Osteoblastoma of the patella, a rare benign bone tumor with an uncommon site: A case report

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Abstract. Osteoblastoma is a rare, benign, bone-forming tumor that is frequently observed in the spine and long tubular bones. There are very few reports available on osteoblastoma of the patella. The present study reported an extremely rare case of a 22-year-old male adult who presented with an osteoblastoma of the patella. He was treated via intralesional curettage of the patella with subsequent bone grafting. After the intervention, he made an uneventful recovery with no recurrence after a follow-up of 2 years. Making an accurate diagnosis of osteoblastoma of the patella is challenging and important for determining the correct treatment modality and prognosis, therefore, the present case may be helpful in the diagnosis and treatment of osteoblastoma of the patella.

Introduction

Osteoblastoma is a rare, benign, bone-forming tumor accounting for less than 1% of all primary bone tumors and 3.5% of benign bone tumors (1); most osteoblastomas frequently arise in the spine (approximately 30-40%) and long (approximately 30%) and small tubular bones of the hands and feet (approximately 10-20%) and infrequently arise in the flat bones and jaw bones (2,3). Osteoblastoma mainly affects adolescents and young adults; 75-90% of patients are aged between 6 and 30 years, with a male-to-female ratio of 2:1 (2,3). The patella is an uncommon site for the occurrence and development of bone tumors. The vast majority of tumors of the patella are benign, with a significant incidence of giant cell tumor and chondroblastoma, accounting for 33 and 16% of all tumors of the patella, respectively (4). Osteoblastoma of the patella is extremely rare, accounting for approximately 2% of all tumors of the patella (4,5). To the best of our knowledge, very few cases of osteoblastoma of the patella have been reported (Table I) (6-10). Therefore, herein, we reported an extremely rare case of osteoblastoma of the patella in a 22-year-old young male adult. The patient was treated with thorough intralesional curettage and with subsequent autologous iliac cancellous bone grafting; he had an uneventful recovery with no clinical and radiological evidence of recurrence after a follow-up of 2 years. The patient was informed that medical documentation and information regarding the case would be submitted for publication, and he provided consent.

Case presentation

A 22-year-old male young adult was admitted to the 940th Hospital of Joint Logistics Support Force of Chinese People's Liberation Army (Lanzhou, Gansu, China) on December 5, 2020, with a 6-month history of intermittent pain in his right knee in absence of trauma. He did not have a significant personal or family medical history. His physical examination showed no obvious swelling or redness, and he had a normal skin temperature on the right knee. However, there was direct tenderness on the right patella. The range of motion of the right knee was full. The floating patella test, patella grind test, McMurray's test, anterior-posterior drawer test, and valgus-varus stress test were all negative. His neurological examination was normal. Laboratory tests revealed that his routine blood, liver function, kidney function, erythrocyte sedimentation rate, C-reactive protein, and tumor marker results were within the normal range. Radiography of the knee showed an osteolytic lesion at the medial superior quadrant of the right patella surrounded by a sclerotic margin with high density (Fig. 1). Computed tomography (CT) scan revealed a circular mixed-density image of the right patella that was surrounded by a thick sclerotic margin with high density (Fig. 2A). Three-dimensional CT showed no pathological fracture or cortical bone breakthrough (Fig. 2B and C). Magnetic resonance imaging (MRI) displayed a low-intensity signal mass on T1-weighted image

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Table I. Summary of all published cases of patellar osteoblastoma to date.

Cases	Year	Language	Age (years)/ Gender	Symptoms	Therapies	Follow-up (months)	Outcomes	(Refs.)
Sicard *et al*	1979	French	N/A	N/A	Patellectomy after failed wide curettage	N/A	Cure	(6)
De Coster *et al*	1989	English	29/Male	Sudden pain and swelling	Intralesional curettage and autologous iliac cancellous bone grafting	18	No evidence of recurrence and a normal incorporation of the cancellous	(7)
Shen *et al*	2001	English	34/Male	Pain and mild swelling, tenderness	Intralesional curettage and allogeneic bone grafting	24	Graft Complete healing of the lesion without evidence of recurrence	(8)
Bhagat *et al*	2008	English	38/Male	Pain	Excision of the lesion	48	Living	(9)
Zhong *et al*	2010	Chinese	20/Female	Pain and swelling, tenderness	Intralesional curettage and autologous iliac cancellous bone grafting	3	No evidence of recurrence and the cancellous graft healed well	(10)
Current case	2023	English	22/Male	Intermittent pain, tenderness	Intralesional curettage and autologous iliac cancellous bone grafting	24	No clinical and radiological evidence of recurrence	

