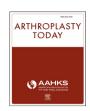
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Arthroplasty in Patients with Rare Conditions

Total Hip Arthroplasty in a Patient With a Large Proximal Femur Aneurysmal Bone Cyst: A Case Report and Literature Review

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ABSTRACT

Aneurysmal bone cysts (ABCs) are rare benign bone lesions with a predilection for the metaphysis of long bones. They are often cystic, expansive, and osteolytic and may result in bony deformity. In general, there remains debate about optimal treatment for ABCs; however, the mainstay typically consists of a combination of curettage, bone grafting, and considering the need for internal fixation and osteotomies. The goals of treatment include preserving bony anatomy while eliminating the lesion. There is sparse literature regarding the treatment of osteoarthritis adjacent to benign bony tumors. If total hip arthroplasty is chosen as a treatment option, diligent preoperative planning is required, and the surgeon must assess the patient's bone stock, account for bony deformity, and utilize specific implants and techniques based on the patient's characteristics. We present a case of an adult patient with proximal femur ABC and symptomatic adjacent hip osteoarthritis who underwent treatment with total hip arthroplasty.

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Introduction

Aneurysmal bone cysts (ABCs) are rare benign bone lesions occurring in 0.14-0.32 per 100,000 individuals per year [1,2]. They include cystic cavities most often in the metaphysis of long bones and are osteolytic, expansive, and hemorrhagic with an etiology that remains unknown [2]. While these lesions are typically encountered in children and adolescents, they may go unrecognized or untreated as patients transition through adulthood [2]. Goals of treatment include eliminating the lesion while preserving as much normal bone as possible; however, optimal treatment for ABCs remains debatable.

Osteoarthritis of the hip adjacent to benign tumors of the proximal femur introduces many complexities. The clinician must first decipher if a patient's symptoms are related to the patient's tumor or their degenerative joint. If arthroplasty is being considered as a treatment option, one must also evaluate the available bone stock, prosthesis design, and hip deformity.

There is limited literature regarding the treatment of osteoarthritis adjacent to benign tumors with most of the research focused

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on fibrous dysplasia. There is even less literature available with respect to treating hip osteoarthritis adjacent to proximal femur ABCs. We present a case of an adult patient with proximal femur ABC and symptomatic adjacent hip osteoarthritis who has failed nonoperative management. To our knowledge, this is the first case report of its kind in the adult population.

Case history

The patient is a 58-year-old female who presented to our orthopaedic clinic for right hip pain. Four years prior to her presentation, she began developing right groin pain, exacerbated by activity and radiation to her knee. She had a known history of right proximal femur bony cyst, which had been stable for over 20 years and was followed radiographically by her primary care physician. In our clinic, an anterior-posterior radiograph of the pelvis demonstrated a large expansive, cystic, and osteolytic lesion of the right proximal femur with end-stage degenerative joint disease of the hip (Fig. 1). She underwent a biopsy as well as a magnetic resonance imaging, which confirmed the diagnosis of ABC. The patient underwent an intra-articular hip injection, resulting in a significant reduction of her right groin pain and hip symptoms, confirming that her symptoms were secondary to osteoarthritis of the hip and not from her ABC. After failing nonoperative management with medications, physical therapy, and lifestyle modifications, the

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Figure 1. AP radiograph of the pelvis demonstrating a large expansive, cystic, and osteolytic lesion of the right proximal femur with end-stage degenerative joint disease of the hip. AP, anterior-posterior.

decision was made to proceed with right total hip arthroplasty (THA). The preoperative plan included curettage of the lesion and placement of a long diaphyseal cemented stem with utilization of the cement to fill the large void about the proximal femur.

The posterior approach to the hip was utilized. About 200 cc of cystic fluid was evacuated from the proximal femur once the femoral neck cut was made and the femoral head was removed revealing a large cystic lesion (Fig. 2). The proximal femur was

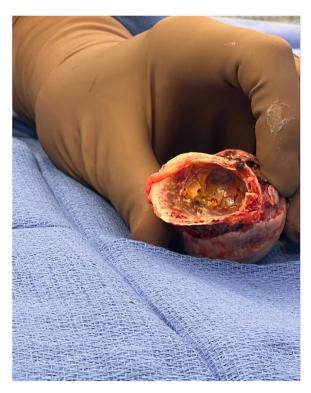


Figure 2. Clinical photograph of the resected femoral head and neck revealing in a large cystic lesion.

