Large Cavernous Hemangioma of the Subscapularis Muscle - A Case Report

Ki-Won Lee, Hyun Il Lee, Chung-Hwan Kim, Tae-Kyung Kim

Department of Orthopedic Surgery, Gangneung Asan Hospital, University of Ulsan College of Medicine, Gangneung, Korea

We report a case of intramuscular hemangioma in the subscapularis muscle and the resulting impairment of shoulder function in an adult patient. A nineteen-year-old female complained of shoulder pain and the development of a mass in the absence of previous trauma. Physical examinations, including lift-off and belly-press tests, showed abnormality. X-ray showed multiple calcifications in the front of the scapula. Magnetic resonance imaging showed a soft-tissue mass occupying almost the entire intramuscular portion of the subscapularis muscle. An arthroscopic examination excluded the possibility of a joint invasion, after which the entire mass was successfully removed by open excision. The displacement of the subscapularis by the mass was relieved after the surgery. Pathological diagnosis of the tissue confirmed a cavernous hemangioma. Both shoulder pain and function was improved after operation. There was no evidence of recurrence even at the 2-year follow-up. Rare forms of hemangioma adjacent to the shoulder joint could be successfully managed with surgical excision. Differential diagnosis, such as synovial chondromatosis, pigmented villo-nodular synovitis, and malignant sarcoma, should also be considered.

Key Words: Shoulder; Rotator cuff; Hemangioma

Intramuscular hemangioma comprises approximately just 1% of all cases of hemangioma, and it occurs more frequently in the lower extremities than in the upper body. It occurs very rarely in the adult shoulder joint and only occasional references of intramuscular hemangioma are reported in the field of radiology or pathology that focus primarily on diagnosis. Furthermore, report of an intramuscular hemangioma in the subscapularis, which is a muscle important for shoulder function, is extremely rare both nationally and internationally. The authors experienced a 19-year-old female patient with a giant intramuscular hemangioma in the subscapularis of the left shoulder joint. In this study, we report the surgical treatment of this case with a literature review.

Case Report

Medical History
A 19-year-old female patient visited the hospital complaining of pain and swelling in the left shoulder joint that began 8 months ago. On the visual analogue scale, the degree of pain was approximately 5 points. The patient experienced intermittent pain even at rest.

Physical Examination
In the affected left shoulder, the range of passive motion was normal and the range of active motion was restricted at 140 degrees in forward elevation, 20 degrees in external rotation, and to L4 in internal rotation, all of which were accompanied with pain at terminal range of joint motion. In the front of the shoulder joint, tenderness was elicited across a relatively broad
area, and a soft mass was detected by palpation, which had an undefined border and an approximate size of 7 cm by 10 cm. The belly-press test and lift-off test showed functional abnormalities in the subscapularis, including pain and muscle weakening. The Korean Shoulder Score (KSS) was 52 points before surgery.

**Radiologic Findings**

Simple radiography showed multiple small round calcifications in the front of the scapular neck, but no other abnormalities indicating the involvement of the bone (Fig. 1). The Magnetic resonance imaging (MRI) showed a soft-tissue mass between the subscapularis recess and the front of the scapular neck, occupying the intramuscular portion of the subscapularis and pushing the subscapularis forward (Fig. 2). The mass was approximately 5.1×5.6×5.5 cm large, with a relatively well-defined border, and comprised of several small nodules organized in a grape-like cluster. Both the T1 and T2-weighted coronal images showed a mass with a heterogeneous signal. The T1-weighted coronal image showed low-signal intensity mass and the signal was similar to the surrounding muscle area. The T2-weighed coronal image showed a mass with a signal intensity that was slightly higher than the surrounding muscles. Additionally, a signal void indicated by low signal intensity was observed in the small nodules of the grape-like cluster (Fig. 2A-C). A part of the tumor is suspected to infiltrate the glenohumeral joint, and contrast-enhancing image showed delayed enhancement of the peripheral structure and of the internal septum (Fig. 2D). Differential diagnosis among hemangioma, pigmented villo-nodular synovitis and

![Fig. 1. Simple radiograph of left shoulder showing multiple small round calcifications in the front of the scapular neck.](image)

![Fig. 2. Preoperative magnetic resonance imaging. (A) T1-weighted coronal image; relatively low-signal intensity mass with heterogeneous signal in subscapularis recess. (B) T2-weighted coronal image; the lesion is defined and lobulated with high-signal intensity. Calcification is demonstrated as foci of signal void (arrows). T2-weighted sagittal image with fat suppression showed high signal intensity in mass ruling-out fatty component (C), T1-weighted axial image with fat suppression after contrast-enhancing (D).](image)
synovial osteo-chondromatosis was required according to MRI.

**Surgery and Rehabilitation**

Arthroscopic examination and excisional biopsy were carried out on the glenohumeral joint on the grounds that the tumor was large, caused persistent pain and functional disability in the patient, and may be malignant. The patient was anesthetized and placed in a beach chair position. First, arthroscopy of the glenohumeral joint was performed to observe the intra-articular space, and showed that no tumor had infiltrated or protruded into the intra-articular space. Then, the lesion was approached by incising the upper 2/3 of the attachment site of the subscapularis into the lesser tubercle after a deltopectoral approach. Then, the subscapularis was pulled towards the front to expose the mass. The tumor was situated in the front of the subscapularis recess and was infiltrating into the intramuscular portion of the subscapularis. The tumor was located in the extracapsular space between the subscapularis and the capsule of glenohumeral joint, and pushing out the subscapularis forwards. The tumor was generally made of soft-tissue and a dense network of blood clot on top of which we observed multiple calcific material and evidence of hemorrhage (Fig. 3). After marginal excision of mass, meticulous hemostasis was achieved and the subscapularis was sutured into its original position. The patient wore an arm brace for 2 weeks after the surgery and began shoulder passive joint range of motion exercise at 2 days after the surgery. The patient was allowed to begin on supportive-active joint exercise at 2 weeks after surgery and on active exercise 4 weeks after this.

