Clinical presentation of an adolescent female synchronized swimmer with a simple bone cyst in the proximal humerus: a case report

Mitchell Savic, BHSc (Hons), DC, FRCCSS(C)1
Alexander Lee, BSc (Hons), DC, FRCCSS(C)2
Ashley Hook, HBSc, DC, DACBR, MSc2

Objective: This case raises awareness for healthcare practitioners who may not suspect a simple bone cyst (SBC) in a pediatric athlete with a high risk of pathological fracture.

Case summary: SBC’s are often considered as asymptomatic lesions which are commonly found incidentally on plain film radiographs. Presented here is a case of a 14-year-old competitive synchronized swimmer with a SBC in the proximal humerus with no MRI evidence of soft tissue pathology or pathological fracture, presenting clinically with refractory posterior shoulder pain.

Summary: Early detection of a SBC in a pediatric athlete is essential so an interdisciplinary care approach can be employed to ensure the appropriate management in those where there is high risk of pathological fracture. Clinicians should be aware of the risk factors for the...
Introduction
A simple bone cyst (SBC), also referred to as a unicameral bone cyst, is the most common benign tumour that is seen in children and adolescents (up to 85% of all cases).  These bone cysts represent approximately 3% of all tumours of the bone and have been reported to occur more frequently in boys than girls (2:1).  The average age of diagnosis is nine years, with a peak incidence between the ages of three and 14 years.  They are cystic, fluid filled cavities that can develop in tubular and flat bones, most typically found in the proximal humerus and femur, which account for greater than 90% of all cases.  These cysts do have a tendency to expand and weaken the bone they reside in, despite not being a true neoplasm.

A definite pathogenesis of a SBC to this day remains controversial. The most widely accepted etiology is a venous outflow obstruction in the cancellous bone causing an increase in intraosseous pressure. This increased pressure results in elevation of inflammatory protein markers released by endothelial cells along the wall of the cyst, causing subsequent osteoclastic activation and bone resorption. Other suggested mechanisms of pathogenesis include: a disturbance in local bone growth, intramedullary (post-traumatic) hemorrhage, and trapped intraosseous nests of synovial cells.

Although SBC’s are common and often considered asymptomatic, it has been reported that approximately two thirds of these lesions will at some point endure pathological fracture. This should be of particular concern in the pediatric athletic population due to the increased load and torque forces that may be placed through the humerus. This suggests that even in a non-contact scenario, the humerus may be at risk for fracture in the presence of a cyst. In sports where body contact with others is more probable, this risk of fracture is only expected to increase.

This raises further concern in such sports particularly if early detection has not been made.

We present an atypical clinical presentation of an adolescent competitive synchronized swimmer who experienced refractory shoulder pain despite a lack of MRI evidence of soft tissue pathology or pathological fracture of a SBC in the proximal humerus. We will also highlight possible complications and risk factors associated with SBC to raise awareness of a condition that may possibly go undiagnosed in young athletes.

Case presentation
A 14-year-old female, competitive synchronized swimmer presented with a four day history of localized, right-sided, posterior shoulder pain sustained while performing a manoeuvre referred to as sculling. Sculling is an action in synchronized swimming, whereby the swimmer treads water solely with their arms, while inverted as they are performing out of water movements with their legs. This action involves heavily resisted and repetitive internal and external rotation of the shoulders.

Upon presentation, the patient complained of a dull ache in the posterior shoulder at rest, which became sharp when she performed sculling. The patient did not report any previous shoulder injuries in her past. On examination, there was full, pain-free right shoulder range of motion (ROM) with no signs of swelling, erythema, or bony malalignment. Empty can, Hornblower’s test and manual muscle testing of external rotation strength at zero degrees shoulder abduction were positive for pain and were rated 4/5 on the right for muscle strength. Palpation of the infraspinatus and teres minor insertions at the posterior aspect of the humeral head reproduced the patient’s chief complaint. An evaluation of generalized joint hypermobility (GJH) using the Beighton Test revealed a
score of 3/9 (+ bilateral hyperextension of 5th metacarpophalangeal joint >90° & + palm of hands touching flat on the ground when knees extended).

An assessment of shoulder instability using the Feagin Test, Load and Shift Test or sulcus sign was not made. Cervical spine range of motion was full and pain free, and foraminal encroachment tests of the cervical spine were negative. Sensation and motor testing of the C5-T1 levels were within normal limits bilaterally and deep tendon reflexes of C5-7 were 2+ bilaterally.

