Intraosseous ganglion cyst of the humeral head in a competitive flat water paddler: case report

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Objective: To present the diagnostic and clinical features of an intraosseous ganglion cyst of the humeral head of a female flat water canoe athlete.

Clinical Features: An 18-year old female flat water canoeist complaining of right shoulder pain following a strenuous paddling training camp.

Intervention and outcome: A trial of passive care was conducted, including soft tissue therapy, spinal manipulative therapy, acupuncture, and rehabilitation. The patient seemed to be responding with treatment, but pain would always resume with paddling. A diagnostic ultrasound displayed mild thickening and effusion in the subacromial/subdeltoid bursae. Continued passive care was not able to resolve the symptoms and she underwent an MRI which revealed an intraosseus ganglion cyst subjacent to the lesser tuberosity and floor of the intertubercular groove. A subsequent MRA was ordered to assess the labrum, which was intact, but the cyst had progressed in size. She was referred to an orthopedic surgeon who performed surgery.

Conclusion: An IOG cyst within the humeral head is a rare, potentially painful condition that can mimic other pathologies including impingement and labral tear. It is important to be aware of the clinical features to obtain Objectif : soumettre un diagnostic et les caractéristiques cliniques d'un kyste ganglionnaire intraosseux de la tête humérale d'une athlète pratiquant le canoë en eau plate.

Caractéristiques cliniques : une canoéiste en eau plate de 18 ans se plaint de douleurs à l'épaule droite suite à un camp d'entraînement très exigeant.

Intervention et résultat : un essai de soins passifs fut mené, notamment la thérapie des parties molles, la manipulation rachidienne, l'acupuncture et la réhabilitation. La patiente semble avoir bien réagi au traitement, mais la douleur revient lorsqu'elle recommence à ramer. Un ultrason diagnostic démontra un épaississement léger et une effusion dans les bourses sous-acromiales/des courts rotateurs de l'épaule. Les soins passifs continus n'ont pu éliminer les symptômes, et elle subit un examen à résonnance magnétique qui révéla un kyste ganglionnaire intraosseux sous-jacent à la tubérosité moindre et *au plancher de la cannelure intertuberculaire. Une* angiographie à résonnance magnétique subséquente fut exigée pour évaluer le bourrelet marginal, qui était intact, mais le kyste avait grossi. Un chirurgien orthopédique dut l'opérer.

Conclusion : un kyste ganglionnaire intraosseux dans la tête humérale est un état pathologique douloureux plutôt rare, qui ressemble à d'autres états pathologiques, notamment la déchirure du bourrelet marginal et le coincement. Il est important de connaître les caractéristiques cliniques afin de diagnostiquer

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a prompt diagnosis and appropriate treatment of this condition. (JCCA 2011; 55(4):294–301)

KEY WORDS: intraosseous cyst, ganglion cyst, paddler, flat water, humeral head, canoe

Introduction

Canoeing and kayaking are two sports that have their origins in North America.¹ Native Americans and Canadians and Inuit developed and used the canoe and kayak for trading, transportation and fishing. The canoe and kayak played a huge role in the history and transformation of Canada into a country when the Voyageurs adapted their use and procured new trade opportunities in the New World.² In a manner similar to the use of horses and horseracing, the mechanization of society diminished the role of the canoe and kayak but their traditions live on through competition.

In competition, canoeing and kayaking are categorized into "whitewater" and "flat water"; whitewater referring to paddling on a moving body of water and flat water a non-moving body of water. Men's flat water canoeing and kayaking debuted in the 1936 Berlin Olympics while women's flat water kayaking was added in 1948.²

The American Canoe Association estimates that there are approximately 24 million Americans involved in canoeing and kayaking.² Another study, done in 2004, showed similar results, indicating that participation had increased from 16.7 million in 1994–95 to 22.6 million in 1999.³ Of the 22.6 million participants, the study indicated 28% were women.³