thoroughly debrided with aggressive curettage. The tissue was biopsied and sent for pathology. The acetabular portion of the procedure was carried out in standard fashion. The femur was broached until appropriate sizing was achieved. The trials were removed, and the femur was copiously irrigated. The canal was then prepped for cementing using the third-generation cement technique. A cement restrictor was placed, and the cement was pressurized using a cement gun. The stem was placed at the planned height and held in position with the appropriate version while the cement hardened. Once the cement had hardened, the appropriate head was placed and the hip was reduced. The patient had a stable range of motion, and equal leg lengths were restored. A chlorhexidine gluconate solution soak was performed, followed by normal saline irrigation. The piriformis and short external rotators were repaired. One gram of vancomycin powder was placed in the wound, and the soft tissues were closed in layers in standard fashion. Pathology of the intraoperative biopsy again confirmed the diagnosis of ABC. Postoperative radiographs are demonstrated in Figure 3. The technical difficulty of this procedure is noted as the cemented stem is in slight varus alignment and ideal cement interdigitation was unable to be achieved due to the smooth cyst walls. We plan to have continued regular follow-up with this patient, given that she is at a higher risk for early aseptic loosening.

The patient did well following the procedure, and her postoperative course was uncomplicated. At the 1-month follow-up, she was doing well, only requiring assistive device use on uneven ground. At 3 months, the patient was nearing her regular activity level with no functional hip pain. At her most recent 6-month follow-up, the patient had no complaints regarding her operative hip and was back to her normal activities, pain-free. Stable radiographs from the patient's 6-month follow-up are demonstrated in Figures 4 and 5.

Discussion

ABCs are rare benign bone lesions. They most commonly occur in the second decade of life; however, many ABCs are discovered incidentally on imaging obtained for other reasons or following trauma [3]. Radiographs typically show an expansive osteolytic lesion about the metaphysis of long bones with no invasion of the cartilage, and symptoms may include pain, swelling, or fracture [1,2]. These lesions have poorly organized structures with vascular gaps and fibrous membranes [3]. The cells within ABCs cause lacunar resorption and osteoclast-like actions [3]. In addition to primary ABCs, they may also be associated with another lesion and denoted as secondary ABCs. Secondary lesions account for 30% of ABCs and may be a reaction to other lesions including giant cell tumors, chondroblastomas, osteoblastomas, chondromyxoid fibromas, fibrous dysplasia, or nonossifying fibromas [2]. The etiology of these rare benign bony lesions remains unknown [1,2]. The natural history of these lesions consists of 4 stages defined radiographically: initial, active, stabilization, and healing. The initial phase consists of a well-defined lesion. The lesion grows rapidly and can progress to bone destruction during the active phase. Next, there is typically a period of stabilization with maturation of the bony shell, and lastly, the healing phase with progressive ossification of the lesion [4].

With respect to imaging, magnetic resonance imaging is more sensitive than computed tomography and will often show lobules or septa with multiple fluid levels on T2-weighted sequences [2]. Computed tomography scans can be more helpful in defining the lesion and the cortices. In addition to the tumors that can be seen with secondary ABCs, the differential diagnosis also includes simple or unicameral bone cysts and telangiectatic osteosarcoma [2]. An



Figure 3. Postoperative AP radiograph of the right hip demonstrating total hip arthroplasty components with a long diaphyseal cemented stem and filling of the large proximal femur lesion. AP, anterior-posterior; PACU, Post-Anesthesia Care Unit.

accurate diagnosis is important to ensure appropriate management; a biopsy is required for a definitive diagnosis [5].

ABCs are benign and may resolve spontaneously; however, they may also become more aggressive and evolve to cause osteolysis and cortical destruction [2]. The goals of treatment include eliminating the lesion while maintaining as much normal bone as possible. The optimal treatment for ABC's is debatable. Despite the number of techniques reported in the literature, there remains a recurrence rate that ranges from 5% to greater than 40%. Currently, curettage and filling the cavity with bone cement or bone substitute is the primary treatment modality used [4]. While many forms of treatment exist, wide surgical resection is the only definite cure. Radiation therapy can be effective but implements the risk of malignant transformation. Isolated embolization can be used, most often in the spine or sacrum. Intracystic injections of demineralized bone powder, bone marrow, calcitonin, or doxycycline have had conflicting results. Intracystic injections of alcohol have been effective, but may require repeated treatments. Cryotherapy has been found to reduce ABC recurrence [1]. Sclerotherapy has been shown to have a similar rate of recurrence compared to curettage; however, given its noninvasive nature, better functional outcomes and fewer complications have been seen [1]. Large defects may be difficult to treat allograft and vascularized bone grafts with internal fixation [6]. Wide resection will prevent local recurrence; however, marginal resection may be a good option for expansile lesions. Simple curettage and filling with graft, cement, or bone substitute



