Case Report

Curettage through a wide cortical window for treatment of a primary aneurysmal bone cyst of the patella

Jincai Zeng1,*, Ming Zhou2,*, Lihua Xu3,*, Lifan Zhu1, Zhanjun Yan1, Weidong Wu1 and Zhenguo Qiao4

Abstract
A 27-year-old man presented with intermittent right knee pain for 1 year with no previous trauma. Physical examination revealed only tenderness over the patella. Typical fluid–fluid levels were visible on magnetic resonance imaging (MRI), which highly suggested aneurysmal bone cyst (ABC) of the patella. After removal of a large window of thin cortical bone, curettage and bone grafting followed by cerclage wiring was performed. Histology confirmed the initial diagnosis of primary ABC of the patella. At the final follow-up visit at 71 months after surgery, the patient had normal joint activity with no pain or evidence of recurrence. Previous publications indicated patellectomy in the initial series, but curettage and bone grafting have more recently provided excellent results and good graft incorporation in most cases, even for aggressive lesions. In our patient, thorough curettage and bone grafting through a wide cortical window followed by cerclage wiring fixation and figure-eight sutures was a successful treatment option for primary ABC of the patella without articular disruption.

Keywords
Patella, bone cysts, aneurysmal, curettage, treatment, case report

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1Department of Orthopaedics, Suzhou Ninth People’s Hospital (Affiliated Wujiang Hospital of Nantong University), Suzhou, China
2Department of Orthopaedics, Wuxi No. 9 People’s Hospital Affiliated to Soochow University, Wuxi, China
3Department of Science and Education, Nantong No. 6 People’s Hospital, Nantong, China
4Department of Gastroenterology, Suzhou Ninth People’s Hospital (Affiliated Wujiang Hospital of Nantong University), Suzhou, China

*These authors contributed equally to this work.

Corresponding author:
Zhenguo Qiao, Department of Gastroenterology, Suzhou Ninth People’s Hospital (Affiliated Wujiang Hospital of Nantong University), 2666 Ludang Road, Suzhou, Jiangsu 215200, China.
Emails: qzg66666666@163.com; qiaozhenguo@ntu.edu.cn
Introduction

An aneurysmal bone cyst (ABC) is a benign aggressive lesion that consists of a lobulated blood-filled cavity, which was first described by Jaffe and Lichtenstein in 1942. ABC of the patella is rare, accounting for less than 1% of all ABC cases, and it includes two types: primary cysts occurring without any precedent lesion, and secondary cysts following preexisting lesions such as giant cell tumor, chondroblastoma, telangiectatic osteosarcoma, and osteoblastoma.

Case report

In February 2011, a 27-year-old man presented with intermittent right knee pain for 1 year with no previous trauma. On physical examination, there was no swelling or signs of local inflammation in the right knee, but there was tenderness over the patella and a full range of active motion. Laboratory test results including hemoglobin, C-reactive protein (CRP), and the erythrocyte sedimentation rate (ESR) were all within normal ranges.

A lateral radiograph of the right knee showed a radiolucent lesion that occupied greater than 80% of the patella with a thin cortex and septations (Figure 1(a)). An axial computed tomography (CT) scan demonstrated an osteolytic lesion with cortical thinning and concealed a cortical breach and endosteal scalloping (Figure 1(b)). Sagittal magnetic resonance imaging (MRI) revealed a large multi-loculated lesion with a high intensity on a T2-weighted image with typical fluid–fluid levels and a thin cortex with an anterior crack, but it was limited to the periosteum and there was no new periosteal bone formation. Normal patellofemoral cartilage structure was also seen (Figure 1(c)). All radiological images highly suggested ABC of the patella.

Intraoperatively, a transverse arc surgical approach was used, and apparent varices were seen over the anterior patella surface, but no cortical bone destruction or tissue mass was found. After removal of a 2.5-cm × 3.5-cm bone window in the anterior patella, a separated cystic lesion filled with serosanguinous fluid was observed. The subchondral bone was found to be very thin. After thorough curettage of the lesion and phenol cauterization, the defect was packed with a combination of autologous cancellous bone that was taken from the iliac crest and synthetic bone substitute (Wright Medical, Arlington, TN, USA). The bone window was then closed using the big bone segment of the patella, which was removed and also previously curetted and cauterized with phenol, and then fixed with cerclage wiring and figure-eight sutures (Figure 1(d)). Histological examination revealed cavernomatous spaces that were lined with endothelial cells and contained red blood cells and capillaries, which confirmed the initial diagnosis (Figure 1(e)).

Postoperatively, the knee was immobilized in a brace for 2 weeks followed by gradually quadriceps strengthening and joint mobilization exercises. Two months later, the patient experienced no pain and had a normal range of knee movement. After 1 year, the cerclage and figure-eight wires were removed under local anesthesia in an outpatient small operation room. At the final follow-up visit 71 months after surgery, the patient had no complaints and the bone graft remained well-incorporated without signs of local recurrence (Figure 2).

