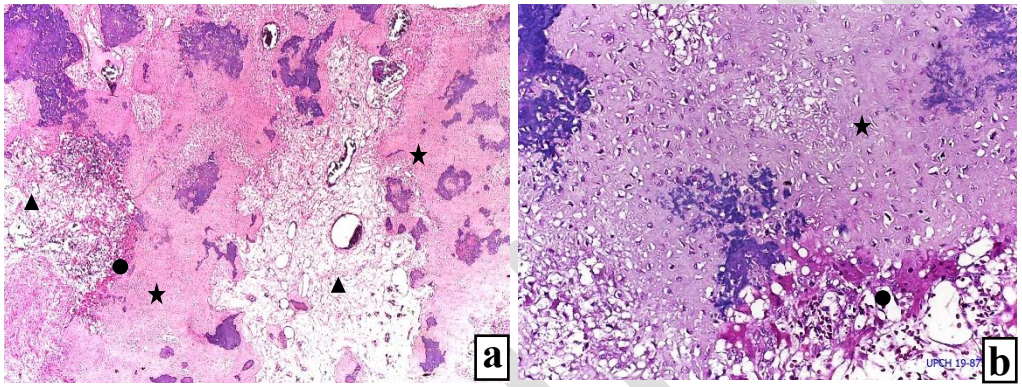


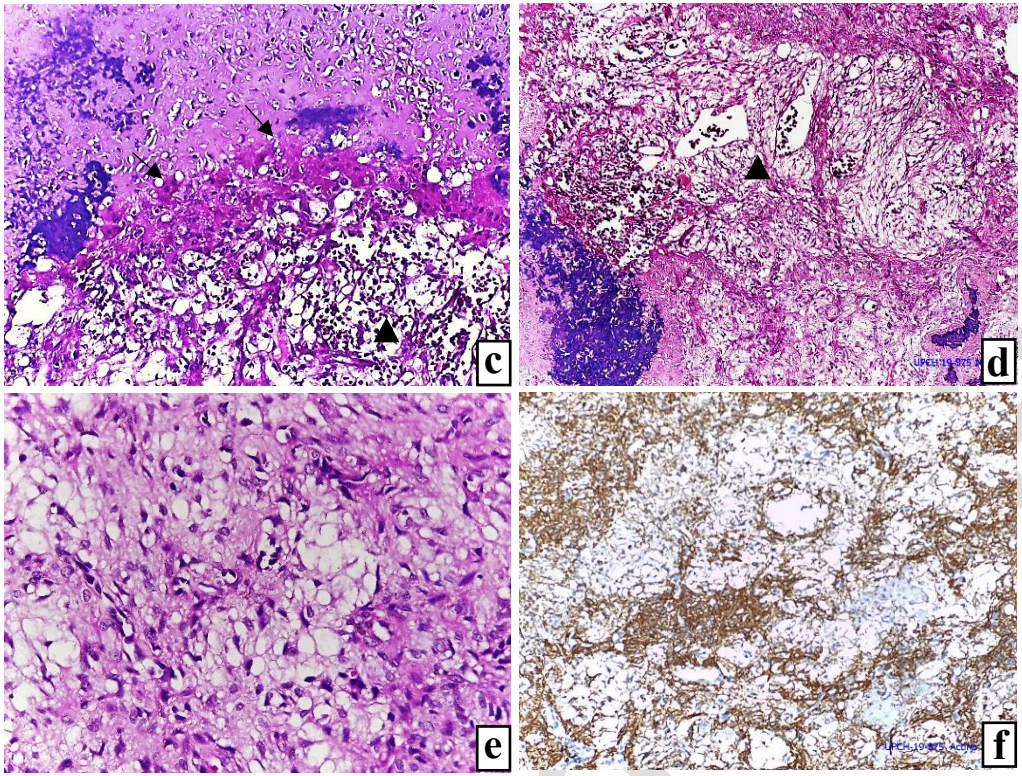
Fig. 1. (a-d).

