World Journal of Advanced Research and Reviews
eISSN: 2581-9615 CODEN (USA): WJARAI
Cross Ref DOI: 10.30574/wjarr
Journal homepage: https://wjarr.com/

(CASE REPORT)

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Periosteal tibial osteosarcoma with pulmonary dissemination and exceptional cardiac metastasis: Case Report and and Imaging Review"

K. Lahlali *, H. Ouazzani, I. Chaouche, A. Akammar, N. El Bouardi, M.Haloua, B. Alami, M.Y. Alaoui Lamrani, M. Maaroufi and M. Boubbou.

Department of radiology, CHU Hassan II, FEZ, Sidi Mohammed Ben Abdellah University, Morocco.

World Journal of Advanced Research and Reviews, 2026, 30(01), 1966-1974

Publication history: Received on 09 March 2026; revised on 19 April 2026; accepted on 22 April 2026

Article DOI: https://doi.org/10.30574/wjarr.2026.30.1.1010

Abstract

Periosteal osteosarcoma is a rare malignant bone tumor belonging to the group of surface osteosarcomas. It differs from conventional forms by its periosteal origin, intermediate grade, and distinct biological behavior.

We report the case of a 14-year-old female patient with no significant past medical history, who presented with progressive leg pain evolving over one month. The pain was inflammatory in nature, associated with unquantified weight loss and limping.

Initial evaluation included plain radiography, followed by computed tomography (CT) and magnetic resonance imaging (MRI) of the left leg, which revealed an aggressive metaphyso-diaphyseal tumor of the left tibia, highly suggestive of periosteal osteosarcoma.

A surgical biopsy was subsequently performed, confirming the diagnosis on histopathological examination.

The patient was managed with neoadjuvant chemotherapy . However, the clinical course was marked by tumor progression with locoregional extension, leading to the indication for transfemoral amputation.

This procedure was refused by the patient's family. The subsequent course was unfavorable, with the development of pulmonary metastases as well as a rare secondary cardiac involvement.

Keywords: Periosteal osteosarcoma**;** Surface osteosarcomas; Biopsy; MRI

1. Introduction

Surface osteosarcomas are rare tumors, accounting for approximately 7% of all osteosarcomas [1]. According to the 2013 World Health Organization (WHO) classification, they are subdivided into three entities in decreasing order of frequency: parosteal osteosarcoma, periosteal osteosarcoma, and high-grade surface osteosarcoma.

Periosteal osteosarcoma is an intermediate-grade malignant bone tumor arising from the surface of the bone, most commonly originating from the periosteum [2]. It accounts for approximately 20% of surface osteosarcomas [3].

This tumor predominantly affects young individuals, typically during the second and third decades of life, with a slight male predominance.

* Corresponding author: K. Lahlali

According to the 5th edition of the WHO classification of soft tissue and bone tumors, the diagnosis is based on a combination of characteristic histopathological and radiological features, including a juxtacortical, subperiosteal origin and a typical diaphyseal location in long bones [2].

Management primarily relies on wide surgical excision, while the role of adjuvant chemotherapy remains a matter of debate.

2. Case presentation

We report the case of a 14-year-old female patient with no significant past medical history, who presented with progressive left leg pain evolving over one month. The pain was inflammatory in nature and associated with unintentional weight loss and limping.

On clinical examination, the patient was conscious, hemodynamically and respiratory stable, and afebrile. Physical examination revealed tenderness over the posterior aspect of the left leg, exacerbated by knee movement, along with localized swelling.

Initial laboratory investigations showed a hemoglobin level of 12 g/dL, white blood cell count of 8,190/mm^3 (neutrophils: 6,010/mm^3, lymphocytes: 1,770/mm^3), and a platelet count of 326,000/mm^3.

Initial radiological assessment revealed an aggressive metaphyso-diaphyseal lesion of the left tibia, highly suggestive of periosteal osteosarcoma.

Plain radiographs of the left leg (anteroposterior and lateral views) demonstrated a heterogeneous bone pattern involving the proximal third of the tibial diaphysis, associated with a Codman triangle periosteal reaction and cortical thickening **(**Figure 1).