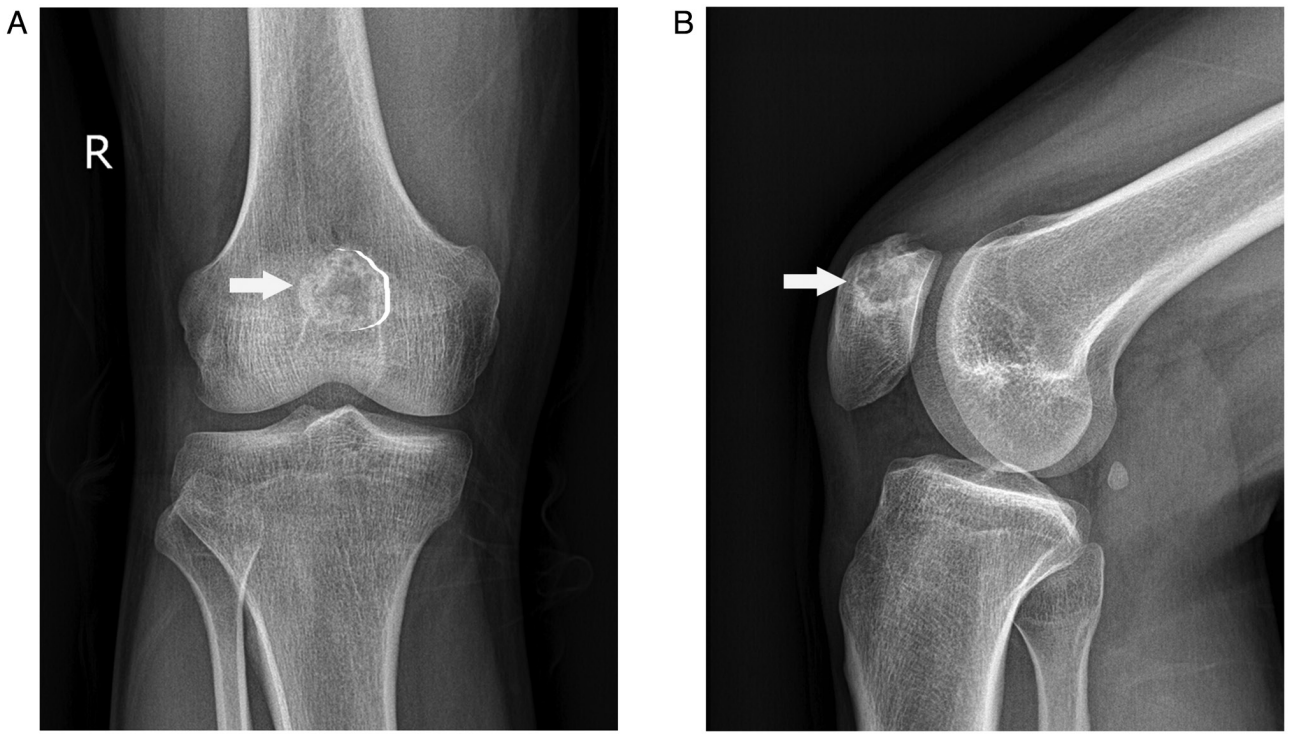

Figure 1. X-ray images showing an osteolytic lesion at the medial superior quadrant of the right patella surrounded by a sclerotic margin with high density. (A) Anteroposterior plane, (B) Lateral plane.

(Fig. 3A) and high-intensity signal mass on T2-weighted image (Fig. 3B, C and D) with a well-defined lesion of the patella. Considering the clinical and imaging manifestations of the patient, benign bone lesions including chondroblastoma, osteoid osteoma, osteoblastoma, and bone abscess, were considered as the initial diagnosis.

