

Solitary Bone Cyst of the Proximal Humerus with a Concomitant Stiff Shoulder Treated with both Arthroscopic Capsular Release and Arthroscope-Assisted Resection of the Bone Cyst: A Case Report

Akira Ando^{1*}, Masashi Koide¹, Yoshihiro Hagiwara², Michimasa Matsuda¹, Eiji Itoi²

¹Department of Orthopaedic Surgery, Matsuda Hospital, Sendai, Japan

²Department of Orthopaedic Surgery, Tohoku University School of Medicine, Sendai, Japan

Email: *andoakir@yahoo.co.jp

How to cite this paper: Ando, A., Koide, M., Hagiwara, Y., Matsuda, M. and Itoi, E. (2018) Solitary Bone Cyst of the Proximal Humerus with a Concomitant Stiff Shoulder Treated with both Arthroscopic Capsular Release and Arthroscope-Assisted Resection of the Bone Cyst: A Case Report. *Open Journal of Orthopedics*, 8, 77-84. <https://doi.org/10.4236/ojo.2018.83010>

Received: January 20, 2018

Accepted: March 2, 2018

Published: March 5, 2018

Copyright © 2018 by authors and Scientific Research Publishing Inc. This work is licensed under the Creative Commons Attribution International License (CC BY 4.0). <http://creativecommons.org/licenses/by/4.0/>



Open Access

Abstract

Solitary bone cysts are benign bone lesions that usually occur in the humerus, calcaneus, and femur of children and adolescents. When present in adults, they are usually diagnosed as incidental findings. In this report, we present an adult case of a solitary bone cyst of the proximal humerus with concomitant refractory stiff shoulder treated with both arthroscopic capsular release and arthroscope-assisted resection of the bone cyst. A 73-year-old woman was referred with a complaint of persistent left shoulder pain and limited range of motion. Magnetic resonance imaging showed an approximately 4.5-cm long diameter cystic bone lesion from the humeral neck to the shaft. At first, arthroscopic synovectomy and pan-capsular release were performed. Next, two 1-cm skin incisions and 8-mm drill holes into the lateral wall of the cyst were created with fluoroscopic guidance. After insertion of the arthroscope through the holes, the white membranous cyst wall was resected with a shaver until bone marrow was observed. Finally, an 8-mm cannulated hydroxyapatite pin was inserted. This is a simple technique that allows direct visualization and complete resection of the cyst wall with two small incisions. Such a technique may be superior to conventional open procedures or those under image guidance.

Keywords

Solitary Bone Cyst, Humerus, Adult, Arthroscope-Assisted Resection, Hydroxyapatite Pin

1. Introduction

Solitary or unicameral bone cysts are benign bone lesions in children and adolescents that are commonly observed in the humerus as well as the femur and the calcaneus [1]. Most patients present with pain due to pathologic or impending fractures or they can be found incidentally [2]. Solitary bone cysts in adults are rare, but are occasionally seen in elderly persons [3]. Treatment options for solitary bone cysts include careful observation, open curettage and bone grafting, steroid or autologous bone marrow injections, various methods of cyst decompression using cannulated screws or pins, and replacement of the cyst cavity using a bone substitute, such as calcium phosphate cement [4] [5] [6]. The treatment of solitary bone cysts has evolved from open curettage and bone grafting to less invasive surgery. The recent development of arthroscopic technology has allowed operators to perform curettage of the cyst wall safely and with greater precision under direct visualization [2] [7] [8] [9] [10] [11]. Using such a minimally invasive surgery for the calcaneus bone is well reported [2] [7] [9] [10], but has rarely been reported for use in treating the humerus [11]. We present an adult case of a solitary bone cyst of the proximal humerus with concomitant refractory stiff shoulder treated with arthroscopic capsular release and arthroscopy-assisted resection of the bone cyst.