Fig. 1. (a) Panoramic radiograph showing a well-defined, corticated, oval-shaped lesion in the region of tooth 46. (b, c) CBCT images reveal a hypodense circular area surrounding a centrally located irregular hyperdense mass, associated with thinning and mild expansion of the buccal and lingual basal cortical plates. (d) The mandibular canal is displaced but remains corticated and intact.

Based on the clinical and radiographic findings, a benign intraosseous lesion was suspected. The differential diagnosis included cemento-ossifying fibroma, benign odontogenic tumors exhibiting mixed radiolucent-radiopaque features, and osteoid osteoma.

Histopathological examination of the incisional biopsy specimen revealed a benign mesenchymal neoplasm composed of lobules containing chondroid and myxoid components, with multinucleated giant cells predominantly located at the interface between them. Distinctive calcified deposits of variable morphology, exhibiting a red-purple coloration, were identified within the chondroid component (Fig. 2A).

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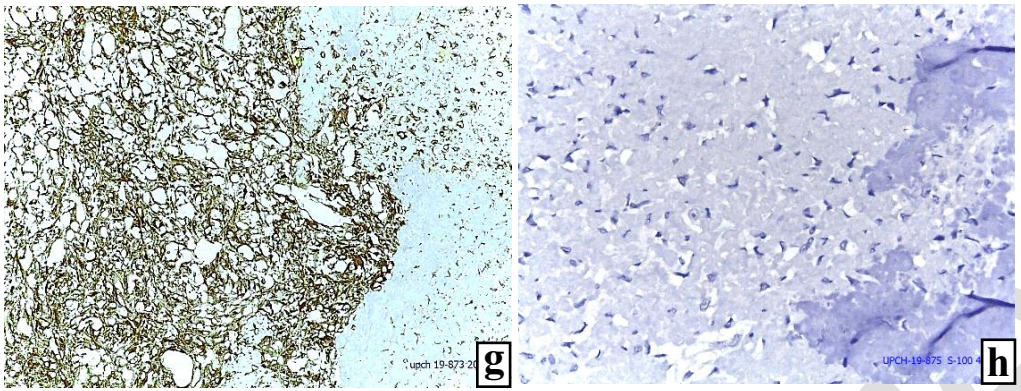

Fig. 2. (a-h).

Fig. 2. (a) Histopathological image showing chondroid areas with dark-purple calcifications (★), myxoid areas (▲), and myxoid regions bordered by multinucleated giant cells (●). (b) Chondroid areas containing irregular dark-purple calcifications (★), with multinucleated giant cells located at their periphery (●). (c) Higher-magnification view of chondroid areas containing stellate cells, bordered by multinucleated giant cells (➞), and adjacent myxoid matrix with a mononuclear inflammatory infiltrate (▲). (d) Well-defined myxoid area located between two chondroid matrices (▲). (e) Myxoid area composed of eosinophilic spindle-shaped cells embedded within an abundant clear to slightly eosinophilic extracellular matrix. (f) SMA immunohistochemical staining demonstrating strong positivity in the myxoid areas. (g) Vimentin immunostaining showing positivity in both myxoid and chondroid components. (h) S-100 immunostaining highlighting stellate cells within the chondroid areas.

The chondroid component consisted of stellate cells embedded within a basophilic matrix. At the periphery of the lobules, a rim of osteoclast-like multinucleated giant cells was observed (Fig. 2B, C). The myxoid component was situated between the chondroid lobules and was composed of eosinophilic spindle-shaped cells set within an abundant myxoid extracellular matrix, which appeared clear to slightly eosinophilic on hematoxylin and eosin staining (Fig. 2D, E).