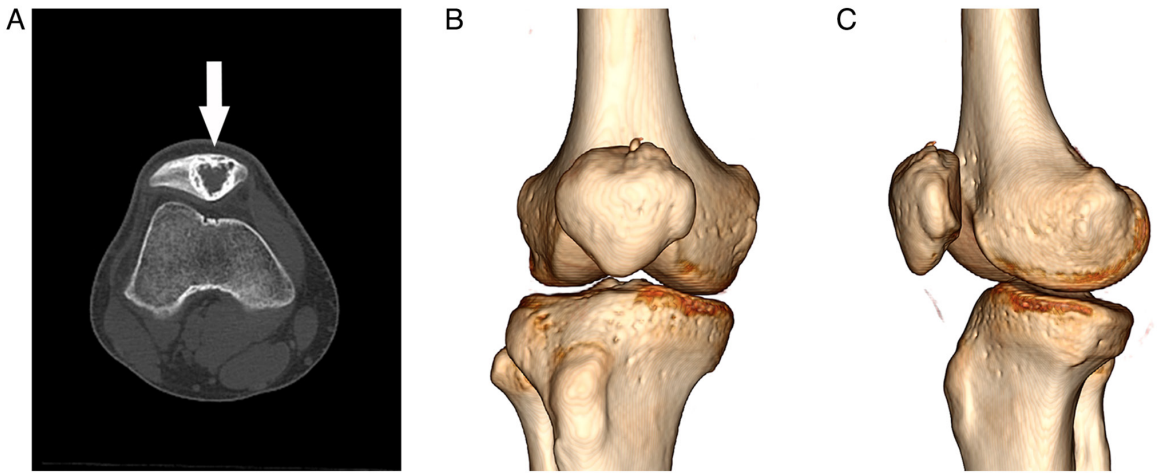

Figure 2. CT scan showing a circular mixed-density image of the right patella surrounded by a thick high-density sclerotic margin with no evidence of extra-articular invasion. (A) Axial plane. (B) Frontal view of three-dimensional CT. (C) Lateral view of three-dimensional CT. CT, computed tomography.