Computed tomography (CT) of the left leg, with axial, coronal, and sagittal reconstructions, revealed a large soft tissue mass arising from the surface of the proximal metaphyso-diaphyseal region of the left tibia. The lesion appeared heterogeneous following contrast administration, with extensive areas of necrosis and internal septations, resulting in an irregular cortical bone appearance consistent with moth-eaten osteolysis (Lodwick type III). The mass extended into the anterior and posterior muscle compartments. Additionally, heterogeneous involvement of the cancellous bone was noted, along with a periosteal reaction characterized by a Codman triangle medially and a “sunburst” pattern laterally (Figure 2).

Magnetic resonance imaging (MRI) of the left leg demonstrated a locally advanced, ill-defined lesion involving the metaphyso-diaphyseal region of the left tibia. The lesion showed isointense signal on T1-weighted images, heterogeneous hyperintensity on T2-weighted and STIR sequences, and marked heterogeneous enhancement after gadolinium administration. It measured approximately 100 × 145 mm (transverse × craniocaudal dimensions). The lesion was associated with cortical destruction and T2 hyperintensity within the medullary cavity extending to the junction of the middle and distal thirds of the tibia (Figure 3).

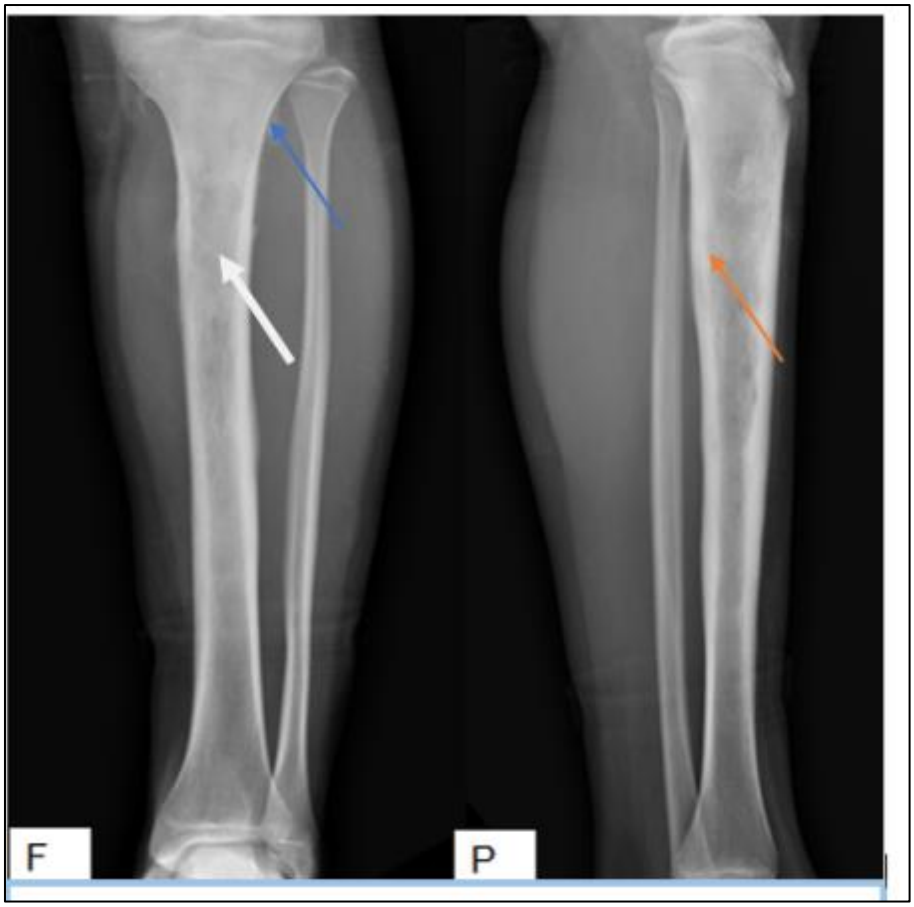
A surgical biopsy was performed, confirming the diagnosis on histopathological examination.

The patient was managed according to the OS 2005 protocol for non-metastatic osteosarcoma, consisting of neoadjuvant chemotherapy followed by a planned amputation.

However, the clinical course was marked by tumor progression with locoregional extension. Follow-up MRI demonstrated an increase in tumor size at the metaphyso-diaphyseal region, with extension toward the growth plate and no evidence of skip metastases (Figure 4).