2. Case Report

A 73-year-old woman was referred to our clinic with a complaint of persistent left shoulder pain and a limited range of motion despite 3 months of conservative treatment. She had been treated with nonsteroidal anti-inflammatory medication and injection of hyaluronan, but the pain continued to worsen. From her examination, both active and passive shoulder motions were restricted at 120° in flexion, 90° in abduction, 20° in external rotation, and at the 5th lumbar vertebra in internal rotation. Tenderness of the rotator interval was observed, but not in the lateral aspect of the humerus. There was no history of previous shoulder pain or trauma. There was no abnormality in the plain radiographs (**Figure 1**), but magnetic resonance imaging showed an approximately 4.5-cm long diameter cystic bone lesion without a septum from the humeral neck to the shaft with T1 low and T2 high signal intensities (**Figure 2**). Effusion of the glenohumeral joint and thickening of the rotator interval were also observed. Thinning of the cortex was not apparent by computed tomography (**Figure 3**). Laboratory findings were all in the normal range, including a white blood cell count (5400/ μ l), erythrocyte sedimentation rate (11 mm/hour), and C-reactive protein (0.11 mg/dl). Culture of the effusion of the glenohumeral joint was negative. Steroid injection into the glenohumeral joint was effective, but limited.

Diagnosis at that time was a refractory stiff shoulder with an incidentally observed solitary bone cyst of the proximal humerus. Surgery was performed in a beach-chair position under general anesthesia. Passive shoulder motion was also restricted under general anesthesia. At first, arthroscopic synovectomy and

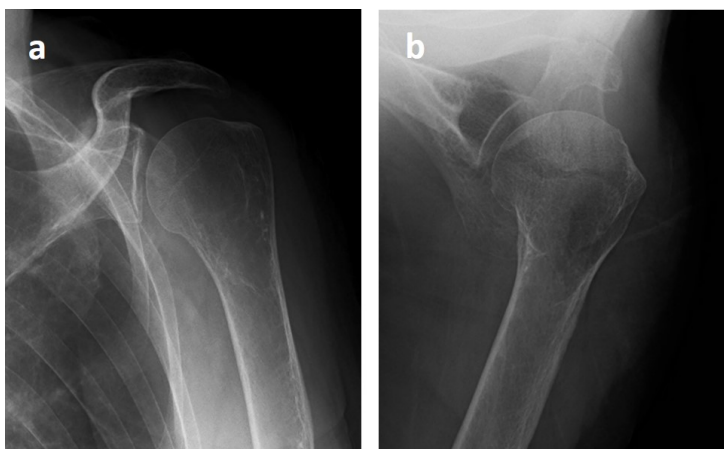


Figure 1. Plain anteroposterior (a) and axial (b) radiographs of the left shoulder showing no apparent bone lesions.