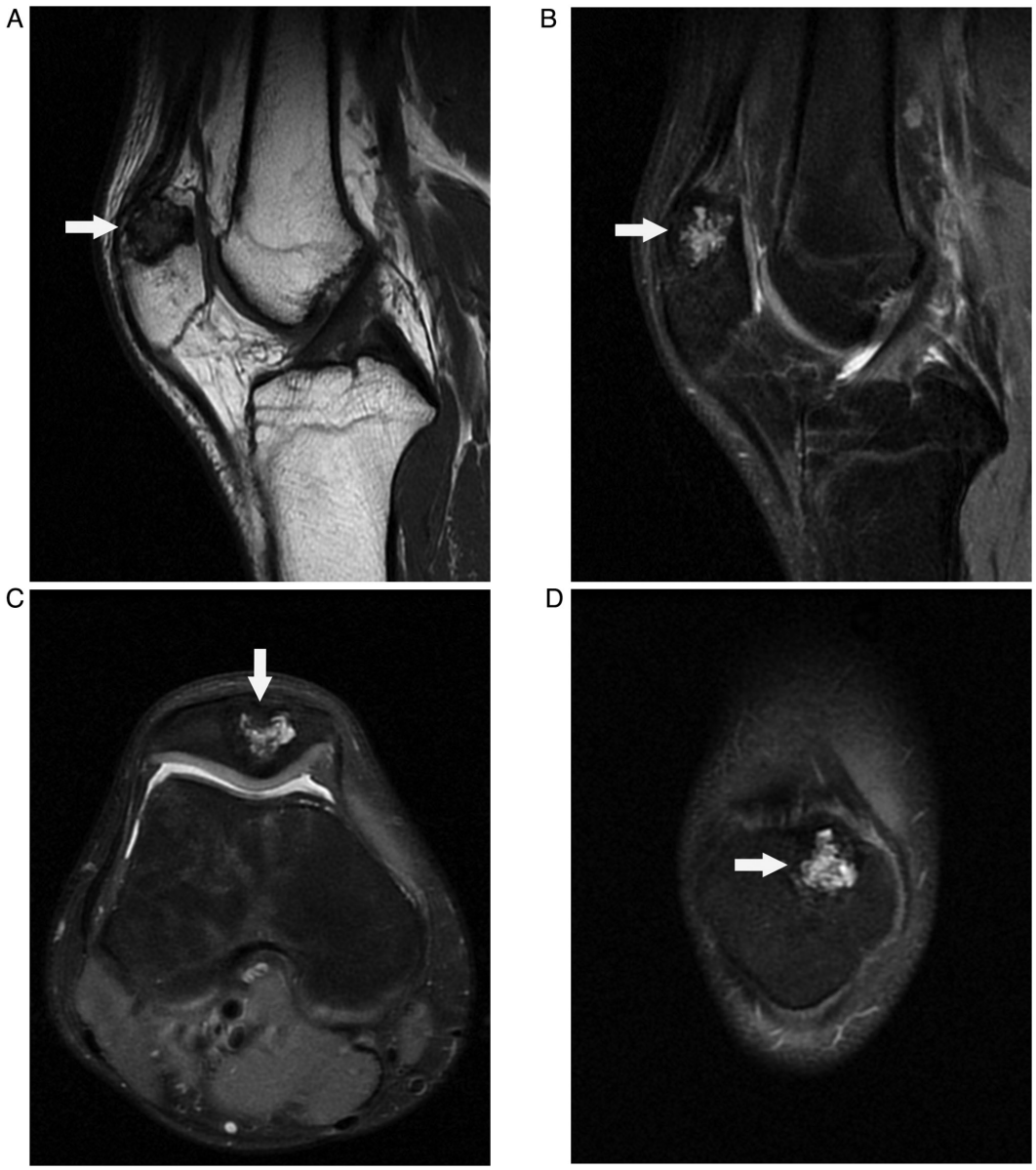

Figure 3. Magnetic resonance imaging showing a low-intensity signal mass on the T1-weighted image and a high-intensity signal mass on the T2-weighted image with a well-defined lesion of the patella. (A) Sagittal plane of the T1-weighted image. (B) Sagittal plane of the T2-weighted image. (C) Axial plane of the T2-weighted image. (D) Coronal plane of the T2-weighted image.

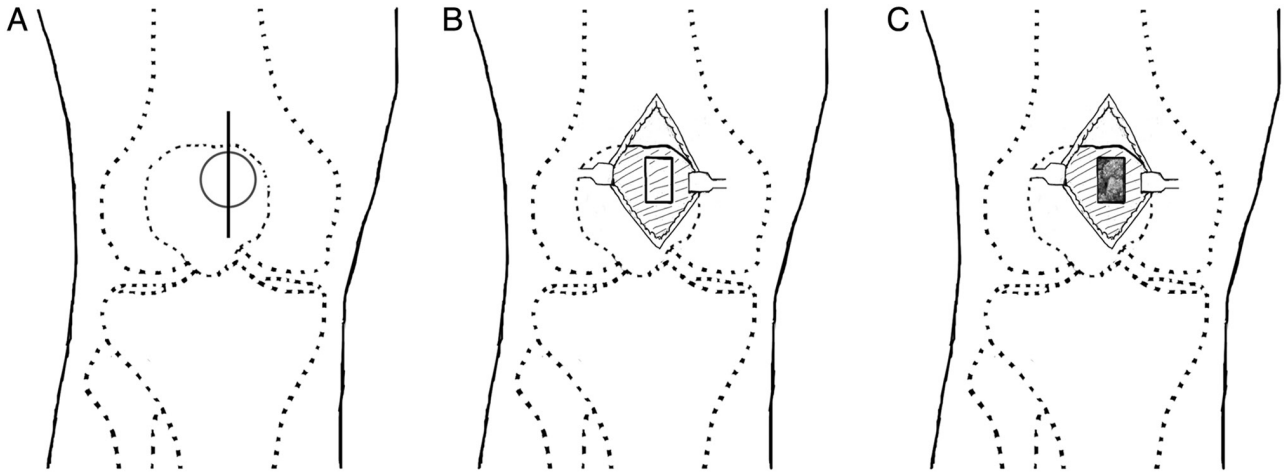

Figure 4. Surgical procedures. (A) Lesion located on the medial superior quadrant of the right patella and a 4-cm longitudinal anteromedial incision was made. (B) The bone window (0.5x1.0 cm) was located on the medial superior quadrant of the patella. (C) Intralesional curettage and with subsequent autologous iliac cancellous bone grafting.

