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Original Article

Periosteal chondroma of the proximal tibia: report of three cases

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Abstract: Periosteal chondroma is a relatively rare benign cartilaginous neoplasm usually seen in young adults. We presented three cases of periosteal chondroma in the proximal tibia of a 7-year-old girl (Case 1), a 12-year-old boy (Case 2) and a 15-year-8-month old boy (Case 3). Meticulous analysis of initial and follow-up plain radiographs, Computed Tomography (CT) and Magnetic resonance imaging (MRI) of these painless lesions suggested the diagnosis of periosteal chondroma. This was confirmed by biopsy for Case 1 and Case 2. Local resection with curettage of the adjacent cortex and bone grafting were performed for Case 1 and Case 2. No biopsy or treatment was performed for Case 3. Recovery was uneventful in all three cases. There was no recurrence at the three years and eight months (Case 1), six months (Case 2), and seven years and one month (Case 3) follow-ups.

Keywords: Periosteal chondroma, tibia

Introduction

Periosteal (juxtacortical) chondroma is a slow growing benign cartilaginous tumor, arising within or underneath the periosteum, external to the cortical bone. The lesion was originally described by Lichtenstein and Hall in 1952, who reported six cases [1]. Most lesions are small, ranging from 1-3 cm [2]. Jaffe proposed the term juxtacortical chondroma and reported nine cases in 1956 [3]. Although it can occur at any age, this tumor predominantly occurs in children or young adults, with a male predilection [4] at approximately 2:1 [5]. Periosteal chondromas appear to be quite rare, with only 46 cases reported in a series of more than 7,000 primary bone tumors (0.7%) in a MayoClinic study, and representing less than 2% of all primary bone neoplasms in another survey [6]. Tubular bones are most commonly affected, with humeral and femoral lesions accounting for approximately 70% of cases [2, 4]. The bones in the hands and feet are affected in approximately 25% of cases [2].

In this manuscript we present the clinical and radiographic features of periosteal chondroma in the proximal tibia of a 7-year-old girl, a 12-year-old boy and a 15-year-8-month old boy, with a review of the relevant literature. All patients and their families have given consent to have any information concerning their cases to be published.

Case 1

A 7-year-old healthy girl presented with a painless firm swelling over the anterior aspect of the right proximal tibia which appeared six months ago. There was no history of trauma or systemic symptoms. Physical examination revealed a firm, immobile, non tender and subcutaneous mass, contiguous with the anterior aspect of the right proximal tibia. There was no restriction of knee motion, and neurovascular examination was normal. Laboratory data were within normal limits. Plain radiographs of the right knee revealed a soft tissue swelling with intralesional calcifications adjacent to the right proximal tibia (**Figure 1A**). Observation care was recommended by the treating physician. Three months later, she was re-evaluated for a report of slight enlargement of the mass. Radiographs demonstrated cortical scalloping and remodeling, with