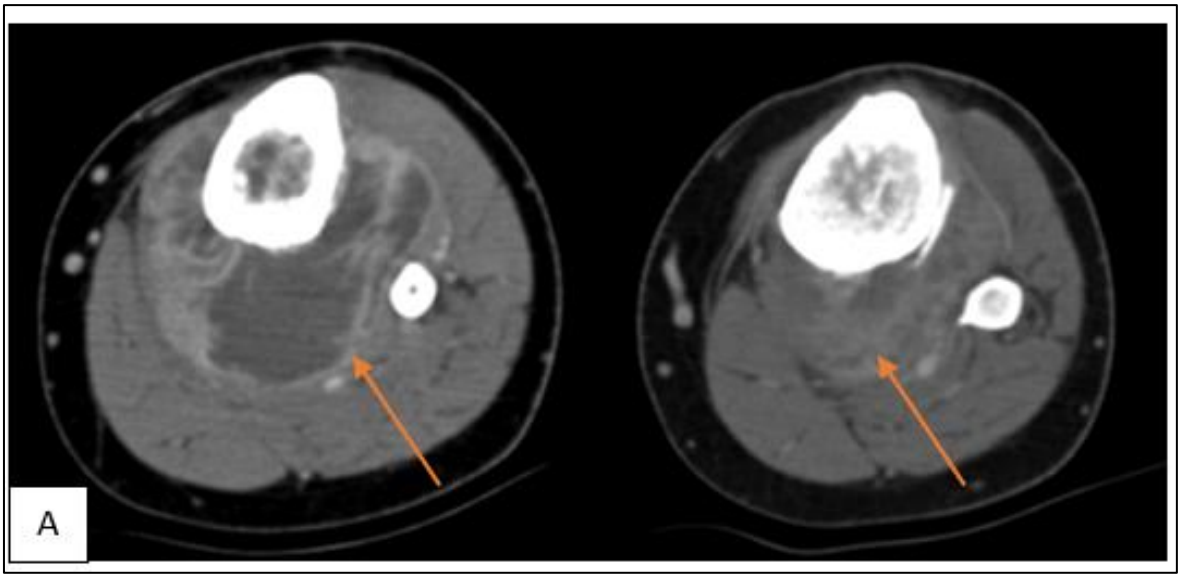
Amputation was indicated; however, the procedure was refused by the patient’s family.

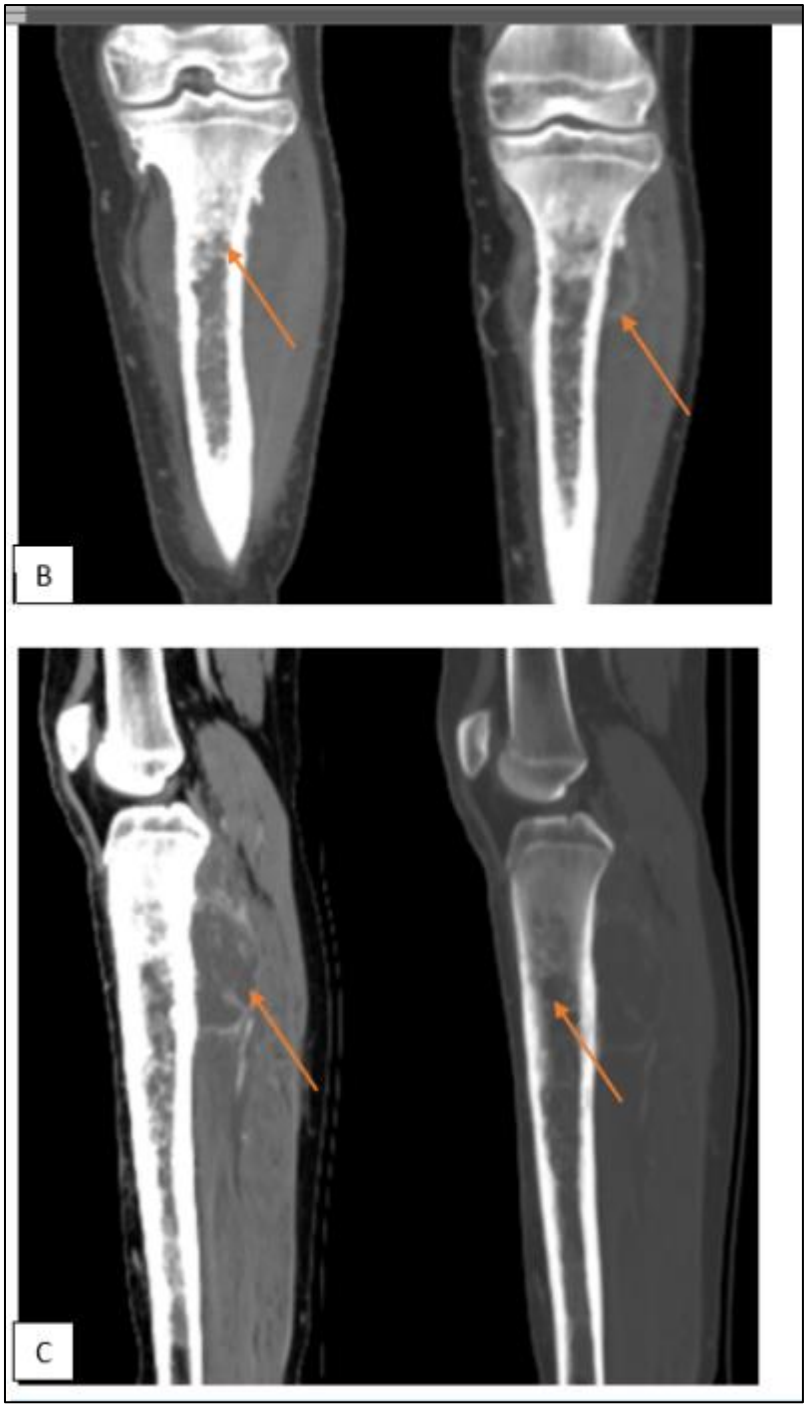
Subsequent thoraco-abdomino-pelvic CT scans revealed an unfavorable outcome, with the development of pulmonary metastases as well as a secondary cardiac involvement (Figure 5).