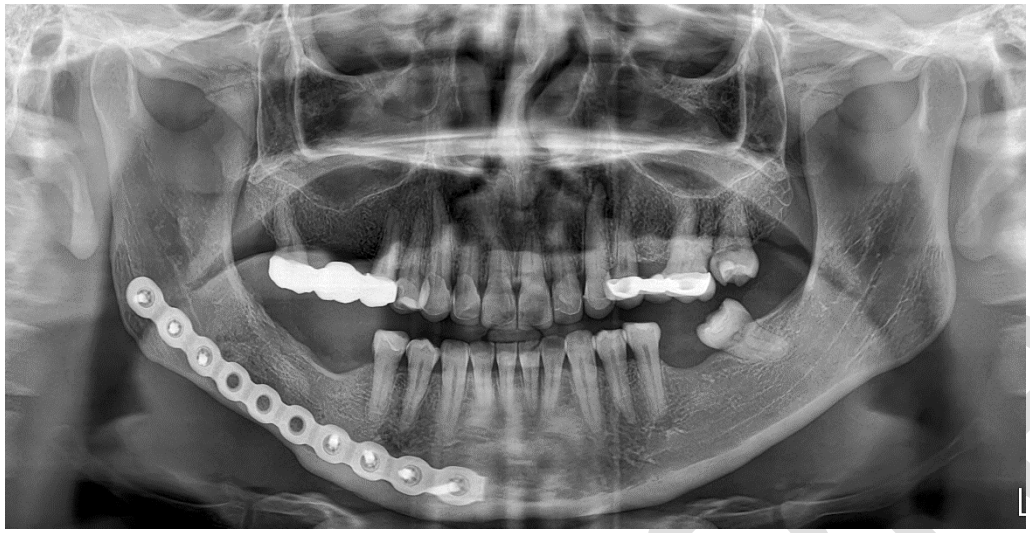

Fig. 3. Panoramic radiograph taken six years after surgical treatment. Complete bone regeneration of the affected area is observed.

Immunohistochemical analysis demonstrated strong cytoplasmic positivity for smooth muscle actin (SMA) (Fig. 2F) and vimentin (Fig. 2G) within the myxoid component, whereas the chondroid component was negative for both markers. S-100 protein expression was absent in both the chondroid and myxoid components; however, the stain enhanced the visualization of cellular morphology within the chondroid areas (Fig. 2H). No immunoreactivity for CD10 or p53 was detected, and Ki-67 labeling was absent.

Based on the clinical, radiographic, and microscopic examination of H&E-stained sections, a diagnosis of chondromyxoid fibroma was established. Immunohistochemical analysis contributed to a better characterization of the histological components of the lesion, while the absence of p53 and Ki-67 expression confirmed its benign nature. Surgical treatment consisted of complete conservative excision of the lesion together with extraction of tooth 47. To preserve mandibular continuity and promote bone regeneration, reconstruction was performed using an autogenous iliac crest bone graft combined with platelet-rich fibrin and stabilized with a reconstruction plate.

At the six-month follow-up, no postoperative complications were observed, and bone healing was satisfactory. The only sequela reported by the patient was mild paresthesia in the right mental region, likely related to the reconstruction plate. Panoramic radiography and cone-beam computed tomography (CBCT) performed six years after surgery demonstrated complete regeneration of the affected mandibular bone, with no evidence of recurrence or residual symptoms (Fig. 3).

3. Discussion

Chondromyxoid fibroma (CMF) of the jaws is an exceptionally rare benign chondrogenic neoplasm. Our review of publications from 2010 to 2025 identified 19 well-documented cases involving the jaws (Table 1), confirming the rarity of this tumor at this anatomical site.[3–21] The mandible was the most commonly affected location (12/19 cases), particularly the posterior body, angle, and ramus.[2,8,17] Reported patients ranged from 1 to 88 years of age, with no significant sex predilection (Table 1).

