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The Sign of the Rising Sun: An Unsequent Periostic Reaction

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Summary

The sign of the "rising sun" has been described as a periosteal reaction that tends to appear in pathologies of a benign or malignant nature, but characterized by being an infrequent reaction, mostly in those pathologies described as malignant, such as In the case of osteosarcoma, which is a type of primary malignant tumor prevalent in children under 25 years of age, it is more frequently located in the femur, tibia and fibula, presenting a destructive bone lesion that has an aggressive periosteal reaction to the pattern of sun rays, when Being one of the most important radiological signs, the use of radiological diagnosis is of vital importance for its correct evaluation accompanied by histopathological studies that allow addressing the suspicion of a bone tumor.

Keywords: Rising sun, Osteosarcoma, Chondrosarcoma, Ewing's sarcoma

Introduction

The periosteum is a thin membrane that covers most of the bony structures, except for the intra-articular surfaces that are covered by cartilage and sesamoid bones. It plays an important role in growth, bone repair and in the irrigation of bone tissue [1]. When it is compromised, it responds by modifying the morphology of the bone. This reaction is known as periosteal reaction, or periostitis, these can be categorized according to their pattern (continuous and interrupted) and their identification helps to define the differential diagnosis, since these reactions are an inaccurate radiographic finding that indicates periosteal irritation. (Table 1). In turn, there are different models within each continuous and interrupted reaction (Figure 1). They can also be broadly characterized as benign or aggressive [2]. Generally, the malignant ones have an accelerated growth and produce laminated, amorphous and irregular reactions. Among them, the "rising sun" sign is striking, characterized by septa arranged perpendicular to the bone cortex that can extend to the epiphysis and diaphysis, due to its infrequency and, at the same time, because it is a tuberculation pattern. For this reason, by growing rapidly and firmly, the periosteum will not be able to regenerate the tissue, causing the formation of tiny fibroelastic that connect the periosteum to the bone and when these ossify, the periosteal reaction occurs in the kicking of the "rising sun". Therefore, its usefulness is important for the identification of the etiology of the lesion, as it reflects acute periostitis, which can be benign such as osteomyelitis, meningioma, hemangiomas, subperiosteal hematomas and less frequently odontogenic or malignant myxomas as in osteosarcoma, sarcoma of Ewing and osteoblastic metastases (Figure 2) [3].

Table 1: Periosteal reaction patterns

Continuous periosteal reaction	Lesions with presence of the cortex	Reaction of a layer	Benign lesions. Osteomyelitis. Eosinophilic granuloma
		Multilayer reaction	Malignant and benign lesions
	Lesions with presence of the cortex	Soft shell	Giant cell tumor
Interrupted periosteal reaction	Interrupted periosteal reaction		Osteomyelitis. Eosinophilic granuloma. Fractures Ewing's sarcoma. Osteosarcoma.