Reference: Radiology department CHU HASSAN II, FEZ, Morocco

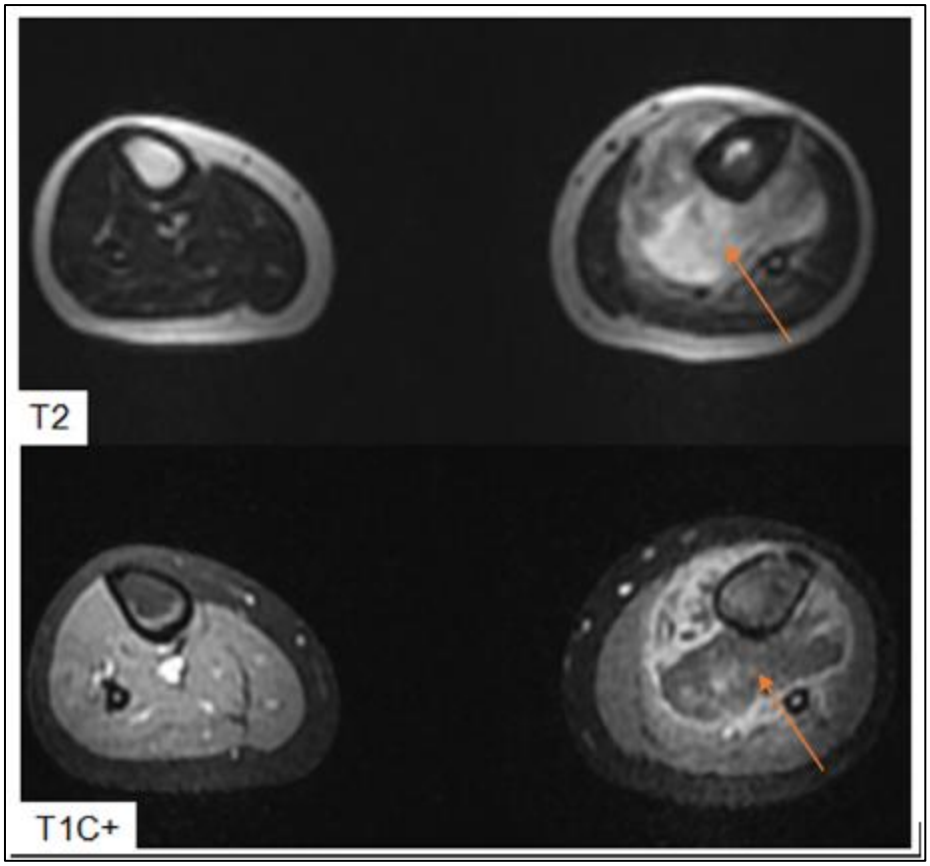
Figure 1 Radiographs of the left leg (anteroposterior and lateral views) : Heterogeneous bone pattern involving the proximal third of the tibial diaphysis (Black arrow), associated with a Codman triangle periosteal reaction (Blue arrow) and cortical thickening (Orange arrow)