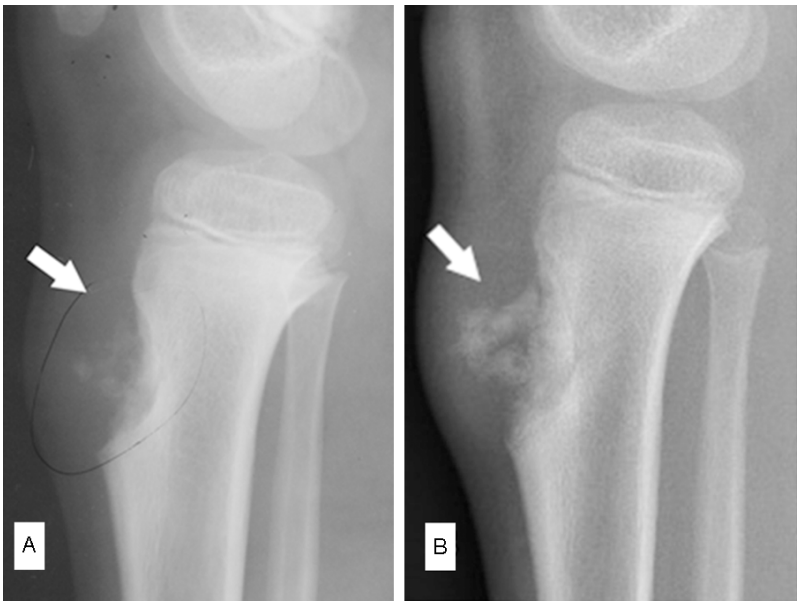

Figure 1. A. Initial lateral radiograph of the right knee. Note the presence of a soft-tissue swelling with intralesional calcifications in the proximal tibia (arrow). B. Three-month-follow-up lateral radiograph of the right knee shows more abundant superficial cartilaginous calcifications (arrow), cortical erosion, saucerization of the underlying cortex, and a rim of sclerosis on the anterior surface of the right proximal tibia.

more abundant calcifications within the lesion itself (**Figure 1B**). A radiographic survey confirmed the lesion to be solitary. MRI was performed, and revealed a lobular heterogeneous juxtacortical cartilaginous mass, adjacent to the tibial tubercle physis. The mass was hypointense on T1-weighted images but hyperintense on T2-weighted images (**Figure 2**). The mass did not appear to invade the intramedullary canal. CT confirmed calcifications within the cartilaginous mass (**Figure 3**).

Microscopic examination showed a hyaline cartilaginous tumor arranged in a lobular pattern. There was no evidence of an infiltrative growth pattern as seen in low-grade chondrosarcoma, confirming a diagnosis of periosteal chondroma (**Figure 4**). Local resection and curettage with iliac crest autograft was performed. At the three years and eight months postoperative follow-up, there was no radiographic evidence of recurrence (**Figure 5**).

Case 2

A 12-year-old boy noted a painless lump below the right knee a month before presentation to our institution. The mass did not interfere with his normal activities, and he seemed otherwise in excellent health. Physical examination revealed a firm, non-tender, non-mobile mass over the right proximal tibia. The affected knee had full range of motion and there was no neurovascular deficit.

Laboratory examinations and chest radiography were normal. Plain radiographs of the right knee revealed a partially calcified soft-tissue mass confluent with the anterior proximal tibia. The underlying cortex demonstrated scalloping and endosteal sclerosis (**Figure 6**). A radiographic survey revealed no other skeletal lesions. MRI confirmed a well-circumscribed, juxtacortical mass arising from the proximal tibia and extending anteriorly with intermediate signal intensity on T1-weighted images and high signal intensity on T2-weighted images. No soft tissue involvement or extension into the medullary cavity was seen. CT findings were similar to Case 1 (**Figure 7**). Biopsy and microscopic appearance were identical to Case 1, and a diagnosis of periosteal chondroma confirmed. Local resection and curettage along with autograft and allograft bone graft were performed. Six-month follow-up revealed no clinical or radiographic evidence of recurrence (**Figure 8**).

Case 3

A 15-year-8-month old boy noted a painless lump below the right knee for 8 years and 8 months before presentation to our institution. His normal activities were not limited and were healthy. Physical examination revealed an almost normal appearance of the right proximal tibia. The affected knee had full range of motion and there was no neurovascular deficit.

The initial plain radiograph of the right knee revealed a partially calcified soft-tissue mass at the anterior proximal tibia. The underlying cor-