The patient provided written informed consent for publication of this report. This was a retrospective case report, and therefore, institutional review board approval was not required.

Discussion

The development of primary ABC in the patella is rare. An English literature review (Table 1) indicated that there
were only 25 reported cases of primary ABC of the patella, including the present case, over the past 60 years.

Most primary ABC of the patella occur in patients who are younger than 20 years old, and there is a slight female predominance; ABC is often associated with a coincidental history of trauma and subsequent pain and swelling. However, Table 1 shows that the cysts are mostly found in men (19 cases) and only 7 of 25 cases were younger than 20 years, bringing the average age to 25 years (range, 9 to 56 years), which is appreciably higher than the previous report. There also seems to be a slight preponderance of non-trauma patients, which suggests that trauma is probably the excitant, but not the originator of the ABC’s pathological process.

Although bone scans may assist with the diagnosis of ABC of the patella, it is accepted that CT and MRI are particularly useful in delineating the expansile osteolytic lesions, with a thin shell of cortical bone and multi-loculated cavities. The presence of a fluid–fluid level and intralesional septations on MRI are typical radiologic features for this lesion. The diagnosis of primary ABC of the patella was finally confirmed by histopathology. Because the

Figure 1. (a) Lateral radiograph showing a radiolucent lesion occupying greater than 80% of the patella with a thin cortex and septations. (b) Axial computed tomography (CT) scan showing an osteolytic lesion with cortical thinning, a suspicious breach in the subchondral bone on the lateral articular facet, and endosteal scalloping. (c) Sagittal magnetic resonance imaging (MRI) showing a multi-loculated lesion with high intensity on a T2-weighted image with a typical fluid–fluid level and thin cortex with an anterior crack, which is limited to the periosteum. (d) Anteroposterior and lateral radiographs showing satisfactory bone graft and cerclage wiring fixation of the patella 2 days postoperatively. (e) Histological feature showing cavernomatous spaces lined with endothelial cells and containing red blood cells and capillaries making up the aneurysmal bone cyst. (hematoxylin–eosin stain; original magnification ×20).
Characterized features were visible on MRI, a benign patella tumor was considered, so we did not perform an open biopsy preoperatively to exclude any malignancy.

Most published cases describe “latent” (stage I), “active” (stage II), or aggressive (stage III) lesions based on the Enneking staging system for benign skeletal tumors. Treatment of primary ABC of the patella includes partial or total patellectomy, incisional curettage with bone grafting, and arthroscopic excisional biopsy of the cyst and curettage, followed by filling the cavity with bone cement at a second stage. Based on our review, among these nine cases that were before Mercuri et al.’s report in 1991, total patellectomy was performed in seven patients and partial patellectomy was performed in one patient. Since then, there have been no patients who were treated with patellectomy even for aggressive (stage III) lesions, except for those reported in the recent publications by Saoji et al. in 2014 and Çetinkaya et al. in 2016. Patellectomy was most frequently used in the initial patients, but this seemed
| Study                      | No. of patients | Sex/age (years) | Trauma | Site | Fracture | Duration (months) | Treatment | Follow-up (months) | Recurrence |
|---------------------------|-----------------|-----------------|--------|------|----------|------------------|-----------|--------------------|------------|
| Besse et al.\(^2\)        | 1               | M/15            | N/A    | R    | No       | N/A              | TP        | N/A                | N/A        |
| Copeland et al.\(^3\)     | 2               | M/22, M/29      | No, Yes| L, R | No, No   | 3, 24            | TP, TP    | 50, 4              | No, No     |
| Linscheid and Dahlin\(^4\)| 1               | M/15            | Yes    | L    | No       | 12               | TP        | 120                | No         |
| Park and Chung\(^5\)      | 1               | M/22            | Yes    | L    | No       | 36               | TP        | 24                 | No         |
| Srivastava et al.\(^6\)   | 1               | F/30            | Yes    | L    | Yes      | 6                | TP        | 41                 | No         |
| Faris et al.\(^7\)        | 1               | M/22            | Yes    | L    | No       | 1                | C+BG      | 24                 | No         |
| Mapelli et al.\(^8\)      | 1               | M/33            | Yes    | N/A  | No       | N/A              | PP        | N/A                | No         |
| Mercuri et al.\(^9\)      | 1               | M/31            | N/A    | N/A  | No       | N/A              | TP        | 25                 | N/A        |
| Pevny and Rooney\(^10\)   | 1               | F/15            | No     | R    | No       | 3                | C+BG      | N/A                | N/A        |
| Castro and Irwin\(^1\)    | 1               | M/14            | Yes    | L    | No       | 5                | C+BG      | N/A                | N/A        |
| Kumar et al.\(^11\)       | 1               | F/16            | N/A    | L    | N/A      | N/A              | N/A       | N/A                | N/A        |
| Oh et al.\(^12\)          | 1               | M/30            | No     | R    | No       | 10               | C+BG      | 18                 | No         |
| Nydick et al.\(^13\)      | 1               | M/11            | Yes    | L    | Yes      | N/A              | C+BG      | 10                 | No         |
| Reddy and Sathi\(^14\)    | 1               | F/27            | No     | L    | No       | 9                | C+BG+CW   | 48                 | Yes        |
| Balke et al.\(^15\)       | 1               | M/56            | No     | L    | Yes      | 2                | C+BC      | 17                 | No         |
| Hsaio et al.\(^16\)       | 1               | M/24            | No     | L    | No       | 6                | C+BG      | 12                 | No         |
| Traoré et al.\(^17\)      | 1               | F/28            | No     | N/A  | No       | 12               | C+BG      | 31                 | No         |
| Arrouda et al.\(^18\)     | 1               | M/9             | No     | R    | No       | 3                | C+BG      | 10                 | No         |
| Plaikner et al.\(^19\)    | 1               | M/23            | Yes    | L    | Yes      | N/A              | C+BG      | 1.5                | No         |
| Sandokji\(^20\)           | 1               | M/43            | No     | R    | No       | 3                | arthroscopy C+BC | 60 | No |
| Sajoji et al.\(^21\)      | 1               | M/20            | No     | R    | No       | 4                | TP        | 48                 | No         |
| Henderson et al.\(^22\)   | 1               | M/33            | No     | L    | No       | N/A              | C+BG      | 24                 | No         |
| Çetinkaya et al.\(^23\)   | 1               | F/32            | No     | L    | No       | N/A              | TP        | 22                 | No         |
| Current study             | 1               | M/27            | No     | R    | No       | 12               | C+BG+CW   | 71                 | No         |