Reference: Radiology department CHU HASSAN II, FEZ, Morocco

Figure 2 Computed tomography (CT) of the left leg with axial (A), coronal(B), and sagittal (C) reconstructions, revealed a large soft tissue mass arising from the surface of the proximal metaphyso-diaphyseal region of the left tibia. The lesion appeared heterogeneous following contrast administration, with extensive areas of necrosis and internal septations, resulting in an irregular cortical bone appearance consistent with moth-eaten osteolysis (Lodwick type III). The mass extended into the anterior and posterior muscle compartments. Additionally, heterogeneous involvement of the cancellous bone was noted, along with a periosteal reaction characterized by a Codman triangle medially and a "sunburst" pattern laterally



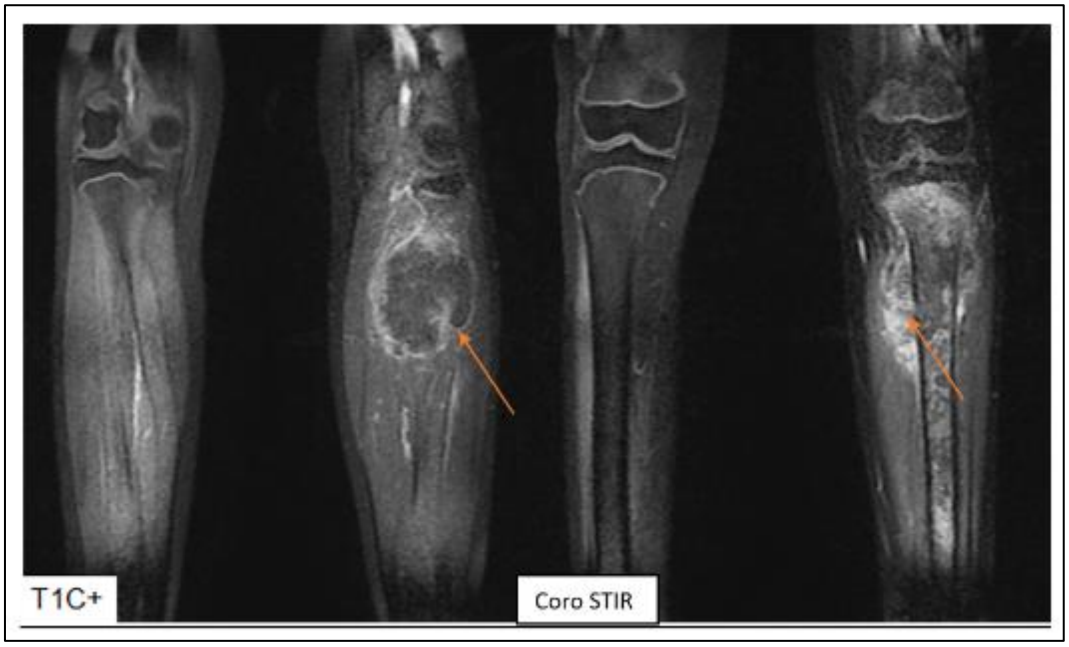


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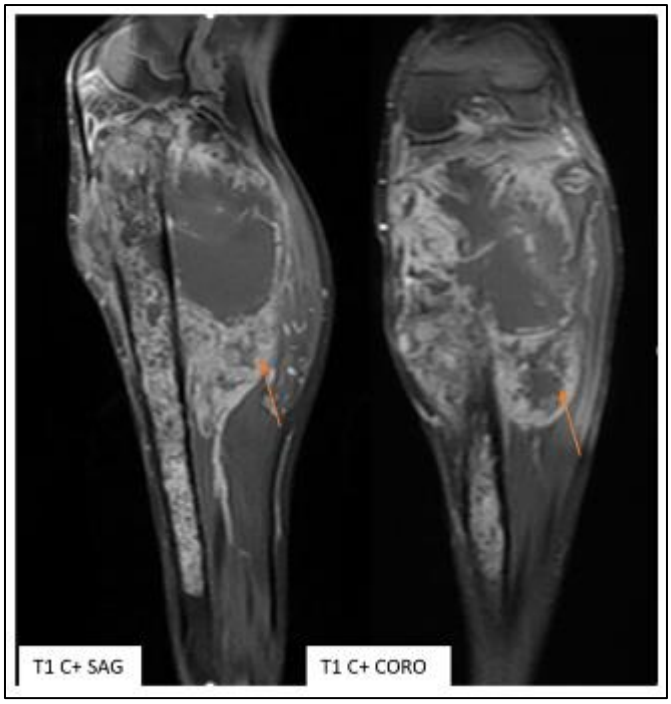
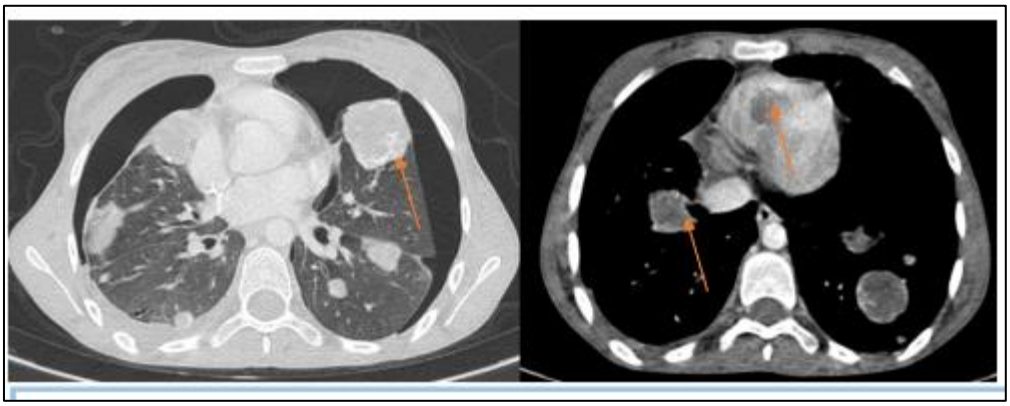
Figure 3 Magnetic resonance imaging (MRI) of the left leg demonstrated a locally advanced, ill-defined lesion involving the metaphyso-diaphyseal region of the left tibia. The lesion showed isointense signal on T1-weighted images, heterogeneous hyperintensity on T2-weighted and STIR sequences, and marked heterogeneous enhancement after gadolinium administration. It measured approximately 100 × 145 mm (transverse × craniocaudal dimensions). The lesion was associated with cortical destruction and T2 hyperintensity within the medullary cavity extending to the junction of the middle and distal thirds of the tibia