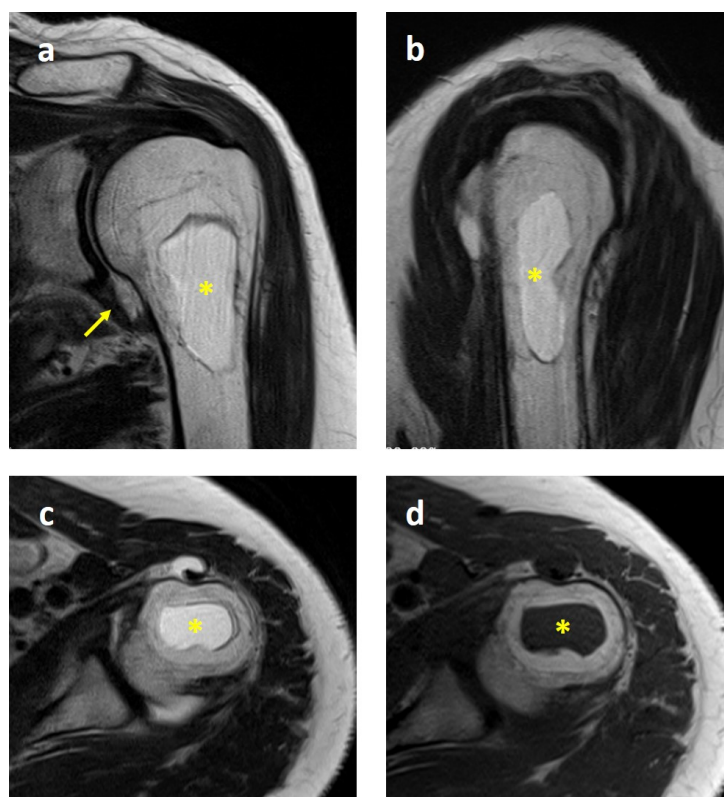


Figure 2. Magnetic resonance imaging of the T2 coronal (a), sagittal (b), axial (c), and T1 axial (d) planes of the left shoulder, thereby indicating a cystic bone lesion without a septum from the humeral neck to the shaft (asterisk). Effusion of the glenohumeral joint was also observed (arrow).

pan-capsular release were performed using anterior and posterior portals. Active synovitis and stiffness of the capsule were observed (**Figure 4(a)**). After the procedure, normal range of motion was obtained. In the next step, two 1-cm skin incisions and 8-mm drill holes into the lateral wall of the cyst with fluoroscopic guidance were created with meticulous care not to damage the axillary nerve

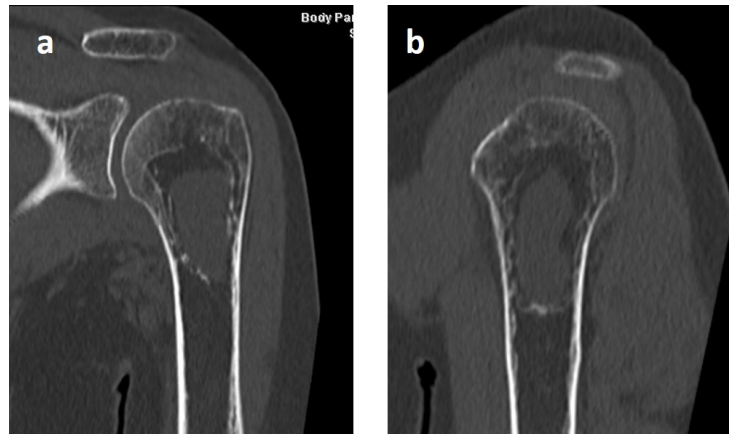


Figure 3. Computed tomography of the coronal (a) and sagittal (b) planes showing a bone lesion without thinning of the cortex.

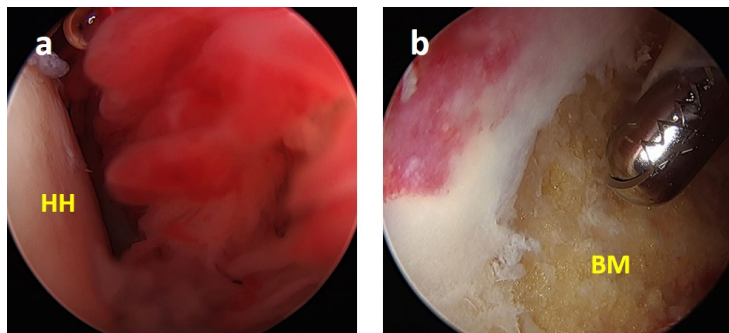


Figure 4. Posterior viewing of the glenohumeral joint showing active synovitis of the rotator interval (a). Viewing from the portal created to the lateral wall of the bone cyst (b). Membranous cyst wall was resected with a shaver until bone marrow was observed. HH, humeral head; BM, bone marrow.

(**Figure 5**). After insertion of the arthroscope through the holes, the white membranous cyst wall was observed and resected with a shaver until bone marrow was observed (**Figure 4(b)**). An arthroscope was introduced to the other drill hole, resection was similarly performed, and sufficient curettage of the inner surface of the cyst was confirmed endoscopically. Finally, an 8-mm cannulated hydroxyapatite pin (Japan Medical Next, Osaka, Japan) was inserted for continuous cannulation (**Figure 6(a)**). Histologic analysis showed a fibrous tissue membrane with occasional giant cells, which was coincident with a finding of the solitary bone cyst. After surgery, no remarkable complications were observed, and passive shoulder exercise was allowed the following day. At 1-year after the operation, pain was relieved, and motion was improved to 170° in flexion and abduction, 60° in external rotation, and to the 10th thoracic vertebra in internal rotation. Ossification of the cyst was insufficient, but enlargement of the cyst was not observed (**Figures 6(b)-(d)**). Written informed consent was obtained from the patient for publication of this case report and accompanying images.