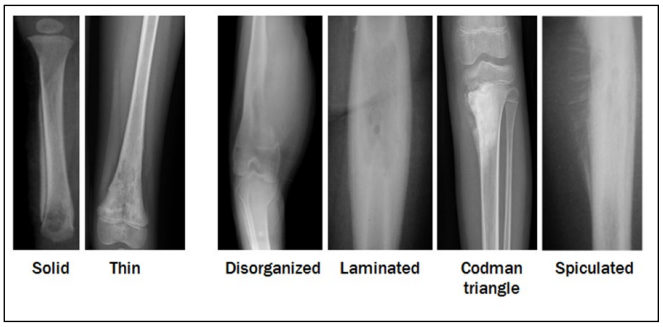

Figure 1: Periosteal reactions

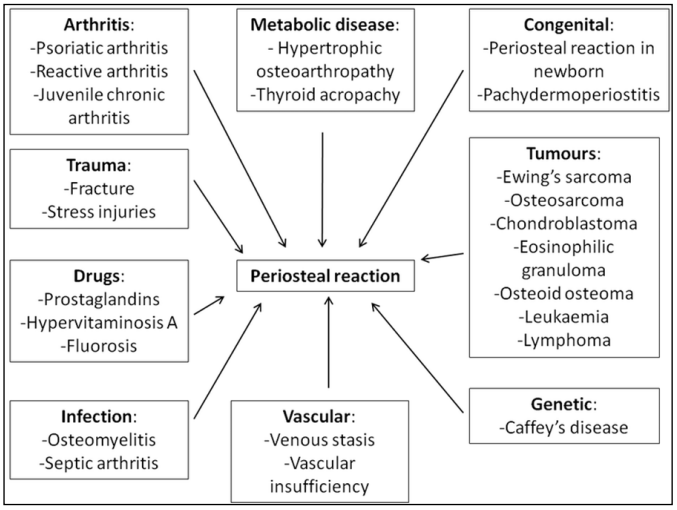

Figure 2: Pathologies where a "rising sun" sign is present

The bone lamellae characteristic of this sign and the presence of mineralized bone matrix are best evaluated with radiography, magnetic resonance imaging, and computed tomography. They are visualized as thin and straight spicules. Cone beam computed tomography probably shows the speculated pattern with greater precision due to its ability to show fine internal structures of a lesion than does a conventional radiograph (Figure 3) [4]. Thus, osteosarcoma, metastasis (especially to the rectum and sigmoid colon), Ewing's sarcoma, hemangioma, and, rarely, fibrous dysplasia are the main differential diagnosis of the periosteal sunburst reaction [5]. Osteosarcoma has its own radiographic characteristics, we can find dense sclerosis of the metaphysis (almost all), extension of soft tissues (75%), radiant calcification; "Sun rays" (60%), osteosclerotic lesion (45%) and lytic lesion (30%). While in chondrosarcoma calcified eccentric osteolytic lesion prevails. Unlike Ewing's sarcoma where there is bone destruction (75%), soft tissue extension (64%), reactive bone formation (25%), laminated periosteal reaction; "Onion skin" (23%), radiant calcification; "Sun rays" (20%) or, failing that, the radiograph may be normal [6].

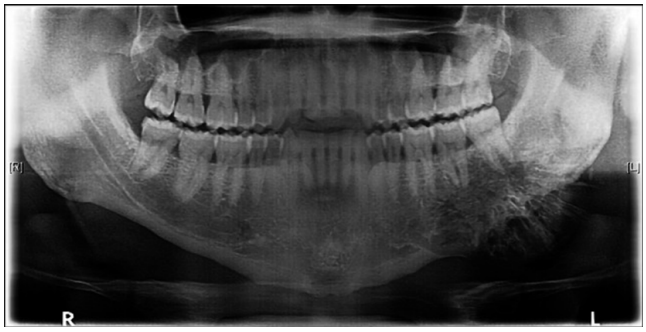

Figure 3: Panoramic X-ray of a 34-year-old male patient, presenting a periosteal reaction in the "rising sun" of the left posterior mandible that encompasses the premolar and molar region, as a consequence of odontogenic myxoma.

Materials and methods

A detailed bibliographic search of information published in the databases pubmed, Elsevier, scielo, national and international libraries is carried out. The following descriptors were used: Rising sun, Ewing's sarcoma, chondrosarcoma, osteosarcoma. The search for articles was carried out in Spanish and English, it was not limited by year of publication.