Reference: Radiology department CHU HASSAN II, FEZ, Morocco

Figure 4 Follow-up MRI demonstrated an increase in tumor size at the metaphyso-diaphyseal region, with extension toward the growth plate and no evidence of skip metastases



Reference: Radiology department CHU HASSAN II, FEZ, Morocco

Figure 5 Thoraco-abdomino-pelvic CT scans revealed pulmonary metastases and secondary cardiac involvement

3. Discussion

Periosteal osteosarcomas are intermediate-grade osteosarcomas with a predominantly chondroblastic component, with a variable prognosis and long-term survival rates ranging from 55% to 80% [3].

It is the second most common type of surface osteosarcoma after parosteal osteosarcoma, accounting for approximately 1–2% of all osteosarcomas [4,5].

It can occur over a wide age range, from the first to the seventh decade, with a peak incidence during the second decade of life [4].

Periosteal osteosarcomas typically arise from the inner cambium layer of the periosteum and are attached to the underlying cortex. Their cytological grade is intermediate, higher than that of parosteal osteosarcoma but lower than that of conventional osteosarcoma [4].

Classically, these tumors are located in the diaphysis of long bones, most commonly involving the tibia (40%) and femur (38%), particularly along the anteromedial aspect of the tibial diaphysis [6]. Other less frequent sites include the humerus and ulna (5–10%).