Injury data for these two sports is difficult to ascertain due to a lack of published studies. In a survey done in 2000 on whitewater canoe and kayak injuries, the estimated incidence of any type of injury was 4.5 injuries per 1000 days paddled with an incidence of significant injuries (an injury for which medical evaluation/care was sought) at 1.9 injuries per 1000 days paddled.³ They also found a median average for days paddled was 50 days per year with a range of 5 to 300 days.³ A similar survey for flat-water paddlers could not be found and makes any extrapolation to this type of paddling difficult. Precipitating factors in paddling injuries include intense, highvolume on-water training, less-than-optimal paddling rapidement cet état pathologique et de prescrire un traitement adéquat. (JCCA 2011; 55(4):294–301)

MOTS CLÉS : kyste intraosseux, kyste ganglionnaire, canoéiste, eau plate, tête humérale, canoë

stroke mechanics and maximum strength training during dry-land workouts.⁴

Intraosseous ganglion (IOG) cysts of the humerus is a rare and often painful condition.^{5–9} There is only one other case in the literature describing an IOG cyst of the humerus, while most papers describe cystic lesions of the carpal and tarsal bones.^{6,10,11} Symptomatic IOG cysts present with intermittent pain, occasional swelling and it is aggravated via movement of the affected area.^{6,12} IOG cysts are often misdiagnosed or there is a delay in diagnosis due to the variable presentation.^{6–8,11–13} Early diagnosis is important to avoid mismanagement of patients.

Another case of a ganglion cyst affecting the shoulder in a canoeist was reported in a 15 year old female.¹⁴ The athlete complained of pain, reduced abduction and external rotation of her left shoulder with intermittent parasthesia over the left scapula. An EMG and MRI revealed an entrapment neuropraxia of the left suprascapular nerve due to a ganglion cyst. The authors felt the repetitive nature of the paddling stroke caused an otherwise asymptomatic, pre-existing ganglion cyst to become symptomatic.

The purpose of this case report is to: present a rare case of intraosseus ganglion cyst (IOG) of the humerus diagnosed in a flat water canoe athlete; outline the difficulties in its diagnosis due to an IOGs ability to mimic other pathologies; outline the sports' biomechanics predisposing the athlete to other, more likely, pathologies (impingement syndromes, rotator cuff tendinopathy ...); the imaging needed to diagnose an IOG; and the subsequent surgical management of the patient.

Case Presentation

An 18 year old female national level flat water canoe athlete was assessed for a right anterior shoulder and upper back complaint that started following an intensive paddling training camp three weeks prior. She paddles on the left side with her right being the high hand on the paddle. The pain was progressing and not improving. She reported a previous, similar right shoulder complaint two years prior that had resolved within three weeks with chiropractic care. She also complained of some numbness and tingling into the posterolateral arm that she had not had previously.

On examination, her cervical spine range of motion and neurologic exam were within normal limits. During orthopedic evaluation of the shoulder, both empty and full can were found to be positive, while crank test elicited pain and clicking. Passive joint play of the glenohumeral joint demonstrated mild to moderate laxity. Joint play of the cervical and thoracic spine revealed locally tender restrictions. An upper limb tension test of the radial nerve recreated the numbness and tingling in the arm, doorbell test was locally tender and Hoffman's test was negative.

The patient was tentatively diagnosed with a rotator cuff tendinopathy and radial nerve entrapment (posterior interosseous nerve syndrome), with differential diagnoses of labral tear, supraspinatus tear, and cervical spine radiculopathy. Treatment of this patient included soft tissue therapy (myofascial release) of the affected right shoulder musculature and radial nerve, spinal manipulative therapy of the cervical and thoracic spine, rehabilitative exercises and a progressive return to paddling. Her MD had prescribed Celebrex but she was unsure if it was helping. She was also referred for a diagnostic ultrasound of the right shoulder.

The diagnostic ultrasound performed within 1 week of referral (4 weeks post-injury) suggested that all rotator cuff musculature was intact and no effusion was visualized in any of the tendons. However, mild thickening and effusion was noted in the subacromial/subdeltoid bursae.