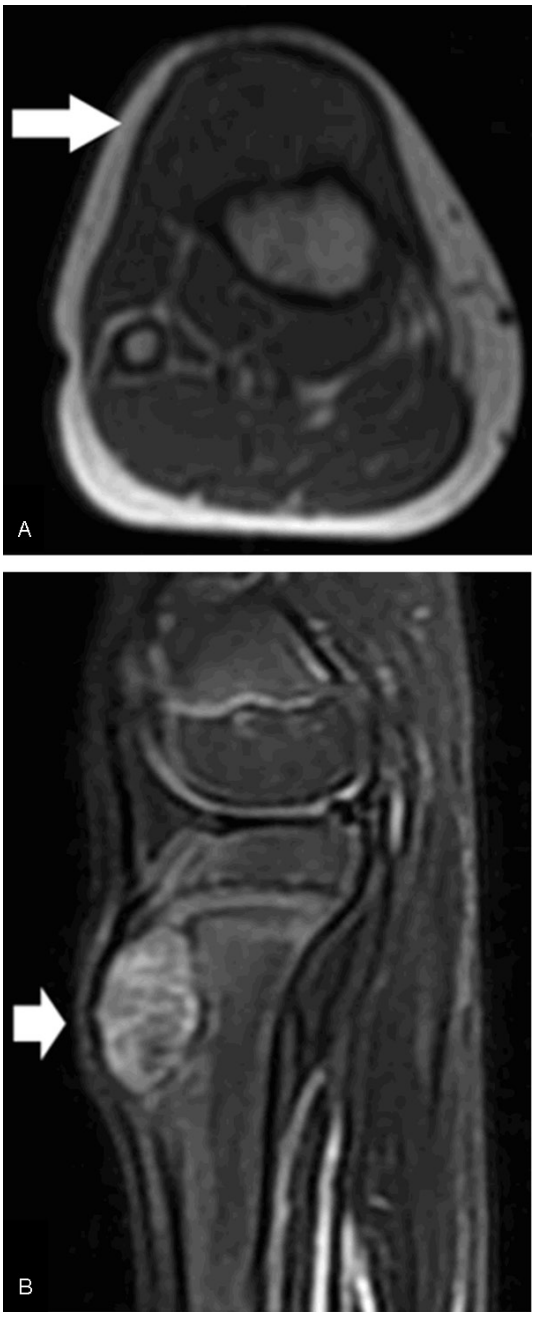

Figure 2. MR imaging of the right knee. Axial T1-weighted image (TR 380/TE 6.46) (A) and Sagittal T2-weighted image (TR 3680/TE 103.28) (B). Note a well-delineated juxtacortical mass (arrow). The mass was slightly hypointense to muscle on T1-weighted MR images (A), and markedly hyperintense to fat on T2-weighted images (B). No evidence of marrow invasion was seen.

tex demonstrated scalloping and endosteal sclerosis (**Figure 9A**). No treatment was performed for this patient. The follow-up radiographs showed the tumor almost healed spontaneously after 25 months initial presentation (**Figure 9B**), and thoroughly healed after 7 years and 1 month (**Figure 9C**).

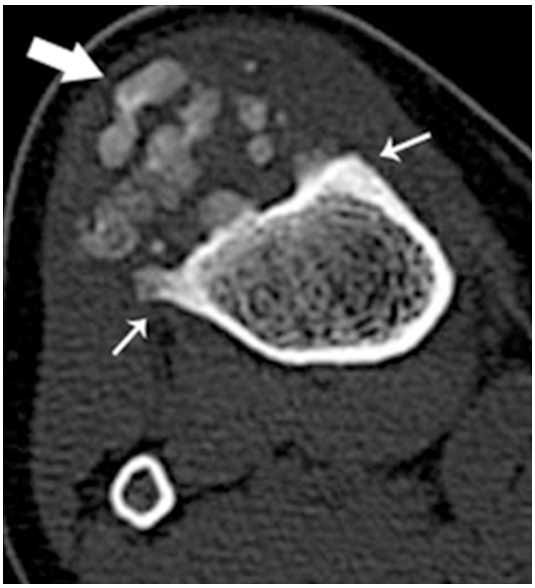

Figure 3. CT images of the right knee. Axial CT bone window image demonstrates the cartilaginous matrix calcification as dots (thick arrow) and cortical scalloping, sclerosisas well as an overhanging cortical margin (thin arrows).