Figure 4. AP right demonstrating stable total hip arthroplasty components without evidence of failure. AP, anterior-posterior.

has shown good results when the tumor leaves a stable bony wall; however, recurrence can still be high from 11%-31% [3]. Langston et al. describe a case of staged THA in a patient with hip dysplasia and an adjacent proximal femur cyst where the patient underwent



Figure 5. Lateral right hip radiographs demonstrating stable total hip arthroplasty components without evidence of failure.

curettage and bone grafting 1 year prior to THA [7]. They had success with this, and bone stock was able to be preserved. They were then able to use a primary tapered press-fit femoral stem once the bone graft had grown in. This process was considered for our patient as well; however, we worried about the high risk of complications given the expansile nature of her lesion and the lack of integrity of her cyst walls.

With respect to standard treatment of ABCs, reconstruction with hip arthroplasty is not common and often reserved for aggressive tumors in the pediatric population; however, there is sparse literature on patients undergoing THA for osteoarthritis adjacent to the proximal femur ABC.

Fibrous dysplasia also commonly occurs in the proximal femur and is a developmental anomaly of bone formation that accounts for around 7% of benign bone tumors [8]. It results from an abnormality in lamellar bone formation and can lead to proximal femur deformity, the classic "shepherd's crook," or coxa vara deformity [8,9]. This is secondary to bone weakness and the large forces acting across the proximal femur. The resulting deviation of the mechanical axis of the hip in turn may lead to the development of adjacent hip osteoarthritis, and a significant number of these patients may require a THA in their lifetime [9,10]. Being aware of the patient's proximal femur deformity is important for surgical planning with respect to femoral implants and considering osteotomies to address and prevent varus remodeling of the femur.

Current controversies and future considerations

Similar to fibrous dysplasia, bone weakness is also seen in ABCs, and the deformity in these patients may vary [11,12]. THA in patients with underlying fibrous dysplasia of the proximal femur has been successful and is well described in the literature. Sierra et al. discuss how historically long-term fixation of the femoral component has been of concern in these patients and how better outcomes have been seen with cemented vs cementless femoral implants [8]. If cemented implants are chosen, meticulous technique should be utilized with care to obtain appropriate alignment and a satisfactory cement mantle [12]. On the contrary, Garceau et al. demonstrated low complication and revision rates with cementless components and routine use of cortical strut and proximal femoral allografts [10]. They also report that with modern conical fluted stems providing better diaphyseal fixation, improved results may be possible with enhanced cementless technology [10]. In our case, we decided to utilize a long calcar to replace the cemented femoral stem for increased stability, to bypass the lesion, and to provide an extra modality of treatment in hopes of combating recurrence. A calcar-replacement stem was chosen for additional calcar support over a traditional taper-slip long stem as we felt the expansile nature of the ABC had eroded too much of the patient's calcar. When utilizing this stem, one may consider using a burr to clear any sclerotic bone as well as the use of intraoperative radiographs to ensure appropriate stem placement. In the future, literature regarding long-term implant survivorship and local recurrence would be very helpful in determining ideal management for these patients.

Summary

Osteoarthritis of the hip adjacent to an ABC is rare, and literature regarding this topic is scarce. When pursuing THA as a treatment option, many complexities must be considered. Careful preoperative planning and selecting appropriate surgical treatment are essential for successful patient outcomes.

Key Points

- The accurate diagnosis of proximal femur lesions and the identification of the true source of hip symptoms are essential for the appropriate management of these patients.
- Treatment of patients with proximal femur lesions, or ABCs, and adjacent hip osteoarthritis should be individualized based on the patient's needs and anatomy.
- When considering total hip arthroplasty techniques in these patients, a variety of implants and allograft materials should be available and considered during preoperative planning.

Conflicts of interest

The authors declare there are no conflicts of interest. For full disclosure statements refer to https://doi.org/10.1016/j.artd.2024.101392.

Informed patient consent

The author(s) confirm that written informed consent has been obtained from the involved patient(s) or if appropriate from the parent, guardian, power of attorney of the involved patient(s); and, they have given approval for this information to be published in this case report (series).

CRediT authorship contribution statement

Jacob Shermetaro: Writing — review & editing, Writing — original draft. **Lawrence Jajou:** Writing — review & editing, Writing — original draft. **Aaron Seidman:** Writing — review & editing, Supervision, Conceptualization. **Daniel McCall:** Writing — review & editing, Supervision, Conceptualization.

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