Her shoulder responded moderately well to treatment but would re-aggravate following a paddling session longer than 10 or 15 minutes. Upon further evaluation by the primary author, she had tenderness of the long thoracic, dorsal scapular, and suprascapular nerves with palpation and tension of the nerve with palpation increased the pain. (Author's Note: In the primary author's experience, these peripheral nerve findings are consistent with other nerve entrapments that involve predominantly the C5 nerve root.) She also had pain in the shoulder with shoulder flexion and abduction. Treatment of the entrapment sites of these nerves using myofascial and vibrationassisted myofascial release and acupuncture was added to her plan of management as well as acupuncture to the arm and shoulder including Li4, 11, 12, 15, TW14, Lu5, GB20 and 21. Nerve flossing exercises of the affected nerves were added to her treatment program as well. With the addition of these therapies, the posterolateral arm tingling resolved and her pain free shoulder range of motion increased.

Paddling continued to aggravate her condition and following a particularly painful training session several SLAP lesion tests were now positive including: O'Brien's, Biceps Load 2 and anterior slide. She was recommended to get an MRA, ideally to assess the labrum, or an MRI and reduce the paddling as much as possible.

An MRI performed 5 months post-injury revealed an 8 mm \times 6 mm intraosseus ganglion cyst subjacent to the lesser tuberosity and floor of the intertubercular groove (see Figure 1). She was told by the medical doctor that the cyst was not the reason for her pain. Due to its close proximity to the biceps tendon in the intertubercular groove, it was suggested that the cyst may be causing a tendinopathy and she received soft tissue treatment and rehabilitation. All other structures in the shoulder were intact and no other effusion was noted in the shoulder or bursal complex upon MRI evaluation.

The patient's shoulder complaint was not improving and she was referred to an orthopedic clinic at the university she was attending. The patient was prescribed physical therapy that mainly consisted of exercises which re-aggravated her condition. She was subsequently referred for an MRA. An MRA performed 8 months postinjury showed that the labrum was intact but the cyst had progressed to a size of 12 mm (see Figure 2). It also showed a partial thickness tear of the bursal surface of the supraspinatus tendon, and fraying of the subscapularis tendon. She was referred to an orthopedic surgeon who performed surgery 9 months post injury.

The orthopaedic surgeon assessed the shoulder arthroscopically and reported an intact rotator cuff with slight fraying of the subscapularis that was felt to be minimal and required no intervention. A small incision was made in the anterior shoulder, the cyst site was identified due to a small hole noticed in the bone deep to bicep tendon. The area was drilled and a curette was used to remove any material but little was extracted. The site was then injected with a small amount of demineralized bone matrix. The surgeon's prognosis in terms of recovery time was

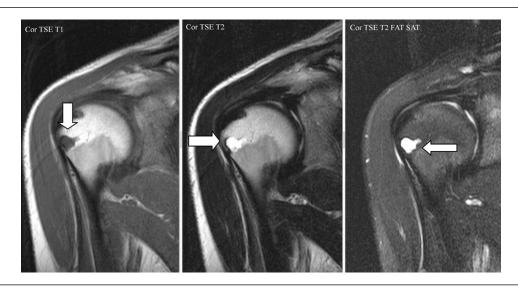
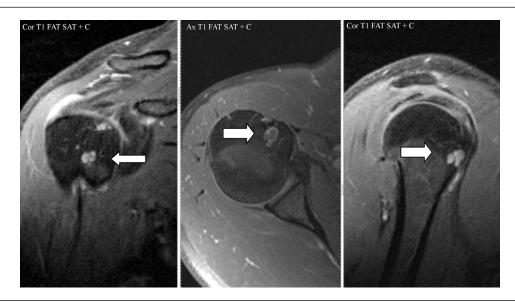


Figure 1 MRI revealing intraosseous ganglion cyst. The white arrows indicates the IOG cyst.

Figure 2 MRA revealing the intraosseous ganglion cyst. The white arrows indicates the IOG cyst.



unknown – he had not seen a similar case previously. The patient has returned twice for follow-up with the surgeon who feels she is making good progress.

Physical therapy was initiated approximately 2 weeks after surgery that included range of motion exercises, postural/scapular setting, and kinesiotaping to aid proper positioning. The program was progressed to light strengthening and lower body cardiovascular exercise. At 14 months post injury (5 months post surgery), the patient reported full range of motion and that she was progressing through her weight training program.