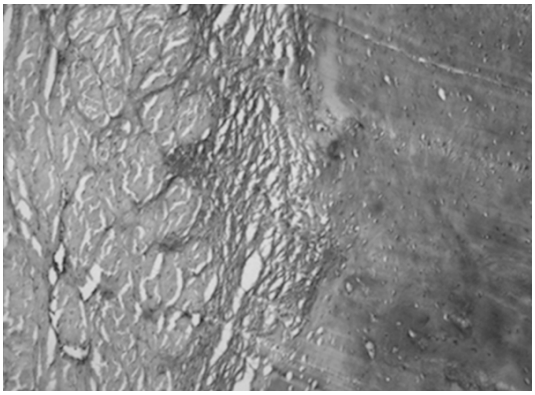

Figure 4. Low-power view of the tumor showing a lobular growth pattern covered by a fibrous periosteal capsule.

Discussion

Table 1 summarizes the literature concerning periosteal chondroma [1-48]. **Table 2** summarizes the location of lesions as reported in the available literature. Fifty-five in the humerus, 37 in the femur and 57 in variable locations in the hand and the wrist have been reported.

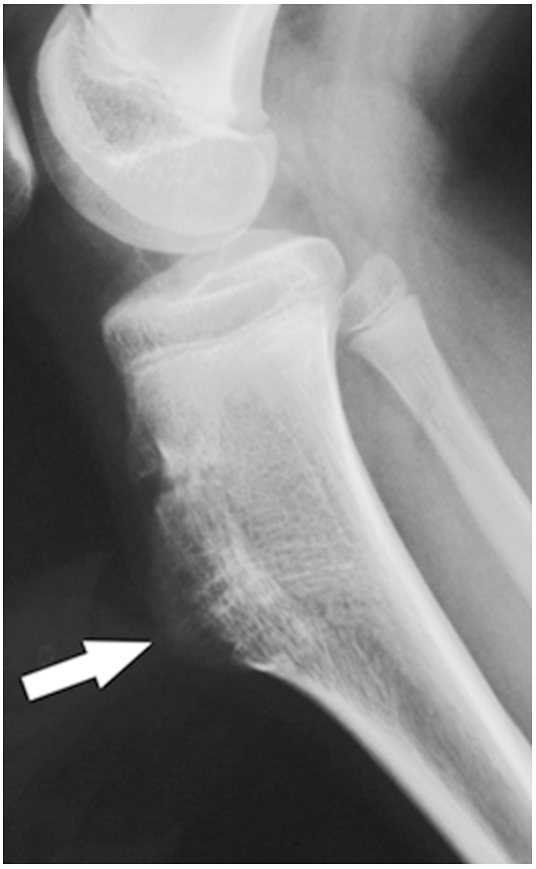

Figure 5. Three-year and eight-month follow-up lateral radiograph of the right knee shows no recurrence (arrow).

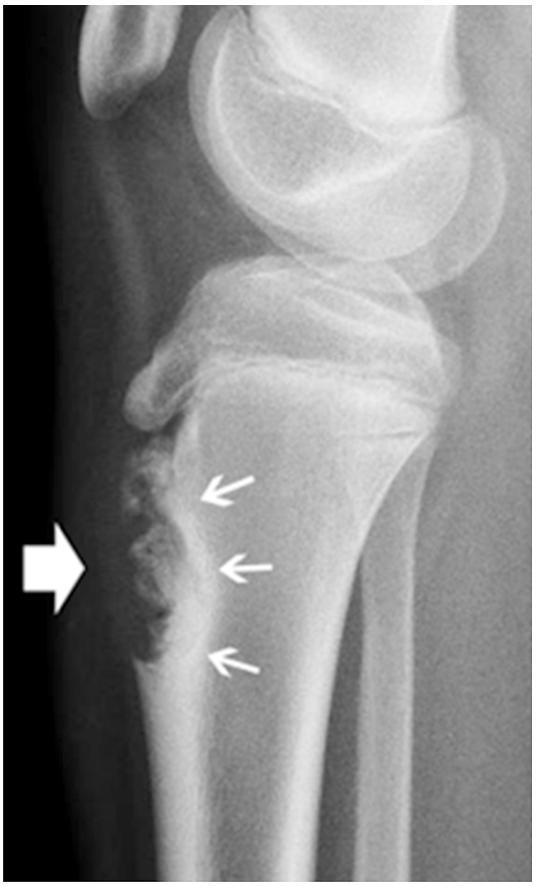

Figure 6. Lateral radiograph of the right knee of case 2 shows abundant superficial cartilaginous calcifications (thick arrow) and thickening of the cortex at its proximal and distal margins (thin arrows), in keeping with a "cortical buttress sign".