Results

Osteosarcoma

Osteosarcoma is a primary malignant tumor with a worldwide incidence of 3.4 per million people per year, more frequently in children and adolescents and with a greater distribution in Latin America and Asian populations [7]. Histological evaluation indicates osteoid production in association with malignant mesenchymal cells. According to their histological classification, they can be central, intramedullary and superficial tumors, with several subtypes in each group and according to the characteristics of the tumor and the predominant stromal differentiation (osteoblastic, fibroblastic, chondroblastic, microcytic, superficial and extraskeletal high-grade telangiectatic) [8]. Osteosarcoma is believed to arise from malignant primitive mesenchymal cells that differentiate into osteoblasts, which in turn produce a malignant osteoid matrix. Osteosarcomas can arise in any bone, but classically develop in the metaphyses of the long bones. Almost 60% occur in the distal femur, proximal tibia, and proximal humerus. The metaphysis of a bone contains the growth plate, which is responsible for the active formation and elongation of bone. Therefore, osteosarcomas tend to occur at the age and location where bone growth is most active and when cells are vulnerable to mutations [9]. Eight syndromes in which osteosarcoma occurs more frequently in the pediatric population are currently known: Li-Fraumeni, retinoblastoma, Rothmund-Thomson anemia, RAPADILINO, Werner, Bloom and Diamond-Blackfan. Unlike the molecular abnormalities in pediatric patient osteosarcoma, adult osteosarcoma is distinguished by genetic amplification and hypermuted regions, that is, by chromothripsis and ketaegis, respectively [10,11]. Patients with this condition present pain in the limb due to the involvement of the periosteum before tumor growth, as well as inflammation due to the new formation in soft tissue, also with decreased range of motion and sensitivity in the area, even in some cases detect palpable masses and pathological fractures. In addition to weight loss, fever and in case of metastasis they present adenopathy [12]. The classic imaging findings are the sun ray pattern and represents reactive ossification resulting from the action of normal osteoblasts rather than tumor cells, probably the result of bone morphogenetic protein production by osteosarcoma cells [13]. Osteosarcoma causes a destructive bone exion with a wide transition zone and aggressive periosteal reaction such as the sun ray / laminate pattern with "cloud-shaped" or "spongy" matrix calcifications [14]. As in the case of a 9-year-old male patient with an expansive mass that compromises the roots of a lower left first molar with radiographic characteristics of cementoblastoma, osteo injury and osteosarcoma, the latter due to the pattern of "sun rays" and reabsorption "Pointed" of the roots of the affected teeth [15]. The same pattern occurs in the intercrestal osteosarcoma of a male patient with inflammation of teeth # 26 and # 27 together with knife-edge root resorption and widening of the periodontal ligament [16]. Although osteosarcoma of the jaws is a rare tumor, a case report was also presented of a female patient with bone swelling in the posterior region of the left mandible with a radiographic presentation of sunburst appearance, which together with a histopathological examination was confirmed

the diagnosis of this neoplasm [17]. This shows that one of the most common sites are the bones of the face and jaws, although a predilection is shown for the metaphysis of the long bones, particularly the distal femur, the proximal tibia and the proximal humerus. Areas where aggressive periosteal reactions in the "sunburts" pattern often originate. However, osteosarcoma can present at the craniofacial level with the presence of this pattern of injury in combination with massive bone destruction probably derived from a very aggressive tumor originating in the ethmoidal area of the medial wall of the orbit [18]. In addition, in one study it provided imaging clues for the differentiation of dedifferentiated and low-grade parosteal osteosarcoma. Among the most important criteria is the periosteal reaction in a "rising sun" pattern. The periosteal reaction of solar rays was visualized in two cases of the low-grade type (12.5%) and four cases of the dedifferentiated type (57.1%) ($p = 0.025$) of parosteal OGS [19].

Condrosarcoma

Chondrosarcomas have an incidence of one in every 500,000 inhabitants, with a higher presentation in males and a predominance between the fourth and sixth decades of life. The most frequent presentation sites are the lower limbs, pelvis, ribs, sternum and clavicle. Of its histological varieties, conventional chondrosarcoma constitutes 85% of all cases, the rest are made up of clear cells, undifferentiated, myxoid and mesenchymal types [20]. The radiographic pattern of chondrosarcomas includes single or multiple radiolucent areas. These lytic changes are prominent in more advanced cases. Other findings include pacification of air spaces, densely calcified bone mass, and root resorption. Some authors have reported a uniform widening of the periodontal membrane space. Additionally, it can reveal a frosted glass appearance or a sunburst appearance [21]. As in the case of a 51-year-old patient with Mazabraud's syndrome, where the standard radiograph revealed medullary opacity and heterogeneous erosion of the anterior cortex with periosteal sunburst reaction and the chondroma was identified as grade II in the biopsy at the level of the femur [22]. Likewise, in a woman prior to the diagnosis of mesenchymal chondrosarcoma, the panoramic radiograph revealed a decrease in radiodensity in the left maxillary tuberosity and in the area of the branch. In addition, it had a typical sun-ray appearance in the infratemporal area involving the maxillary tuberosity, infratemporal bone, and the mandibular ramus area. They also resorted to taking a computed radiograph that revealed the entire infratemporal fossa involving the maxillary tuberosity, the infratemporal surface of the temporal bone, and the area of the mandibular ramus. The axial section revealed a non-enhancement mass with mottled calcification measuring 5 × 5 cm in size. Likewise, the classic appearance of sun rays is appreciated [23]. Figure 4 shows the perisotic sun-ray pattern reaction in the fibula of a 22-year-old woman with primary mesenchymal chondrosarcoma [24].