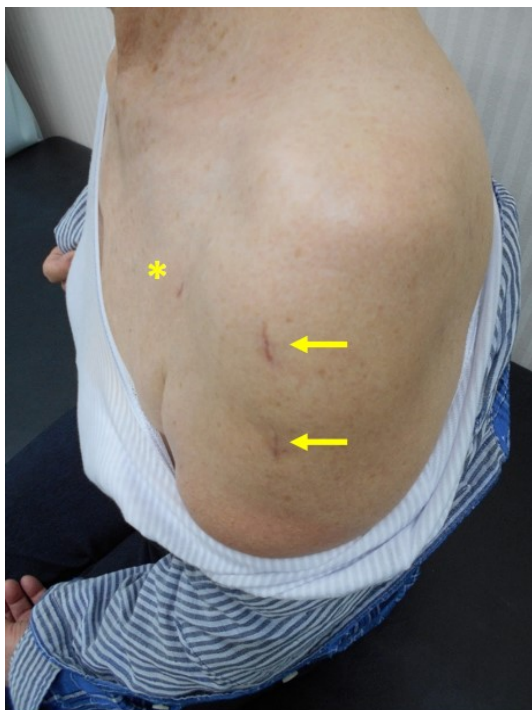


Figure 5. Macroscopic picture of the lateral aspect of the shoulder showing two portals created for cyst resection (arrows). Asterisk indicates an anterior portal.

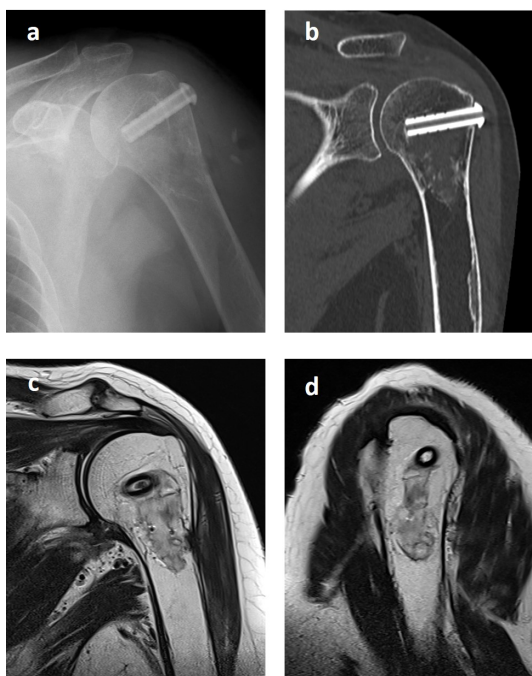


Figure 6. Postoperative anteroposterior plain radiograph showing an inserted cannulated hydroxyapatite pin (a). Computed tomography (b) and magnetic resonance imaging ((c), (d)) at one year after the operation indicating insufficient ossification without enlargement of the bone cyst.

3. Discussion

Solitary bone cysts are benign bone lesions that usually occur in the humerus, calcaneus, and femur of children or adolescents. When present in adults, they are usually diagnosed as incidental findings [1]. The bone cyst observed in this case was found incidentally in relation to the concomitant stiff shoulder. It was successfully treated with arthroscope-assisted resection of the bone cyst and insertion of the hydroxyapatite pin for continuous cannulation. This simple technique allowed direct visualization and complete resection of the cyst wall with two small incisions. Such a technique may be superior to conventional open procedures or those under image guidance.