The age of patients at presentation ranged from 2 weeks to 78 years [1-48]. Five cases of local recurrence have been reported [7, 9, 41].

Periosteal chondroma usually arises at the osseous insertions of tendons and ligaments or at the metaphyseal region of the long tubular bones [4]. The cells of the periosteum, rather than the physis, appear to be the source for juxtacortical chondroma [8]. Clinical manifestations include a painless swelling with progressive onset [4] or mild local pain and swelling and possibly a palpable mass [2, 6, 34]. Unlike other benign cartilaginous lesions, such as osteochondromas or enchondromas, the tumor may continue to grow after skeletal maturity [2].

Plain radiographic features include an extracortical radiolucent soft tissue mass, with scalloping or remodeling of the underlying cortex, with an endosteal border of sclerosis, matrix calcifications (occurring in approximately 50% of patients), and a "cortical buttress sign" of the adjacent cortex [6, 9]. Irregularity of the osseous surface may be misinterpreted as a malignant tumor. Although similar in shape to a "Codman's triangle", which is most often seen in relation to osteosarcoma and aneurysmal bone cyst, the "periosteal buttress sign" delineates the proximal and/or distal extent of a benign-looking juxtacortical lesion, rather than an aggressive-appearing intramedullary lesion with soft tissue extension. Histologically, it presents periosteally produced woven to lamellar bone encasing hyaline cartilage [8].

CT is superior to plain radiographs in demonstrating these diagnostic features, and cortical scalloping and intralesional matrix calcifications [6, 8].

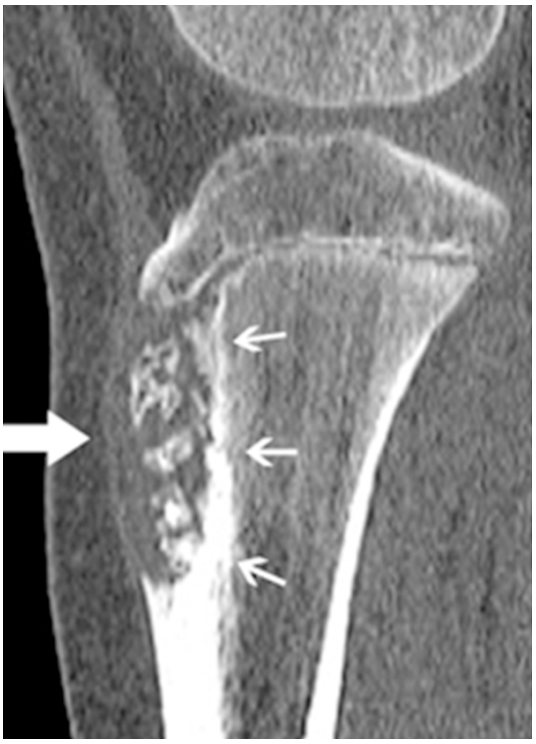

Figure 7. CT images of the right knee. Sagittal CT bone window image demonstrates cortical saucerization (thin arrows) and a small soft-tissue mass with the cartilaginous matrix calcification (thick arrow).