She also reported a return to paddling one time per week for 1 hour sessions with no return of the original pain.

Discussion

This is the second case known to the authors to describe IOG cysts of the humeral head and the first in a flat water paddler. The discussion will outline the reasons why the shoulder is an oft injured area, including the biomechanics of the canoe stroke (see Tables 1), as well as IOG cysts in bones. Because IOG cysts in the humeral head are very rare, the IOG cyst discussion will primarily be based on information in bones more commonly involved.

Shoulder injuries in paddlers are very common.^{4,15,16} Berglund and McKenzie¹⁶ suggest that the most common types of shoulder injuries in paddlers are impingement syndrome, bicipital tendonitis, and glenohumeral dislocations/subluxations although there were no statistics or references related to the prevalence of these conditions. They report that impingement syndrome is a constant source of lost training or missed competition, however, degeneration and subsequent tearing of the rotator cuff is very rare in the paddling athlete.

A condition known as "paddler's shoulder" is a soft tissue injury of the shoulder that includes shoulder impingement syndrome, bicipital tendonitis and subacromial bursitis.⁴ Predisposing factors for paddler's shoulder include maintaining a high (upper) pivot arm in the canoe paddling stroke as well as hypertrophy of the musculotendinous structures of the rotator cuff that fills the subacromial space.⁴ Interestingly, a comparison of canoe athletes between the 1976 and 2000 Olympics showed that both male and female athletes were 5 kg heavier although having similar skinfold measurements, and had increased shoulder breadth and chest girth indicating hypertrophic changes in these and other muscle groups.¹⁷ These changes were attributed to "advances" in off and on water training programs. Although suggestive of a predisposing factor,⁴ no injury data for comparison was available.

Our patient was initially diagnosed with a rotator cuff tendinopathy secondary to shoulder impingement and this differential remained high on the list even after the MRI showed an intraosseus ganglion cyst. It was not until the surgery and the lack of tendon damage being found that rotator cuff tendinopathy could be seemingly ruled out.

Ganglion cysts most commonly exist in soft tissue, but there are rare occurrences where they have been diagnosed within the bone termed as an intraosseous ganglion cyst.⁵ IOG cysts present as solitary well-defined, sharply marginated lytic lesions that are commonly found within the subchondral region and epiphyseal areas of long and short tubular bones.^{6,7,8} They come in close proximity to the joint but rarely perforate the joint cavity or articular surface.^{6,7,8} These cysts form near joints that do not undergo significant degenerative changes.⁷ They most often occur in the lower end of the tibia, medial malleolus, femoral head, and carpals.^{6,7,9} However, they rarely affect the neck of the scapula, head of the humerus, ulnar head and acetabulum.^{6,7,9}

The exact pathogenesis of ganglion cysts is still unclear but a few theories exist. One theory suggests that ganglia are due to intramedullary metaplasia of mesenchymal cells into synovial type cells as an idiopathic process in response to an unknown stimulus.^{7,8,12} The second theory describes penetration of an extra-osseous synovial cyst which begins to erode into the bone and eventually becomes isolated within the bone by new bone formation filling the intracortical gap.^{6,7,8,11,13} The last theory proposes a microvascular disorder that leads to the development of cysts after focal ischemic bone necrosis.⁸

The clinical presentation of IOG cysts is quite variable and nonspecific, which can impact obtaining prompt diagnosis.^{7,13} In most cases there is intermittent mild localized pain, aggravated with activity of the affected area.⁶ There may or may not be associated swelling and soft tissue masses depending on the location of the cyst.^{6,13} Pain may be present for many months due to a delay in diagnosis¹¹ which can vary from two months to three years.^{6,7,8,10,11,18} In the case presented, the patient was told by the MD that the pain was not due to the IOG cyst, which delayed adequate treatment. Interestingly during the surgery, the surgeon noted that there was a small hole in the bone leading into the cyst suggesting that there was communication between the biceps tendon and the ganglia. If the cyst was enlarging during and after activity, and its contents were pushing through the communicating channel, the biceps tendon may have been forced anteriorly potentially increasing the risk of impingement.