Clinically, patients usually present with nonspecific symptoms such as localized swelling and/or pain evolving over several weeks to months.

The diagnosis is based on the combination of imaging findings and histopathological analysis obtained through surgical or image-guided biopsy.

Histopathologically, periosteal osteosarcoma is characterized by a predominant cartilaginous component with transition to osteoid matrix formation. It typically shows lobules of atypical cartilage, intervening bands of reactive bone, and a periosteal reaction with linear reactive bony spicules separated by cartilaginous matrix. The tumor also contains primitive mesenchymal cells with mitotic activity and nuclear atypia [4–7].

Plain radiography of the limb demonstrates a fusiform or broad-based mass arising from the bone surface, associated with cortical thickening. The periosteal reaction may be solid, lamellar, or spiculated, and may occasionally be associated with a Codman triangle. The lesion typically involves approximately 50% of the cortical circumference. Intramedullary extension is uncommon. The overall appearance is usually heterogeneous [5-6–8].

Computed tomography (CT): demonstrates a soft tissue mass with a broad-based attachment to the cortical surface, observed in nearly all cases. A mineralized periosteal reaction oriented perpendicular to the cortex is present in approximately 95% of cases, as reported by Murphey et al. [6]. A Codman triangle, reflecting the aggressive nature of the lesion, may also be observed.

The underlying cortex may appear either thinned or thickened as a result of the periosteal reaction, related to the osteoblastic activity of the inner periosteal layer. Cortical disruption is uncommon.

Matrix calcifications may be present, reflecting the chondroblastic histological nature of this tumor [6].

Magnetic resonance imaging (MRI): reveals a heterogeneous lesion on T2-weighted sequences, with areas of marked hyperintensity corresponding to the cartilaginous matrix component [6]. The lesion is typically well delineated by the superficial layer of the periosteum.

Following contrast administration, heterogeneous enhancement is observed due to the presence of mineralized components. Septal and lobulated enhancement patterns may also be seen, reflecting the underlying chondroblastic matrix.

Medullary involvement is uncommon and, when present, does not appear to significantly impact prognosis. However, reactive (non-tumoral) bone marrow signal abnormalities are frequently observed adjacent to the lesion. In such cases, the cortical bone remains intact, appearing as a continuous low-signal line on all sequences [2].

The differential diagnosis includes aggressive-appearing juxtacortical lesions of periosteal origin, as well as other surface osteosarcomas in their atypical forms.

Periosteal chondrosarcoma [9] typically affects older patients, usually in the fourth to fifth decades of life. It is commonly located in the metaphysis and is often not associated with a perpendicular periosteal reaction.

Periosteal Ewing sarcoma is an extremely rare tumor. It may mimic periosteal osteosarcoma due to similar patient demographics and anatomical location.

The prognosis of intermediate-grade periosteal osteosarcoma is generally more favorable than that of conventional osteosarcoma, but less favorable than that of parosteal osteosarcoma. Reported 5- and 10-year survival rates are approximately 89% and 77–86%, respectively [2]

Medullary involvement is uncommon; however, when present, it is associated with a poorer prognosis.

The mainstay of treatment is wide surgical excision. The role of chemotherapy remains controversial, as it does not appear to significantly improve prognosis or overall survival. In rare cases, it has been associated with the development of secondary malignancies, particularly pulmonary [2].

Local recurrence, metastatic spread, and death typically occur within the first three years following diagnosis [2].

4. Conclusion

Accurate diagnosis relies on a rigorous correlation between imaging findings and histopathological examination. In the presence of an aggressive juxtacortical lesion with a diaphyseal location, associated with a malignant periosteal reaction in a young patient, periosteal osteosarcoma should be strongly suspected.

List of abreviations:

- **CT** : Computed tomography
- **MRI** : Magnetic resonance imaging
- **WHO** : World Health Organization

Compliance with ethical standards

Disclosure of conflict of interest

The authors do not declare any conflict of interest

Statement of informed consent

Written informed consent was obtained from the patient. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Availability of data and materials

The data sets are generated on the data system of the CHU Hassan II of Fes, including the biological and radiological data.

Author's contribution

- KL Is the corresponding author, she participated in the organization and writing of the article.
- Professor MB supervised the working and validated the figures.
- Professor and chief of department of radiology MB and MM red and allowed the article for publication.
- All authors read and approved the final manuscript.

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