Table 1. Cases of Chondromyxoid Fibroma of the Jaws (2011–2025)

N°	Author(s)	Year	Age	Sex	Location	Sub-location	Imaging Characteristics	Treatment	Recurrence
1	Castle & Kernig	2011	15	M	Mandible	Mandibular angle	Multilocular radiolucency, soap-bubble appearance	Wide curettage	No
2	Baranović et al.	2012	13	F	Mandible	Posterior body	Well-delimited expansive osteolytic lesion	Curettage	No
3	Strek et al.	2013	27	F	Mandible	Mandibular ramus	CT: lobulated hypodense mass with cortical thinning	Segmental resection	No
4	Fomete et al.	2014	30	F	Mandible	Right mandibular body	Expansive radiolucent lesion 9 cm, no calcifications	Mandibular disarticulation	No
5	Pintor et al.	2015	65	M	Maxilla	Zygoma (zygomaticomaxillary region)	CT: expansive lytic lesion with adjacent bony erosion	En-bloc resection	No
6	Cardemil et al.	2016	15	F	Maxilla	Left posterior maxilla	CT: well-defined hypodense lesion, cortical expansion, no calcifications	Total left maxillectomy	No (6 months)
7	Khatib & Pogrel	2018	43	M	Mandible	Right body (premolar–molar)	CBCT: irregular unilocular radiolucency with buccal cortical resorption	Surgical enucleation	No
8	Florescu et al.	2019	21	F	Mandible	Right posterior body	Panoramic and CT: expansive multilocular radiolucent lesion	Marginal resection	No
9	Terzic et al.	2020	10	M	Mandible	Mandibular body and angle	CT: expansive osteolytic mass with cortical breakthrough	Segmental resection + reconstruction with 7th rib flap (Eve procedure)	Yes (after prior curettage)
10	Hassan & Ghali	2020	35	F	Maxilla	Anterior maxilla	CT: well-delimited hypodense lesion with nasal floor erosion	Wide curettage	No

11	Elseyoufi et al.	2021	43	M	Mandible	Right body (teeth 44–46)	CBCT 3D: well-defined irregular unilocular radiolucency, buccal cortical resorption and apex of tooth 45; IHC: S-100 positive in chondroid area	Enucleation and local curettage	No
12	Panucci et al.	2021	1	M	Maxilla	Left anterior maxilla	CT: well-defined hypodense mass compressing ipsilateral nasal cavity and maxillary sinus; MRI: well-delimited T2 hyperintense lesion; IHC: vimentin+, CD10+, focal α-SMA+, S-100–, Ki-67 6%	Conservative surgical excision	No (1 year)
13	Elzouiti et al.	2022	88	M	Maxilla	Right zygoma (zygomatic arch)	Axial CT: osteolytic lesion of zygomatic bone with cortical destruction; MRI: T1 hypointense, T2 hyperintense, heterogeneous enhancement	Intraoral bone curettage under general anesthesia	No (5 months)
14	Jadaun et al.	2023	13	M	Mandible	Parasymphysis–angle–right condyle	OPG: unilocular lesion 15×12 cm, right premolar to subcondylar region; CECT: thin-walled cystic lesion 15–19 HU, bony thinning, neurovascular bundle displacement	Hemimandibulectomy + 2.4 mm reconstruction plate	No (2 years)
15	Moreno-García et al.	2023	17	M	Mandible	Right ascending ramus	CT: lobulated hypodense mass, well-defined borders, no calcifications	Segmental resection + reconstruction	No
16	Watabe et al.	2024	1	M	Maxilla	Left anterior maxilla	CT: mass displacing and compressing ipsilateral nasal cavity	Surgical excision	No
17	Li & Gong	2024	56	F	Mandible	Right TMJ – condyle/pterygoid space	CT: vermicular osteolysis of right condyle, ground-glass opacity, middle cranial fossa thinning; MRI: irregular mass T1 hypointense, T2 heterogeneous, peripheral ring enhancement; IHC: S-100+, Ki-67 ≤1%	Preauricular resection + CAD/CAM titanium mesh + temporalis myofascial flap	No (1 year)
18	Shahriar Rubel et al.	2025	24	F	Maxilla	Right upper maxilla (premolar–molar, hard palate)	Contrast CT: expansive lucent lesion 4×3.5 cm, inhomogeneous dense content, bony border thinning with dehiscence, marginal enhancement, indenting right maxillary antrum; OPG: no significant findings	Right class III maxillectomy under general anesthesia	Yes (prior surgery 2019, same region)
19	Lodhia et al.	2025	38	M	Mandible	Angle and mandibular ramus	CT: expansive multilocular lytic lesion, cortical thinning	Segmental resection	No

n = 19 gnathic cases.

