CASE REPORT AND REVIEW OF LITERATURE

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Intramuscular ganglionic cyst in the extensor digitorum longus muscle of the leg: A case report and review of literature

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ABSTRACT

Ganglionic cysts are benign soft-tissue lesions with unclear etiology and pathogenesis commonly occurring on the wrists and hands. Although their diagnosis and management in these characteristic locations are easy, rare occurrences in atypical locations are difficult to diagnose. Ultrasonography and magnetic resonance imaging (MRI) are helpful in diagnosing and differentiating a ganglionic cyst from other soft-tissue lesions. Here, we report a case of 60-year-old male patient with intramuscular ganglionic cyst, measuring 68 × 41 × 48 mm, arising in the extensor digitorum muscle of the left leg, which was diagnosed using MRI and treated using excision and biopsy.

Key words: Ganglion; Cyst; Extensor digitorum muscle; Intramuscular ganglionic cyst; Excision

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INTRODUCTION

Ganglionic cysts are common benign non-neoplastic soft-tissue masses. They have no true epithelial lining and are filled with gelatinous material composed mainly of hyaluronic acid. The etiology and pathogenesis of ganglionic cysts remain unknown. They may be secondary to degenerative changes and chronic damage that leads to liquefaction and cyst formation. This is followed by the formation and proliferation of a fibrotic and compact wall arising from the surrounding connective tissue.[1] The gelatinous material has been proposed to be produced by injured mesenchymal cells.[2]

Ganglionic cysts are more common in females and can occur at any age. They usually form over a joint, particularly the scapholunate joint of the wrist, and to a lesser extent from the tendon sheath. They are commonly seen in the hand, wrist, and foot, and most of them are found in the wrist on either the dorsal or volar side. Although ganglionic cysts formed in these locations can be easily diagnosed clinically, those arising at unusual anatomic locations present a diagnostic challenge. Ganglionic cysts have been reported to originate from cartilage, nerve, and muscle. Intra-articular ganglionic cysts in the knee involve the tendon sheath, joint capsule, menisci, or anterior and posterior cruciate ligaments. However, intramuscular ganglionic cysts have rarely been reported in the literature.[3-5] Ganglionic cysts have also been reported as intraosseous ganglions in the distal tibia and other bones as well as from peripheral nerve sheath, most commonly common

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peroneal nerve and less frequently radial, ulnar, median, and sciatic nerves.[6,7]

In most of cases, patients with ganglion cyst present with mild symptoms, such as swelling, which might sometimes cause mild pain and nerve compression. The etiology and pathogenesis of ganglion cysts are obscure and they are believed to be generated by mucoid degeneration of the joint capsule, tendon, or tendon sheath.[8] Ultrasonography and magnetic resonance imaging (MRI) are helpful in diagnosing and differentiating a ganglionic cyst from other soft-tissue lesions and provide excellent information on the location; however, the diagnosis needs to be confirmed by biopsy.[9]

Here, we report a case of multilobulated intramuscular ganglionic cyst within the extensor digitorum longus muscle of the left leg.

CASE REPORT

A 60-year-old male presented at our hospital with swelling in the lateral aspect of the proximal part of left leg in the past 6 months. He had no history of trauma or infection. The swelling was painless and did not cause any discomfort to the patient. He had a history of two surgical interventions but the swelling had recurred. On physical examination, a palpable cystic mass present over the lateral aspect of the proximal part of the left leg was noted. The swelling was non-tender and skin over the swelling was normal in color and texture, except for a scar mark of previous surgery over the swelling (Figure 1). No functional or neurological deficits were noticed.

The radiographic finding of the leg was normal. MRI revealed a multiloculated, fluid-filled large cystic mass lesion, measuring 68×41×48 mm, which was hypointense on T1WI and hyperintense on T2/STIR-weighted images, with its epicenter within the extensor digitorum longus muscle in the upper left leg region (Figure 2).