Diagnosis of IOG cysts is achieved via radiographic or special imaging. Upon x-ray analysis, IOG cysts appear as a single or multiple confluent radiolucent, eccentric defects measuring 1–2 millimeters surrounded by a narrow zone of sclerosis.^{6,7,12,18} The lesion, however, usually does not extend deep into the bone.^{6,10} CT scans display details of spatial orientation of the lesions, confirms the cysts liquid aspect and is helpful in detecting any cortical defect

 Table 1
 Canoe biomechanics^{1,4,19}

Canoe stroke position	Anterior view	Lateral view
Set up The canoeing stroke involves the athlete kneeling on one knee, with the contralateral hip and knee flexed to 90 degrees. The bottom arm is held at approximately 100 degrees of shoulder flexion, with the hand positioned just above the blade while the top arm is at 170 degrees of shoulder flexion, with the hand at the top of the paddle (A-frame position). The stroke side hip and torso are rotated forward to allow the bottom arm to reach as far forward as possible.	Note the Neer impingment position.	
Catch The torso flexes forward to allow the blade to enter the water. The top and bottom arms drive downward, with the blade entering the water at approximately 60 degrees.		
Draw or Pull The pulling motion is initiated by the stroke side hip rotating back away from the bottom hand. The elbows stay extended throughout the pull phase. The body then rotates posteriorly, keeping both elbows extended. Both shoulders will move into extension, with the top shoulder adducting and crossing the midline of the body.		

Table 1 (Concluded)

Exit As the blade nears the hip, the bottom arm will flex at the elbow to allow the blade to exit the water.		
Recovery The top arm quickly flexes upward as the body rotates towards the front of the boat to place the blade back in the water. The forward rotation is initiated by the stroke side hip rotating forward as the blade exits the water.	Return to Set up position	Return to Set up position

or articular communication.¹⁰ MRI scans exhibit a welldefined fluid collection with low intensity of T1-weighted images and very high intensity on T2-weighted images.¹¹ In our present case, the initial MRI clearly shows the IOG on the T2 weighted images (see figure 1) and the MRA (see figure 2).

Differential diagnoses of IOG cysts include: aneurysmal bone cyst, osteoblastoma, giant cell tumour, fibrous dysplasia, chondromyxoid fibroma, osteiod osteoma, rheumatoid arthritis, simple bone cyst, enchondroma, chondroblastoma, and subchondaral bone cyst.^{6,12} Many of these differentials can be ruled out based on patient age, symptomatolgy, and location. IOG cysts are commonly confused with subchondral cysts, which communicate with the joint, while IOG cysts rarely do so and are not associated with a degenerative a process.^{6,7,8}

Conservative management is always considered the first step to treatment. There are a limited number of reports outlining the conservative treatment of IOG. Conservative care typically consists of NSAIDs and splinting of the affected area.¹⁸ In our present case, the patient received physical therapy, chiropractic care (16 visits), acupuncture and appropriate activity modification. This

regimen was moderately successful in reducing the pain when the patient was not paddling but the pain quickly returned following a session on the water. It was due to this unusual recurrence pattern, atypical for a normal soft tissue/overuse injury, that further investigation was initiated.

If conservative therapy fails, surgery is an option. There are several indications to guide operative intervention. These include: i) failure of conservative modalities to provide adequate relief of symptoms, ii) suspicious x-ray changes, iii) the cyst progressively entering and replacing the cancellous substance of the bone eroding the cortex. Even if the lesion is not painful, it weakens the bone putting it at risk for fracture.^{12,18} The surgical procedure with the lowest recurrence rate is known as curettage^{6,18} which involves excision of the cyst followed by bone grafting to prevent recurrence and the risk of collapsing fracture.^{12,18}

Conclusion

The biomechanics of the canoe stroke and the hypertrophy of the musculotendinous contents of the subacromial space seemingly predispose the paddling athlete to shoulder overuse/impingement injuries.⁴ Combine these factors with an acute episode of extreme training,⁴ as in our case, and this can lead to a shoulder injury. Whether the extreme training caused the intraosseus ganglion cyst to become symptomatic is debatable but has been suggested previously¹⁴ although the mechanism is unclear.