CT = computed tomography; CBCT = cone beam CT; MRI = magnetic resonance imaging; OPG = orthopantomogram; CECT = contrast-enhanced CT; IHC = immunohistochemistry; TMJ = temporomandibular joint.

The present case involved a 29-year-old woman with a slowly enlarging mandibular lesion. Radiographically, the tumor appeared as a well-defined mixed-density lesion with cortical thinning and mild expansion, features suggestive of a benign fibro-osseous process.[24] No radiographic signs of malignancy, such as ill-defined borders, cortical destruction, or periosteal reaction, were observed. In some cases, CMF may exhibit cortical erosion and bone destruction, thereby mimicking chondrosarcoma or osteosarcoma and posing diagnostic challenges, particularly when only limited biopsy specimens are available.[13,22,28]

The final diagnosis in the present case was established primarily on the basis of conventional histopathological findings, whereas immunohistochemistry provided supportive ancillary information. The strong expression of SMA and vimentin within the myxoid component suggests myofibroblastic differentiation of the tumor cells, while the absence of p53 expression and Ki-67 labeling is consistent with the low proliferative activity typically observed in CMF. The multinucleated giant cells identified at the interface between the chondroid and myxoid components exhibited an osteoclast-like morphology and may contribute to matrix remodeling during tumor growth. In addition, the distinctive calcified deposits observed within the chondroid matrix represent a recognized histopathological feature of CMF and may also be encountered in other benign and malignant cartilaginous neoplasms.

An additional finding of surgical relevance was the absence of a true capsule separating the lesion from the surrounding bone. Instead, a thin layer of vascularized connective tissue was present between the tumor and adjacent normal bone. Similar observations have been reported previously and support complete surgical excision with adequate osseous margins.[5,7]

Treatment ranges from curettage to marginal or segmental resection, depending on lesion size, cortical involvement, and recurrence risk.[12,16,20,21] Because recurrence is primarily associated with incomplete excision,[12,20] conservative marginal resection was performed. At six years of follow-up, complete bone regeneration and absence of recurrence supported the effectiveness of this approach for anatomically favorable mandibular CMF.[12,16,21]

4. Conclusion

Chondromyxoid fibroma of the jaws is a rare benign chondrogenic neoplasm that may mimic fibro-osseous lesions, mixed odontogenic tumors, or other benign osteogenic tumors. Some cases may be mistaken for low-grade chondrosarcoma. In the present case, the clinical presentation and imaging findings were consistent with a benign lesion. The definitive diagnosis was established through histopathological examination of H&E-stained sections. Immunohistochemistry was not essential for the diagnosis. This case highlights the importance of clinicopathological correlation and demonstrates that complete conservative surgical excision can result in excellent long-term outcomes without recurrence.

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AUTHORS' CONTRIBUTIONS

WADA: Investigation, Methodology, Supervision, Validation, Visualization, Writing – original draft and Writing – review & editing.
JOHP: Resources and Validation.
HDGM: Methodology and Validation.
HAO: Methodology and Resources.
MGCP: Resources and Visualization.
NLGP: Resources, Supervision and Validation.
KBTS: Conceptualization, Investigation, Methodology, Supervision, Validation, Visualization, Writing – original draft and Writing – review & editing.

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ETHICS STATEMENT

The authors declare that there are no ethical issues to disclose.

CONSENT

Informed consent was obtained directly from the patient

CONFLICT OF INTEREST

The authors declare no conflicts of interest.

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