**Pathologic Findings**

The biopsy showed a cavernous space, vascular endothelial tissue surrounding this space, and an organized fibrinoid material that suggested recanalization of the blood capillary around the cavernous space (Fig. 4A). According to immune-histochemical tests, the sample was positive for the vascular endothelial cell-specific protein CD31 (Fig. 4B). Embedded mineral in the calcific material is turned out to dystrophic calcification suggesting typical phlebolith. The overall observation of the biopsy indi-

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**Fig. 3.** Gross appearance of mass from the excision shows calcified material and the main tumor. The main tumor shrank after blood was evacuated from the mass.

**Fig. 4.** (A) Histology slides shows cavernous vessel (black arrows), skeletal muscle bundle (arrow head), and adipose tissue (white arrow) (H&E, ×200). (B) Endothelial lining cells stained with CD31 antibody (black arrows) and organized small capillaries (white arrow) in immunohistochemistry (×200).

**Fig. 5.** Postoperative radiograph at 2 years after operation shows no abnormal findings.
icated this mass as a cavernous hemangioma.

Clinical Course after Surgery

One month after surgery, the pain in the shoulder disappeared and the range of active internal rotation was recovered. One year after surgery, clinical tests did not show tenderness or palpation of mass. The KSS was 94 points, the function of the subscapularis was normal as indicated by the near-to-normal lift-off and belly-press tests, and muscle strength was almost normal, although still weaker than the right. The most recent follow-up examination at 2.4 years (27 months) after surgery showed no evidence of recurrence via physical or X-ray examinations (Fig. 5).

Discussion

Hemangiomas are histologically classified into capillary hemangioma, cavernous hemangioma, arteriovenous hemangioma, venous hemangioma and mixed types. The type observed in this case report is cavernous hemangioma, of which typical histologic characteristics are hyperplasia of the blood vessel, abnormal fatty proliferation and the presence of phlebolith.1,4 Intramuscular hemangiomas are very rare and comprise just 1% of all hemangioma, therefore, their literatures reported in Korea are also rare.6 In the past, 24 case series of hemangiomas in the upper limbs have been reported in Korea, of which one case occurred in the deltoid muscle of the shoulders. However, neither the exact size of the tumor or its functional effects was reported.8 Tang et al.7 have reported an analysis of surgical treatments carried out in 89 cases of soft-tissue hemangioma. In the report, the average size of the tumor was 6 cm (0.8-12.0 cm), the average age of the patients was 30 years old (7-71 years old), the common locations of the tumor were the thighs (36%), the lower legs (17%), the forearms (12%), pain from pressure were seen in 58% of the patients, and phlebolith was observed in 15% of the patients by X-ray. The surgical treatments carried out were marginal or intra-lesional excision. Recurrence was observed in 19% of the patients, most of which were detected within 2 years of surgery. Phlebolith is the most common form of calcification, which occurs when the blood clot calcifies and is characteristic of hemangioma. It can be detected in 15-50% of hemangioma by simple radiography.7,8

Memis et al.5 reported that the accurate diagnosis of hemangioma by radiologic examination can be achieved by MRI scans. They also reported that that performing surgery after locating the exact boundary through MRI study helps to limit recurrence. Griffin et al.9 reported that an MRI scan for intra-muscular hemangioma had identified the precise boundary of the lesion, the lobulation structure. Adipose tissue and vascular channel were observed in 93% and 100% of cases, respectively. Also, in T1-weighted imaging, 70% of the cases had higher signal intensities than in the muscle, and in T2-weighted imaging, it was 96% of the cases. Altogether, the presence of fat, calcification, and internal vascular channels in MRI images are predictable factors towards the preference for diagnosis of hemangioma. In our study, we observed a mixture of signal intensities that indicated calcification and the presence of internal vascular channels, whilst those indicating adipose tissue were largely undetected.

There have been no reports of distant metastasis of the hemangioma, but it is known to be a locally aggressive tumor and so the possibility of its recurrence is ever-present.10 Aggressive hemangioma includes hemangioendothelioma, hemangiopericytoma, and angiosarcoma, which are suggested by the infiltration of the tumor into the bone, the co-morbidity of skin ulcers, rapid growth and so on. However, radiological examination alone is insufficient for diagnosis and a biopsy is for diagnosis strongly required.8

Although the close observation of hemangioma without surgery could be possible because this is a benign tumor, active surgical treatment is required in situation as where aggressiveness is suspected and radiological examination is unable to differentiate between benign and aggressive, there is pain from tumor or aesthetic need is large.1,5,7 In our case, an accurate histological diagnosis was required, and a surgical treatment was carried out due to the increasing size and pain from the tumor. If the tumor is large and therefore excessive bleeding is expected during the surgery, excision may be performed after artery embolic occlusion or the occlusion alone with regular follow-up could also be possible.7,9 Recurrence is related to the size of the tumor and an incomplete excision, so complete excision of the lesion during surgery is the most effective method of reducing the risk of recurrence.7,10

The authors report a case of a large cavernous hemangioma in the subscapularis muscle and the resulting impairment in the function of the shoulder joint. We include a literature review with the case report, in the hope to improve the differential diagnosis and the current treatment methods of hemangioma.

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