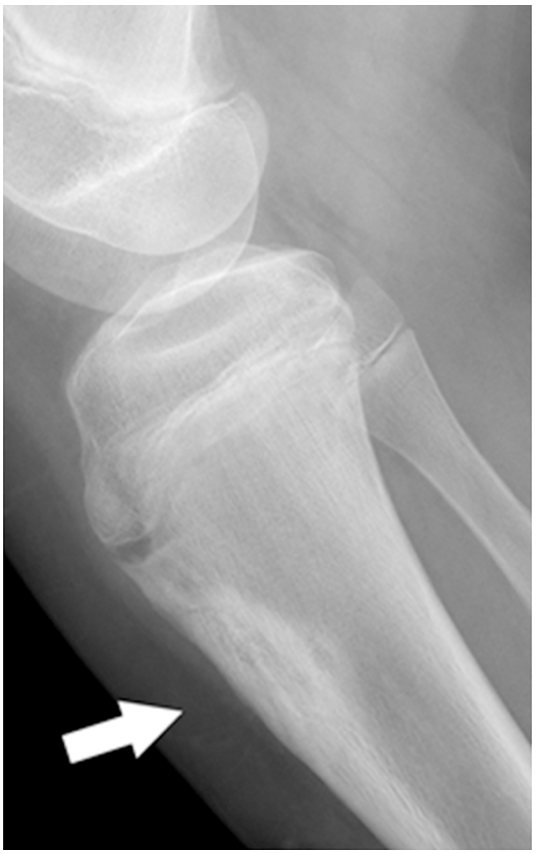

Figure 8. Six-month follow-up lateral radiograph of the right knee shows no recurrence (arrow).

On MR imaging, the typical features of a water-rich cartilaginous tumor are found, consisting of a matrix of hyperintense signal relative to fat on T2-weighted images, and of hypointense to isointense signal relative to muscle on T1-weighted images. Intralesional calcifications can be seen as areas of low signal intensity on both pulse sequences. The lesion is typically well delineated and often bordered by a hypointense rim on T2-weighted images. Neither bone marrow nor soft tissue edema is typically seen. Contrast enhancement is observed predominantly at the periphery of the cartilage nodules (ring-and-arc enhancement) [6, 23].

Radiographic differential diagnoses include other benign and malignant tumor and tumor-like conditions, such as an osteochondroma, enchondroma, and Ewing's sarcoma [2]. An enchondroma is located in the medullary cavity. Ewing's sarcoma is a fast growing osteolytic lesion causing an indistinct cortical margin in the affected region. Large soft tissue extension is typical [2]. An osteochondroma is composed of cortical and medullary bone that extend from the underlying bone and are continuous with the underlying medullary canal and cortical bone [3, 20]. The most important tumor to distinguish from periosteal chondroma is periosteal chondrosarcoma. Periosteal chondrosarcoma presents as a relatively slow-growing malignancy, reactive sclerosis and scalloping may be seen, similar to the appearance of a periosteal chondroma. However, periosteal chondrosarcomas are generally larger, occur in an older population, and tend to permeate the underlying bone with formation of bony spicules extending out from the cortex [2, 17, 34]. The size of reported periosteal chondrosarcomas (range, 3-14 cm; median, 4 cm) is considerably larger than periosteal chondromas (range, 1-6.5 cm; median, 2.5 cm). The size of the lesion is the most reliable indicator in distinguishing between periosteal chondromas and periosteal chondrosarcomas [17]. Other lesions that may present similarly include peri-