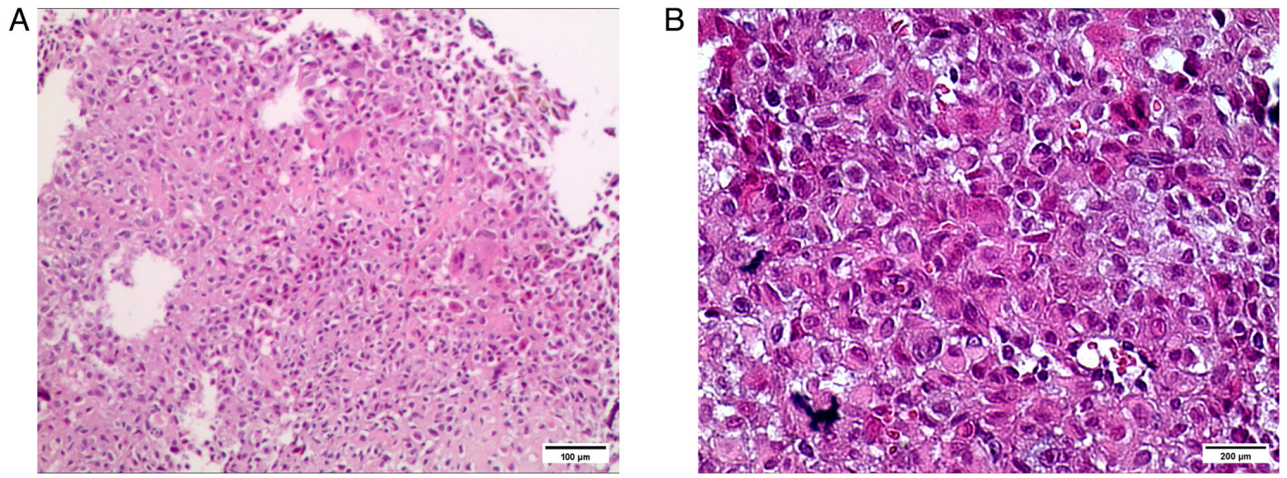

Figure 5. Histopathological examination showing oval osteoblasts were distributed in sheets, in which focal multinucleated giant cells were observed, the histopathological findings are consistent with the diagnosis of benign osteoblastoma. (A) Hematoxylin-eosin stain (magnification, x100). (B) Hematoxylin-eosin stain (magnification, x400).

The preoperative contraindications were excluded. The patient was administered spinal anesthesia in the supine position by applying an air pressure tourniquet on the right thigh (pressure 35 kPa for 90 min). The right knee and homolateral iliac region were routinely disinfected and draped. First, a 3-cm oblique incision along the anterior superior iliac spine was made for cutting the cancellous bone. In total, approximately 2 cm^3 (2.0x1.0x1.0 cm) autologous iliac cancellous bone was removed for subsequent grafting. Then, to expose the right patella, a 4-cm longitudinal anteromedial incision was made (Fig. 4A), and the soft tissue was not invaded by the bone tumor. The bone window (0.5x1 cm) was located on the medial superior quadrant of the patella (Fig. 4B). Thorough intralesional curettage of the bone tumor was performed. The specimen contained a mixture of sand-like, pale-yellow substances. The tumor border was ground by a high-speed burr and cauterized using a high-frequency electrotome. The tumor cavity was inactivated by anhydrous ethanol and washed with distilled water for 20 and 10 min, respectively. To reconstruct bone defects, the tumor cavity of the patella was subsequently filled with autogenous iliac cancellous bone (Fig. 4C). Finally, the intraoperative specimen was submitted for histopathological examination.

Pathologist conducted histopathological examination through hematoxylin-eosin staining. The intraoperative specimen was made into paraffin sections through the procedures of fixation, washing, dehydration, transparency, wax penetration, embedding, sectioning, and then underwent the procedures of dewaxing, hydration, staining (hematoxylin, 15 min), washing, differentiation, washing, re-staining (eosin, 3 min), dehydration, transparency, and sealing before microscopy. The histopathological examination findings showed that oval osteoblasts were distributed in sheets, in which focal multinucleated giant cells were observed. The histopathological findings were consistent with the diagnosis of benign osteoblastoma (Fig. 5).