Treatment strategies for the solitary bone cyst are changing from conventional open curettage and autologous bone grafting to minimally invasive surgeries to decrease the rate of recurrence and to avoid disadvantages, such as donor-site morbidity, increased operative time, and intraoperative blood loss [9]. Dormans *et al.* described successful results of a minimally invasive technique that consists of percutaneous intramedullary decompression, curettage, and calcium sulfate pellets grafting for the solitary bone cysts of variable bones under image intensifier [4]. Alvarez *et al.* first reported a case of a calcaneal bone cyst treated with curettage and bone grafting under arthroscope assistance [7]. Thereafter, several authors reported successful cases of a calcaneal bone cyst with use of an arthroscope [2] [9] [10]. Meanwhile, there are few reports of a bone cyst on the humerus [11] [12]. Otsuka *et al.* reported a case series of arthroscope-assisted resection of the cyst of variable bones, which included a case involving the humerus; however, detailed descriptions about the surgical technique are lacking [11]. Saccomanni reported an atypical case of aneurysmal bone cyst of the humeral head treated with arthroscope-assisted resection of the bone cyst [12]. To our knowledge, this is the first case of treatment of a solitary bone cyst of the humerus that includes a detailed surgical technique under arthroscope assistance.

Optimal treatment after arthroscope-assisted resection of the cyst wall is still unclear. Recent reports have shown the efficacy of continuous decompression using a cannulated screw [5] [6] [13] [14]. This technique may be reasonable because a main hypothesized cause of a solitary bone cyst is accumulation of interstitial fluid within the bone due to venous obstruction [1] [13]. Shirai *et al.* reported good clinical results with this procedure, of which 38 out of 43 cases achieved bone healing [6]. In this case, we used a cannulated hydroxyapatite pin for continuous decompression of the cyst. Such a pin does not require removal and has osteoconductive properties that may stimulate bone healing [6]. However, there has been no evidence regarding the time required to achieve sufficient structural strength using this technique. In this case, ossification of the cyst was insufficient at one year after the operation. As another, less-invasive technique, injection of bone substitute, such as calcium phosphate cement, has been performed [2] [4] [9] [10]. Mainard *et al.* first reported a case of a solitary bone cyst on the calcaneus that was treated using arthroscope-assisted resection of the

cyst and injection of calcium phosphate cement with a successful outcome [9]. Innami *et al.* reported superior results using a similar technique in 16 young athletes with symptomatic calcaneal bone cysts [2]. They found that optimal visualization, which allows complete curettage with minimum violation of the structural integrity, and percutaneous injection of bone substitute instead of autologous bone grafting permit an early return to sports activities in young athletes. Notably, while the maximum strength of the cement is obtained within 3 days, the process of integration and eventual degradation requires several years [9] [10]. Patient age, activity, and cyst site (*i.e.* upper or lower extremities) should be considered for suitable treatment.

4. Conclusion

We present an adult case of solitary bone cyst of the proximal humerus successfully treated with arthroscopy-assisted resection of the bone cyst and insertion of the hydroxyapatite pin for continuous cannulation.

Acknowledgements

We would like to thank Editage (<http://www.editage.jp/>) for English language editing.

Conflicts of Interest

No conflicts declared.

References

- [1] Pretell-Mazzini, J., Murphy, R.F., Kushare, I. and Dormans, J.P. (2014) Unicameral Bone Cysts: General Characteristics and Management Controversies. *Journal of the American Academy of Orthopaedic Surgeons*, **22**, 295-303. <https://doi.org/10.5435/JAAOS-22-05-295>
- [2] Innami, K., Takao, M., Miyamoto, W., Abe, S., Nishi, H. and Matsushita, T. (2011) Endoscopic Surgery for Young Athletes with Symptomatic Unicameral Bone Cyst of the Calcaneus. *The American Journal of Sports Medicine*, **39**, 575-581. <https://doi.org/10.1177/0363546510388932>
- [3] Aycan, O.E., Çamurcu, I.Y., Özer, D., Arıkan, Y. and Kabukçuoğlu, Y.S. (2015) Unusual Localizations of Unicameral Bone Cysts and Aneurysmal Bone Cyst: A Retrospective Review of 451 Cases. *Acta Orthopaedica Belgica*, **81**, 209-212. <https://www.ncbi.nlm.nih.gov/pubmed/26280957>
- [4] Dormans, J.P., Sankar, W.N., Moroz, L. and Erol, B. (2005) Percutaneous Intramedullary Decompression, Curettage, and Grafting with Medical-Grade Calcium Sulfate Pellets for Unicameral Bone Cysts in Children. *Journal of Pediatric Orthopedics*, **25**, 804-811. <https://www.ncbi.nlm.nih.gov/pubmed/16294140> <https://doi.org/10.1097/01.bpo.0000184647.03981.a5>
- [5] Hou, H.Y., Wu, K., Wang, C.T., Chang, S.M., Lin, W.H. and Yang, R.S. (2010) Treatment of Unicameral Bone Cyst: A Comparative Study of Selected Techniques. *The Journal of Bone and Joint Surgery, American Volume*, **92**, 855-862. <https://doi.org/10.2106/JBJS.I.00607>
- [6] Shirai, T., Tsuchiya, H., Terauchi, R., Tsuchida, S., Mizoshiri, N., Ikoma, K., Fuji-