The patient was given a clinical diagnosis of an acute, right-sided rotator cuff tendinopathy and a six-week conservative plan of management consisting of Active Release Therapy (ART®) and a progressive rotator cuff and scapular stabilizer strengthening rehab protocol was implemented. The patient was treated with this approach for 9 visits over 6 weeks. Following this treatment plan, the patient showed minimal clinical change in her symptomatology and was unable to perform sculling due to pain.

A radiograph of the right shoulder was ordered (Figure 2) which revealed a well-defined, osteolytic lesion. Differential diagnoses based off the radiographic findings included: a SBC, aneurysmal bone cyst (ABC), and a chondroid lesion, such as an enchondroma. The patient was...
immediately referred to an orthopedic surgeon with a specialty in bone tumors of the upper extremity, who sent the patient for a magnetic resonance imaging (MRI) of the right shoulder (Figure 3) which demonstrated a slightly expansile, T2 hyper-intense and T1 hypo-intense intramedullary unilocular cystic lesion in the proximal humeral metaphysis causing endosteal scalloping and cortical thinning with no evidence of fracture. All rotator-cuff and long head of biceps tendons appeared to be intact with no sign of pathology. An orthopedic surgeon confirmed the diagnosis of a SBC based off the aforementioned MRI features which are pathognomonic for this lesion, along with the consideration of the patient’s age which is also consistent with its clinical presentation. One possible inconsistency with the clinical presentation of this suspected SBC is that they are not typically painful.

At this point in time (six to eight weeks post initial presentation), the patient was still experiencing intermittent, refractory dull, achy pain along the posterior shoulder, despite her diligence with pain-free rehabilitation exercises, and avoiding all aggravating factors including synchronized swimming. The orthopedic surgeon handling the patient’s case prescribed a conservative “wait and see” approach which simply involved a follow-up MRI, activity restriction, and to continue rehab for the rotator cuff and scapular stabilizers at a recommended physiotherapy clinic.

The patient was contacted a year later, reporting that she had stopped participating in synchronized swimming. She has no pain at rest or during any of her activities of daily living. By this point, the patient decided to not follow-up with any radiographs and/or MRI, and as such, the status of the SBC is currently unknown. Any further follow-up with the patient was lost as she no longer attends the clinic where the assessment and treatments were conducted.

**Discussion**

The diagnosis of a SBC can normally be made with the use of plain film radiographs, however, confirmation can be made with advanced imaging (such as MRI) in cases where there is a strong resemblance to other benign bony lesions. An example of such a lesion, and a strong differential diagnosis is an ABC, which is at higher risk of gross expansion and pathological fracture.1 Although generally not as expansile as an ABC, SBC’s have the capability to expand and thin the cortex that it surrounds. This may compromise the cortex possibly resulting in a pathological fracture, which is not uncommon in the athletic population.10 SBC’s also reside centrally within the medullary cavity, unlike ABC’s which have a more eccentric presentation. The more central the location, the smaller the likelihood of fracture risk and soft tissue involvement.2 Ten percent of those that fracture will present with a fallen fragment sign on plain film radiographs, which represents a small portion of the fractured bone which can separate and sink to the bottom of the cyst.2,11

With that said, it is important to understand risk factors which may further increase fracture risk. A retrospective study by Urakawa *et al.* identified 155 subjects with a SBC, 141 of which exceeded six-month follow-up. The majority of subjects (35%) had a SBC which resided in the proximal humerus, followed by five other locations in the lower extremities. In this study, a multivariate analysis revealed that ballooning of the bone, a cyst located in a long bone, male sex, and multilocular cysts were signifi-
cant risk factors in increasing the incidence of pathological fractures. Kaelin and MacEwan proposed a cyst index formula (cyst area divided by the diaphyseal diameter) and cut-off score to help determine fracture risk. A cyst index greater than 3.38 was predictive of pathological fracture in this particular study. Similar studies have reported other risk factors associated with increased incidence of pathological fracture, which include: a high cyst index, high percentage of bone occupied by the cyst in the transverse plane, thin cortical thickness, and those active phase cysts residing in the upper limb.