At the last follow-up 2 years postoperatively, no clinical and radiological evidence of local recurrence was noted, and

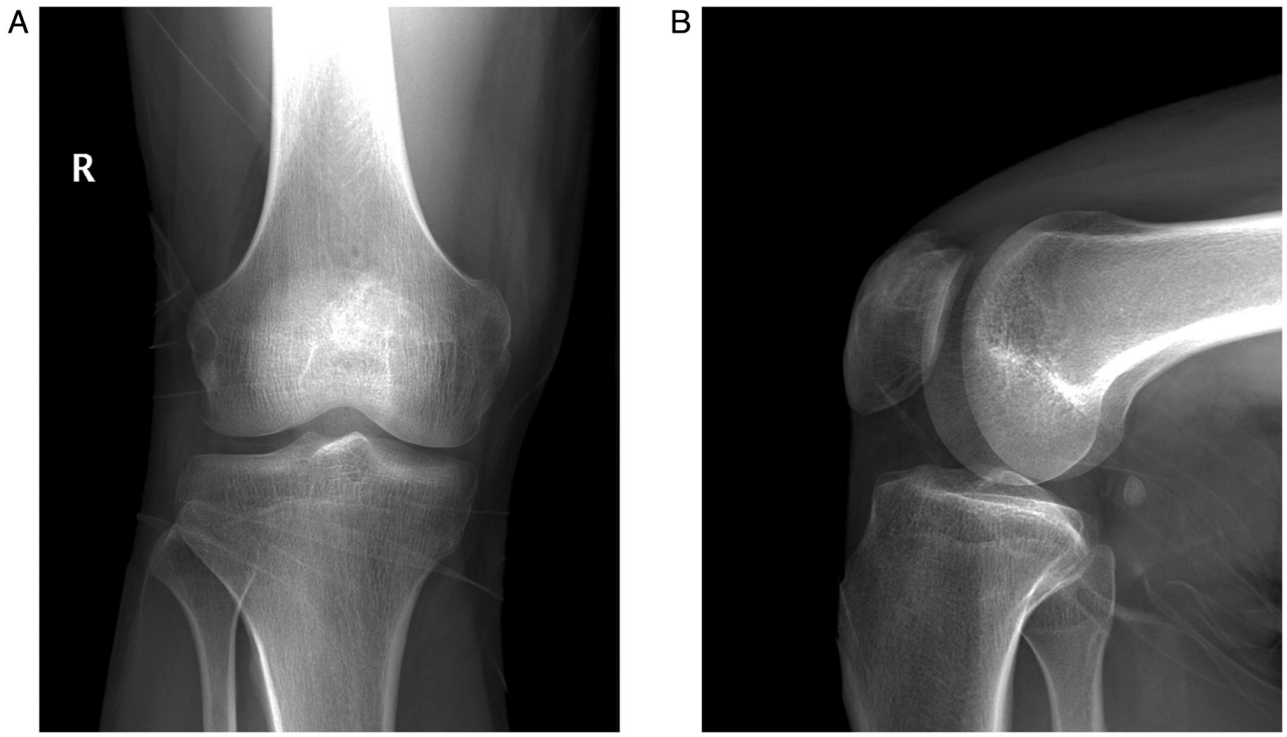

Figure 6. X-ray images of the right knee at the 2-year follow-up showing that the bone graft was blurred and fused; there was no radiological evidence of local recurrence. (A) Anteroposterior plane and (B) lateral plane.

the bone graft was blurred and fused (Fig. 6). The patient's sports and physical functioning domain score was 100 points. In spite of satisfactory outcome, we require a long-term regular follow-up in order to perform an early detection and treatment for a possible recurrence.

Discussion

The clinical presentations of osteoblastoma differ depending on its location. Cranial osteoblastoma may present as an enlarging mass with or without pain and neurological symptoms (11). Osteoblastoma of the spine can present with localized pain, night pain, extremity weakness, and radicular pain (12,13). The aggressive variants of osteoblastoma usually present with more intense pain, which can be due to localized areas of destruction (14). The most common symptoms of osteoblastoma of the patella are pain and swelling (7,8). Local tenderness may be noted during physical examination (8).