- wara, H., Miwa, S., Kimura, H., Takeuchi, A., Hayashi, K., Yamamoto, N. and Kubo, T. (2015) Treatment of a Simple Bone Cyst Using a Cannulated Hydroxyapatite pin. *Medicine*, **94**, e1027. <https://doi.org/10.1097/MD.0000000000001027>
- [7] Alvarez, R.G. and Arnold, J.M. (2007) Technical Tip: Arthroscopic Assistance in Minimally Invasive Curettage and Bone Grafting of a Calcaneal Unicameral Bone Cyst. *Foot & Ankle International*, **28**, 1198-1199. <https://doi.org/10.3113/FAI.2007.1198>
- [8] Dietz, J.F., Kachar, S.M. and Nagle, D.J. (2007) Endoscopically Assisted Excision of Digital Enchondroma. *Arthroscopy*, **23**, 678.e1-678.e4. <https://doi.org/10.1016/j.arthro.2005.12.063>
- [9] Mainard, D. and Galois, L. (2006) Treatment of a Solitary Calcaneal Cyst with Endoscopic Curettage and Percutaneous Injection of Calcium Phosphate Cement. *Journal of Foot and Ankle Surgery*, **45**, 436-440. <https://doi.org/10.1053/j.jfas.2006.09.014>
- [10] Nishimura, A., Matsumine, A., Kato, K., Asanuma, K., Nakamura, T., Fukuda, A. and Sudo, A. (2016) Endoscopic versus Open Surgery for Calcaneal Bone Cysts: A Preliminary Report. *Journal of Foot and Ankle Surgery*, **55**, 782-787. <https://doi.org/10.1053/j.jfas.2016.03.006>
- [11] Otsuka, T., Kobayashi, M., Sekiya, I., Yonezawa, M., Kamiyama, F., Matsushita, Y. and Matsui, N. (2001) A New Treatment of Aneurysmal Bone Cyst by Endoscopic Curettage without Bone Grafting. *Arthroscopy*, **17**, E28. <https://www.ncbi.nlm.nih.gov/pubmed/11536108> <https://doi.org/10.1053/jars.2001.25334>
- [12] Saccomanni, B. (2008) An Atypical Aneurysmal Bone Cyst of the Head of the Humerus, Arthroscopic Treatment: A Case Report. *Archives of Orthopaedic and Trauma Surgery*, **128**, 1279-1282. <https://doi.org/10.1007/s00402-007-0503-8>
- [13] Morii, T., Mochizuki, K. and Satomi, K. (2009) Continuous Decompression Using a Cannulated Ceramic Pin for Simple Bone Cysts. *Journal of Orthopaedic Surgery*, **17**, 62-66. <https://doi.org/10.1177/230949900901700114>
- [14] Tsuchiya, H., Abdel-Wanis, M.E., Uehara, K. and Tomita, K. (2002) Cannulation of Simple Bone Cysts. *The Journal of Bone & Joint Surgery (British Volume)*, **84**, 245-248. <https://www.ncbi.nlm.nih.gov/pubmed/11922366> <https://doi.org/10.1302/0301-620X.84B2.12473>