F, female; M, male; N/A, not available; PP, partial patellectomy; TP, total patellectomy; C, curettage; BG, bone grafting; BC, bone cement; CW, cerclage wiring.
to be overtreatment because in some patients, the articular cartilage surface below the patella was not affected.\textsuperscript{2,3,9} Currently, with a better understanding of primary ABC of the patella and reports of successful clinical outcomes, an increasing number of patients are being treated with curettage and bone grafting. Although the arthroscopic approach appears to be a less-invasive procedure, there is the potential for implantation of tumor cells into the articular cavity.

Based on Campanacci et al.,\textsuperscript{24} when dealing with active or aggressive cysts, the curettage should not be limited to simply opening a wedge in the cortex, and it is necessary to remove a good area of the wall and saucerize the cavity. This viewpoint is consistent with that of Dorman et al.\textsuperscript{25} Therefore, treatment selection depends on the size of the cyst and involvement of the articular surface. If the lesion is small, curettage with bone graft would suffice. However, for a larger cyst, curettage and bone grafting through a wide cortical window may be advised. If the lesion is associated with obvious articular disruption, partial or total patellectomy may be considered to avoid local recurrence. Similar to the case reported by Reddy and Sathi\textsuperscript{14} with Stage III ABC of the patella with cortical breakthrough that occurred because of improper treatment with curettage and autogenous bone grafting, the disease recurred. Additionally, because loss of the patella disturbs the biomechanics of the knee joint, which ultimately leads to early osteoarthritis,\textsuperscript{26} maintenance of all or part of the patella should be attempted first in young patients without significant articular damage if they accept the risk that a recurrence will require a total patellectomy.

Because of cortical thinning with an anterior crack that was limited to the periosteum and the articular cartilage was still intact, our patient had an “active” (stage II) large cyst. Consequently, intralesional curettage with bone grafting through a wide cortical window was performed. Cerclage and tension band wiring were then performed to protect the anterior tension of the patella to prevent patellar fracture. There are several advantages of the technique. First, the patient does not need to wear a cylinder cast postoperatively, which is conducive to early rehabilitation because of the rigid internal fixation. Second, a second surgery to remove the hardware is avoided. Finally, local recurrence seems to be reduced by a thorough curettage through a wide cortical window. Recurrence has been reported in other locations except if a wide resection is performed.\textsuperscript{27} Therefore, because this technique was used, our patient regained normal knee joint function without pain, and the bone graft remained well-incorporated without evidence of recurrence at a longest follow-up of 71 months. This was the second longest follow-up duration that has been reported, behind the study by Linscheid and Dahlin.\textsuperscript{4}

In conclusion, thorough curettage and bone grafting through a wide cortical window followed by cerclage wiring fixation and figure-eight sutures is a successful treatment option for primary ABC of the patella without articular disruption.

Declaration of conflicting interest
The authors declare that there is no conflict of interest.

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ORCID iD
Zhenguo Qiao \(\text{https://orcid.org/0000-0002-9079-956X}\)
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