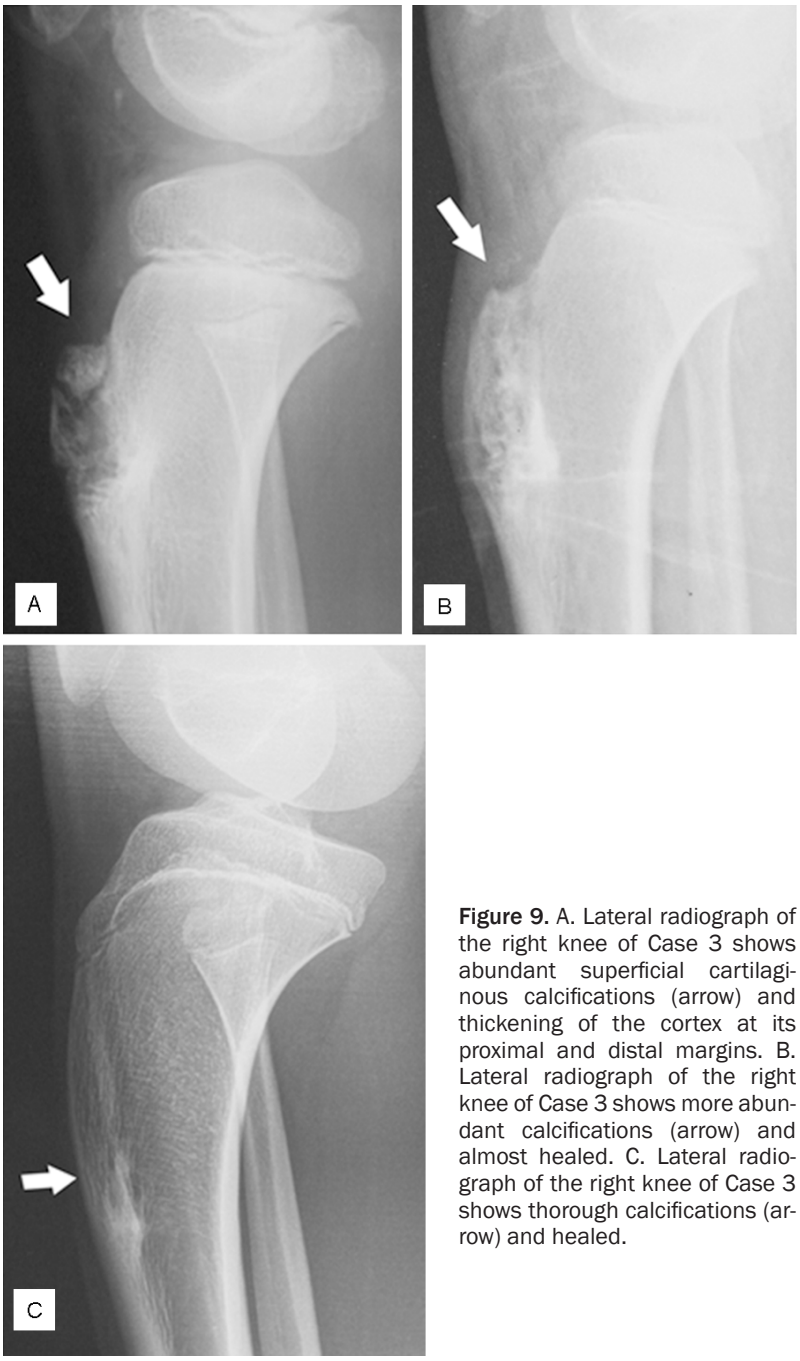

Figure 9. A. Lateral radiograph of the right knee of Case 3 shows abundant superficial cartilaginous calcifications (arrow) and thickening of the cortex at its proximal and distal margins. B. Lateral radiograph of the right knee of Case 3 shows more abundant calcifications (arrow) and almost healed. C. Lateral radiograph of the right knee of Case 3 shows thorough calcifications (arrow) and healed.

osteal osteosarcoma, aneurysmal bone cyst, subperiosteal hemangioma, subperiosteal ganglion, pigmented villonodular synovitis [3], and chondromyxoid fibroma [49, 50].

A periosteal chondroma occurring at the proximal tibial in skeletally immature patients, such as in our cases, may mimic an Osgood-Schlatter's disease. Clinically, Osgood-Schlatter disease is characterized by focal pain at the tibial tuberosity. On plain radiographs, fragmentation, if present, involves the tibial tuberosity itself, instead of the cortical bone underneath the tibial tuberosity. MR imaging may demonstrate an associated infrapatellar bursitis, marrow edema within the proximaltibia, and thickened cartilage anterior to the tibial tubercle [48]. In Case 1 and Case 2, the lesions were distal to the tibial tuberosity.

Gross examination reveals a hyaline cartilage mass with variable calcification juxtaposed to the cortex [8]. Histologically, periosteal chondroma is characterized by lobulated immature cartilaginous tissue, covered by a fibrous periosteal capsule. Pathologic differentiation with a low-grade chondrosarcoma may be difficult. Medullary invasion is the most valuable differentiating finding on histopathology, as this never occurs in periosteal chondroma [23].