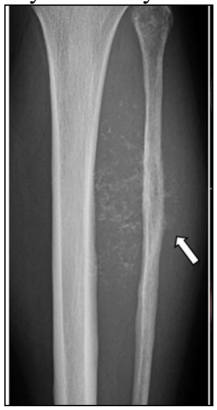

Figure 4: "Sun rays" on the fibula of a 22-year-old woman with primary mesenchymal chondrosarcoma

Ewing sarcoma

Ewing's sarcoma (ES) and Ewing-like sarcomas are highly aggressive round cell mesenchymal neoplasms that occur most frequently in children and young adults [25]. Its exact histogenesis is unknown, but it is believed to be derived from bone marrow cells. However, some believe that it is a round cell cancer of neural origin, similar to the so-called primitive neuroectodermal tumors. Ewing's sarcoma has as a particular characteristic the presence of small blue rounded cells that basically includes: Askin's tumor (bone tumor of the chest wall), skeletal (bone) and extraskeletal (soft tissue) Ewing's sarcoma [26]. On radiography, Ewing sarcoma of bone reveals aggressive features, reflecting the high-grade nature of this malignant lesion. The periosteal reaction is frequent (58% -84%) and generally aggressive in appearance (94%), either laminated (onion skin) or spiculated (sun rays or spiky) [27]. Other reports report that its radiological characteristics are not yet well established. However, in an 11-month-old patient with a confirmed diagnosis of Ewing's Sarcoma, he presented a periosteal reaction with a "sun rays" pattern, so it should be included in the differential diagnosis of some other pathology with this same indicator (Figure 5) [28].

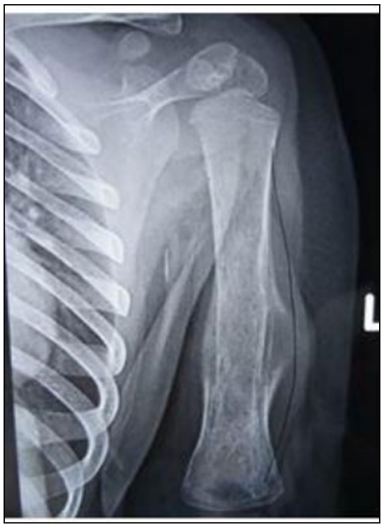

Figure 5: X-ray of the left arm shows a thick periosteal reaction that gives a typical sun-ray appearance with bone expansion, cortical thickening, and endosteal scalloping.

A study by Tsurumoto, Toshiyuki et al. They found a human skeleton of a middle-aged adult man in a Kinoue-Kodo stone coffin from the 5th-6th centuries excavated in the southwestern fringe region of the Oita Plains, northeast of Kyushu, Japan. Where they observed a typical pattern of sun rays on the left scapula. The angle changed regularly from the central area to the marginal region, and its general morphology showed this unusual sign. In addition, on the anterior aspect of the left scapula, mainly at the base of the coracoid, a sunburst-like lesion was also present, although incomplete. Therefore, they suspected that it was a representative primary malignant tumor such as Ewing's sarcoma [29].

Discussion

The periosteal reaction has been described for some years. It occurs when the cortical bone reacts to one of many possible insults. Tumor, infection, trauma, certain medications, and some arthritic conditions can elevate the periosteum of the cortex and form various periosteal reaction patterns. The appearance of the periosteal reaction is determined by the intensity, aggressiveness, and duration of the underlying injury. Furthermore, the periosteum in children is more active and less adherent to the cortex than in adults. Therefore, the periosteal