Furthermore, active phase cysts are a potential risk factor for fracture. It has generally been accepted throughout the literature that cysts residing within 1cm of the growth plate are considered active, minimizing the amount of normal bone between them. Haidar et al. underwent a retrospective analysis whereby a measurement of two centimeters from the growth plate was determined as the cut-off point between active versus latent cysts. The destined progression in the natural history of the cyst is to migrate down into the diaphysis through endochondral ossification where it eventually becomes latent (non-active). These cysts are not commonly seen in adults, owing to the fact that they spontaneously fill and ossify on their own sometime after skeletal maturity is reached. Since SBC’s are typically asymptomatic in the absence of pathological fracture, there is no inquiry for imaging and thus go undetected during pre-adulthood prior to ossification, making natural history difficult to document.

Analysis of the present patient’s SBC on MRI (Figure 2) reveals many of the risk factors mentioned above. The cyst’s location is in a long bone, it is showing properties of expansion (ballooning), a high percentage of the bone is occupied by the cyst in the transverse plane, there is presence of thinning cortex, and it clearly appears to be in an active phase, as it’s shown abutting the epiphyseal growth plate. Furthermore, using the equation proposed by the Kaelin and MacEwan, the patient had a cyst index well above the cut-off score (4.5cm × 3.6cm = 16.2/1.32cm² = 9.30). The only fracture risk factors that do not appear to exist in the present patient’s case are the fact that the cyst is not multilocular in nature, and she is not of male sex. It was also described in the study by Urakawa et al. that males in the study were more likely to be physically active, which may be the main contributor to the increased incidence of fracture within this cohort. The presence of multiple risk factors suggest that the present patient is of high risk of developing a pathological fracture.

As for management, Urakawa et al. suggested that high risk cysts should either be considered for surgery, or at the very least, careful observation. Due to the fact that SBC’s are self-limiting and have a tendency to heal without any intervention, it is common to adopt a more conservative plan of management, especially in children who have asymptomatic, latent cysts. Educating and restricting activity in those athletes with a SBC who are at high risk of fracture should be of utmost importance in those who are following this conservative approach. In a recent survey of members of the European Pediatric Orthopedic Society (EPOS) and Pediatric Orthopedic Society of North America (POSNA) surgery was highly recommended only in those children with symptomatic SBC’s under 10 to 12 years of age (by 71% of members) and in those children with asymptomatic, high risk of fracture SBC’s (by 53% of members). The most commonly reported surgical options used include: curettage with or without bone grafting, percutaneous decompression, corticosteroid injections, or autologous bone marrow injections. It is important to note that many of these interventions have high rates of recurrence, with improved outcomes seen with cysts measuring >2cm from the growth plate.

As a clinician working with pediatric athletes, not only is it important to detect the presence of a bone cyst, or any other bony lesions, it is also important to understand the possibility or risk of other complications that may arise. In the presence of a pathological fracture, one such potential complication is the development of avascular necrosis (AVN) of the humeral head. Urakawa et al. reported one subject with a femoral neck SBC who developed AVN after a displaced fracture. There are other reports in the literature that have described similar instances of AVN of the femoral head as a complication after pathological fracture. Brooks et al. specify that AVN of the humeral head may result after either displaced fractures, or fracture dislocations. Humeral head AVN has not been reported in SBC cases, likely due to the fact that, to the authors’ knowledge, there presently is no report of displaced humeral SBC pathological fractures in the literature.

Another possible complication is growth arrest, especially in those active cysts which are likely communicating with the epiphyseal growth plate. Although extremely
Clinical presentation of an adolescent female synchronized swimmer with a simple bone cyst in the proximal humerus: a case report

As a treating practitioner, early detection of SBCs is central to their management. When identified early, decisions relating to activity restriction (or elimination), and early referral to an orthopedic surgeon can be undertaken. This will help to not only decrease the likelihood of developing a pathological fracture, but will also allow for the development of a plan of management that will be more conducive to treating structures around a weakened bone. That being said, practitioners must be cognizant when treating soft tissue structures surrounding the cyst, in order to help decrease the risk of iatrogenic fracture which could plausibly be caused by manual therapy.

Summary
This case highlights the importance of early detection of a SBC in a pediatric synchronized swimmer. She presented with symptoms consistent with rotator cuff pathology, but was determined to have a SBC that was classified as a potential high risk for pathological fracture. As illustrated
by this case, it is important to determine the risk factors for the development of a pathological fracture, and for other potential complications that may arise thereafter in those with a SBC.

References