The imaging manifestations of osteoblastoma differ depending on its location, and the results lack specificity. The most frequent X-ray manifestations of the osteoblastoma of the patella are that of an osteolytic lesion, with or without matrix mineralization and pathological fracture of the patella, surrounded by a high-density sclerotic margin with no evidence of extra-articular invasion (7,15). Aggressive spinal osteoblastoma can break through the bony cortex and invade the spinal canal and paravertebral soft tissue (16,17). CT scans are superior to MRI in showing calcification of osteoblastoma and thus are used to further confirm a well-defined lesion with fine calcifications. However, the radiological characteristics of CT are atypical. Tang P *et al* reported a case of osteoblastoma of the rib that was misdiagnosed as lymphomatous by CT and was confirmed by pathological examination (18). MRI helps in evaluating bone marrow edema and soft tissue component or extension of the tumor; however, there is no significant specificity to this finding.

The pathological examination findings of osteoblastoma reveal that the lesion is composed of irregular trabeculae of woven bone, lined by active osteoblasts within a copious vascularized fibrous stroma containing focal multinucleated giant cells (7,8). The cartilaginous matrix may be present rarely (19). Osteoblastoma is difficult to distinguish from osteoid osteoma due to their similar imaging and histopathological patterns (20). Some of the histopathological features suggesting the diagnosis of the aggressive variant included surrounding soft tissue invasion, large and more irregular trabeculae of woven bone, ace- or sheet-like osteoid production, abundant large epithelioid osteoblasts, increased mitotic figures, disordered osteoid matrix, less vascularized fibrous stroma (3,21).

The treatment of osteoblastoma depends on the location within the bone and the aggressiveness of the tumor (20,22). In theory, the treatment options available for osteoblastoma of the patella include intralesional curettage followed by bone grafting or bone cement, en-bloc resection of the lesion, and even patellectomy. To treat the osteoblastoma of the patella, intralesional curettage with subsequent allogeneic bone grafting has been previously performed (7,8). Intralesional curettage can be effective but is related to a recurrence rate of 15-25% (23). Patellectomy can be the used for a failure after wide intralesional curettage (6). En-bloc resection is the preferred treatment for osteoblastoma of some irregular bones due to its propensity for local recurrence. Garvayo *et al* performed an en-bloc resection

to treat cranial osteoblastoma and observed no recurrence at 3 years follow-up (11). Mohammadi *et al* performed total surgical resection for treating maxillary osteoblastoma in a 10-month-old infant and found no recurrence during 10 months of follow-up (24). The treatment of unresectable or extensive osteoblastoma is difficult. Osteoblastoma can respond to denosumab. Yamaga *et al* reported a case of unresectable osteoblastoma of the cervical spine that was controlled with denosumab (25). Wong *et al* reported a case of aggressive osteoblastoma with a secondary aneurysmal bone cyst that was effectively treated with denosumab (22). If there is evidence of persistent aggressive behavior of the tumor or recurrence, radiotherapy should be considered. Long-term imaging surveillance after surgical resection is essential due to the risk of local recurrence.

However, there were several limitations in our case that should be noted. First, a intraoperative image was not captured during autogenous iliac cancellous bone grafting. Second, the 2-year follow-up period is relatively short, so we need to conduct long-term follow-up to confirm that our surgical procedures can achieve long-term good outcomes. Third, case report lack representativeness, and the research conclusion of our case cannot be summarized as general conclusion. In order to further study this rare bone tumor with an uncommon site, more case reports are needed.

In summary, our patient underwent thorough intralesional curettage of the patella with subsequent autogenous iliac cancellous bone grafting. This treatment was found to be effective as there was no clinical and radiological evidence of recurrence after a follow-up of 2 years. The accurate diagnosis of osteoblastoma of the patella is challenging and important in determining the appropriate treatment modality and prognosis.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Authors' contributions

HZ, FL, and YQ performed the operation together. FL designed the study, collected data and drafted the manuscript. YQ assisted with study design, data collection and in the editing of the manuscript. FL and YQ contributed to this paper equally. SZ and XS made substantial contributions to data acquisition and manuscript revision. HZ assisted with study design and in the editing and revising of the manuscript. All authors read and approved the final manuscript. HZ and FL confirm the authenticity of all the raw data.

Ethics approval and consent to participate

Not applicable.

Patient consent for publication

Written informed consent was obtained from the patient for publication of the patient's data and images.

Competing interests

The authors declare that they have no competing interests.

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