reaction can occur earlier and appear more aggressive in children than in adults [30]. Consequently, the sign of the "rising sun" has been described by several authors as an infrequent periosteal reaction that tends to arise in pathologies of a benign or malignant nature associated with bone tissue, with a marked presence especially in those bone pathologies of malignant origin. Especially in osteosarcoma, where there is an invasion of bone tissue with subsequent rapid periosteal aggregation that leads to an inflammatory response that causes septae similar to the sun's rays or take the form of spicules that open in the shape of a divergent fan [31, 32]. In osteosarcomas Most injuries occur in patients under 25 years of age, with the femur, tibia and fibula being the most common sites. Sunshine, spiky hair, or Codman's triangle periosteal reaction subtypes are the most commonly seen; however, laminated, solid, thin, or disorganized forms of periosteal reaction may also be present. A large transition zone, cortical rupture, and soft tissue mass are worrisome features that warrant further evaluation [33]. The sign stands out in the classic and telangiectatic osteosarcoma subtypes [34]. For this reason, the radiological diagnosis plays an important role in the evaluation of this type of neoplasms, since we will first find the loss of the normal trabecular bone pattern, accompanied by the mottled appearance that is visualized in the "sunburst" due to the neo bone formation that occurs perpendicular to the axis of the bone, some linear bone spicules are observed; Another characteristic bone change is the Codman Triangle that occurs due to a reaction of the periosteum. In addition, radiographs show osteolytic areas [35]. One study reported that the most important radiological signs were the rising sun sign and the codman triangle when looking for diagnostic concordance between radiological and histopathological findings and found that radiography offers significant values for the diagnosis of osteosarcoma, as it is a method to address the suspicion of a bone tumor, which must be confirmed with histopathological studies [36]. In the case of Ewing's sarcoma, only cortical changes may be evident on radiographs with a permeative or moth-eaten osteolytic component. A large mass of soft tissue can be seen. The periosteal reaction pattern is typically aggressive, with the hairy subtype at the tip. Unlike chondroblastomas where the lesions are typically lytic and may have a sclerotic margin. The periosteal reaction due to chondroblastoma occurs most frequently in large lesions in small or flat tubular bones. The periosteal reaction can be thick, solid, or laminated [37]. However, it can occur in pathologies that do not directly compromise the bone, so it is important to distinguish the periosteal reactions to know the differential diagnoses. This reaction has been associated in 50% with anomalies such as Chiari II, Migration disorders (heterotopias, licencefalia, schizoencephaly), Encephaloceles, Dandy-Walker malformation, Holoprosencephaly and especially, it has been observed in cancer of primary malignant origin especially, Chondrosarcoma, Fibrosarcoma, EWING Tumor and Myeloma [38]. In hydrocephalus, this sign is one of the important clinical criteria in the lactation and neonatal stages, with growth of the cephalic perimeter with delay in the closure of the fontanelles and an increase in their amplitude [39]. In addition, hemangiomas and aggressive osteoblastic metastases can also stimulate this appearance. The periosteal reaction of sun rays in metastasis is rare and is observed in less than 5% of the cases. It is most commonly associated with osteoblastic metastases and approximately 30% are secondary to prostate carcinoma. Less commonly associated primary tumors are bronchi (10%), carcinoid (6%), and breast (4%) [40].

Conclusion

The rising sun sign, defined as the alteration of the periosteum product of an inflammation or pathological lesion that is consequently reflected in radiological images as a speculated and irregular parenchymal mass, which resembles the rays of the sun, constitutes a sign of great importance when performing the differential diagnosis of neoplasms, rooted mainly in long bones, and to a lesser extent in short and flat bones, such as Osteosarcoma, chondrosarcoma and less frequent pathologies such as Ewing's Sarcoma, where studies reveal that the presence of this sign constitutes a determining criterion for when diagnosing this type of neoplastic processes.

This sign mainly determines the development of an underlying bone lesion and its presence is reflected with the change in the morphology of the bone in question, mechanical, physical, chemical and enzymatic changes that alter the microenvironment of the periosteum, however, this is not a sign determinant of malignancy, therefore in the event of a possible diagnosis of a neoplasm with the presence of a rising sun sign in radiological examinations, it is necessary to implement complementary histopathological examinations to determine the etiological origin, whether benign or malignant, of the present neoplasm, which continues to represent an important object of study today.

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