En bloc excision resection of the periosteum, mass and fibrous capsule remains the preferred treatment of periosteal chondroma since local recurrence has been reported with curettage alone [8, 34, 37]. However, when clinical and radiographic findings are inconclusive, a preoperative excisional biopsy is mandatory [8]. Cryotherapy may be used as an adjunct to reduce the likelihood of local recurrence [8]. Observation may also be warranted if there is a firm diagnosis and the patient is symptom free and the latent lesion [34, 37]. Radiation is contraindicated, as the lesion is benign and radiation resistant [47]. No biopsy and no treatment were performed in Case 3 and the diagnostic

Table 1. Literature of periosteal chondroma since 1952 to 2014

Authors	Year	Cases
Lichtenstein et al. [1]	1952	6
Feinberg et al. [49]	1956	2
MEYER et al. [41]	1958	1
MERLINO et al. [46]	1964	1
Cary [26]	1965	1
Nosanchuk *et al.* [42]	1969	2
Rockwell *et al.* [4]	1972	8
Kirchner *et al.* [5]	1978	2
Bauer *et al.* [10]	1982	23
Calderone *et al.* [32]	1982	1
Boriani *et al.* [11]	1983	20
Pazzaglia *et al.* [12]	1985	1
Nojima *et al.* [20]	1985	46
Baber et al. [33]	1988	1
Lewis et al. [18]	1990	10
Abdelwahab et al. [23]	1990	1
Nguyen et al. [2]	1995	1
Morisaki et al. [28]	1996	1
Peidro et al. [37]	1996	1
Ishida et al. [13]	1998	1
Brien et al. [8]	1999	16
Sinha *et al.* [27]	1999	1
Ricca et al. [48]	2000	1
Robinson *et al.* [17]	2001	11
Inoue et al. [31]	2001	2
Kahn et al. [21]	2002	1
Pérez-Alonso et al. [3]	2003	1
Takada *et al.* [9]	2005	5
Matsushima *et al.* [30]	2006	1
Luevitoonvechkij et al. [14]	2006	1
Flint et al. [6]	2007	1
Singh et al. [35]	2008	1
Akiyama et al. [16]	2008	1
Domson et al. [15]	2008	1
Yamauchi *et al.* [44]	2008	1
Al-Qudah et al. [36]	2009	1
Fahim et al. [34]	2009	1
Parodi et al. [47]	2011	1
Yoshimura et al. [45]	2011	1
Sakai et al. [25]	2011	5
Akansu et al. [38]	2012	1
Zhao et al. [29]	2012	1
Santanelli et al. [43]	2013	1
Vancauwenberghe et al. [24]	2013	1
Imura et al. [22]	2014	1
Kosaka et al. [40]	2014	1
Miller et al. [7]	2014	7
Rabarin et al. [39]	2014	24

Table 2. Locations of periosteal chondroma from the literature since 1952 to 2014

Location	Numbers
Humerus	55
Femur	37
Hand	57
Feet	12
Tibia	15
Fibula	4
Radius	8
Ulnar	1
Cervical	3
Rib	8
Clavicle	2
Scapula	1
Ischium	2
Sacrum	3
Ilium	2
Pubic symphysis	1

evidence of periosteal chondroma was not very strong. However, the radiological findings of Case 3 were extremely similar to those of Case 1 and Case 2. The treatment was closely followed observation care. So we propose a hypothesis that some cases of periosteal chondroma may heal spontaneously.

The recurrence rate of periosteal chondroma is low. In the present review of the literature this rate is at 2.3% (5 among 221 cases). No malignant transformation or metastases have been reported [21].

This is the first report of periosteal chondroma of the proximal tibia from the northern part of China. Although there are no published articles for the incidence of this tumor in China, the authors believe that periosteal chondroma is very rare among Chinese people. The clinical findings and the typical radiological features of the presented cases pointed to the diagnosis of periosteal chondroma, which must be confirmed by histological examination.

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Disclosure of conflict of interest

None.

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