IOG cyst of the humeral head is a rare entity that may result in a great deal of pain. It is important for clinicians to be aware that cysts not only exist in soft tissues but also within the bone. Patients may present with enduring pain lasting anywhere between two months to three years and delays in diagnosis can lead to inappropriate management and delays in proper treatment.

References

- 1 Shephard RJ. Science and medicine of canoeing and kayaking. Sports Medicine. 1987; 4: 19–33
- 2 Kenal K, Trela P. Canoeing and Kayaking. In: Drinkwater, BL, editor. Women in Sport. Oxford UK: Blackwell Science LTD; 2008. p.600
- 3 Schoen RG, Stano MJ. Year 2000 Whitewater Injury Survey. Wilderness Environ Med. 2002; 13(2):119–24.
- 4 Pelham TW, Holt LE, Stalker RE. The etiology of paddler's shoulder. Australian J Science Med Sport. 1995; 27(2):43–7.
- 5 Fealy MJ, Lineaweaver W. Intraossrous ganglion cyst of the scaphoid. Ann Plastic Surgery. 1995; 34(2): 215–217.
- 6 Kambolis C, Bullough PG, Jaffe HL. Ganglionic cystic defects of bone. J Bone Joint Surg. 1973; 55: 496–505.
- 7 Pope TL, Fechner RE, Keats TE. Intra-osseous ganglion. Report of four cases and review of the literature. Skeletal Radiology. 1989; 18: 185–187.
- 8 Schrank C, Meirer R, Stabler A, Nerlich A et al. Morphology and topography of intraosseous ganglion cysts in the carpus: an anatomic, histopathologic, and Magnetic Resonance Imaging correlation study. J Hand Surg. 2003; 28A:52–61.

- 9 Tuzner T, Subasi M, Alper M et al. Penetrating type intraosseous ganglion cyst of the lunate bone. West Indian Med J. 2005; 56(6):384–386.
- 10 Mnif H, Koubaa M, Zrig M et al. Ganglion cyst of the carpal navicular. a case report and review of the literature. Orthopaedics & Traumatology: Surgery and Research. 2010; 96:190–193.
- 11 Nishimura T, Tsujii, Kusuzaki, Hoki Y et al. Intra-osseous ganglion of the proximal humerus: case report. J Ortho Surg. 2007; 15(1):102–105.
- 12 Uriburu, Levy VD. Scaphoid and lunate bones: report of 15 cases in 13 patients. J Hand Surg. 1999; 24A:508–515.
- 13 Tan EW, Dharamsi FM, McCarthy EF et al. Intramuscualr synovial cyst of the shoulder: case report. J Shoulder Elbow Surg. 2010; 19:e20–e24.
- 14 Knossalla F, Nicolas V, Tegenthoff M. Suprascapular nerve entrapment in a canoeist. Arch Neurol. 2006; 63(5):781.
- 15 Kameyama O, Shibano K, Kawakita H et al. Medical check of competitive canoeists. J Ortho Science. 1999; 4(4):243–9.
- 16 Berglund B, McKenzie D. Injuries in canoeing and kayaking. In: Renstron, editor. P.A.F.H. Clinical Practice of Sports Injury, Prevention, and Care. Encyclopaedia of Sports Medicine. Oxford; Blackwell Scientific Publications; 1994. P.633–640
- 17 Ackland TR, Ong KB, Kerr DA, et al. Morphological characteristics of Olympic sprint canoe and kayak paddlers. J Science Med Sport. 2003; 6(3):285–94.
- 18 Kural C, Sungur I, Cetinus E. Bilateral lunate intraosseous ganglia: case report. Orthopedics. 2010; 33(7):514.
- 19 Canoe Kayak Canada [Internet]. Ottawa; c2009–2011 [updated 2002 Sep; cited 2011 Apr 11]. CKC Canoe Technical Page. Available from: http://www.canoekayak. ca/files/49/72/Canoe_TECHNICAL_TEMPLATEwebsite_ version.pdf