The lesion was abutting the proximal portion of the fibula posterior, extending anterolaterally and slight inferiorly and tracking into the overlying subcutaneous fat plane. On post-contrast images, mild peripheral enhancement of the capsule of the cystic mass lesion was noted but T2 hyperintense signal, which was tracking into subcutaneous fat plane, and the cyst content was not enhancing, showing rupture (Figure 3).

The patient was treated surgically under spinal anesthesia. Longitudinal skin incision was made over the lateral aspect of the left proximal leg over the swelling (Figure 4). After dissecting the subcutaneous tissue, transparent gelatinous

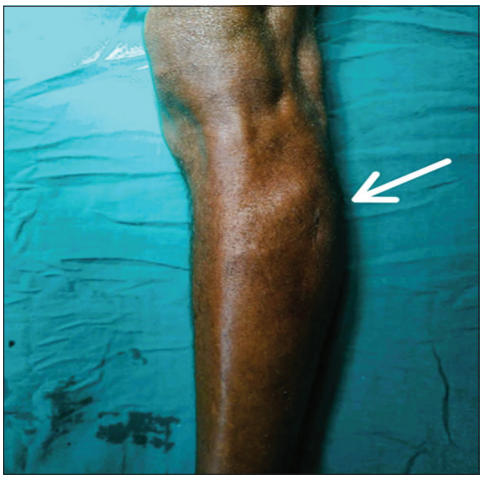

Figure 1: Sixty-year-old male who presented with a diffuse swelling over the lateral side of proximal leg. A linear scar (arrow) from a previous surgery can be seen over the swelling

Figure 2: Magnetic resonance imaging shows a multiloculated fluid-filled cystic lesion, which is iso- to hypointense on T1W1 (a) and hyperintense on T2/STIRW-weighted image (b and c) with its epicenter within the extensor digitorum longus muscle in the upper part of leg and abutting the proximal fibula posteriorly. It is also extending anterolaterally and slight inferiorly, tracking into the overlying subcutaneous fat plane

fluid spilled from the mass, indicating a ruptured ganglionic cyst. The cyst was completely excised and the sample was sent for histopathological examination, the findings of which were consistent with ganglionic cyst.

DISCUSSION

Intramuscular ganglionic cyst involving skeletal muscle usually represents an extension from a nearby joint. It

Figure 3: Magnetic resonance imaging shows post-contrast image T1 contrast FAT-SAT image. A mild peripheral enhancement of the capsule of the cystic lesion is noticeable, with the content remaining iso- to hypointense, showing no significant enhancement

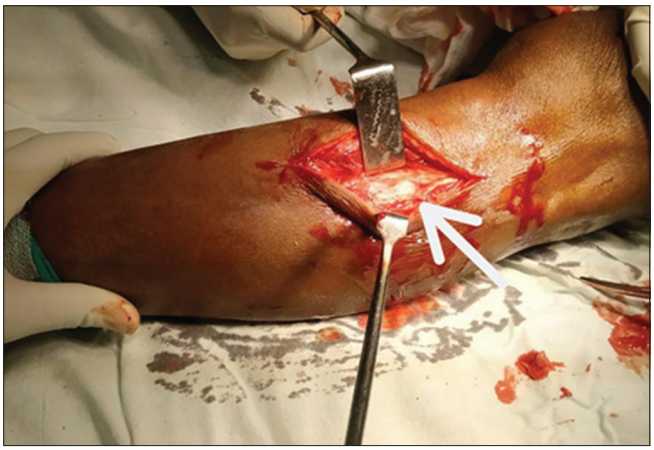

Figure 4: A linear incision made over the lateral side of the proximal part of the left leg showing a multilobulated swelling (arrow) within the extensor digitorum muscle; a thick myxoid gelatinous fluid oozed out after the incision

is proposed that a tear in the muscle sheath leads to the dissection of fluid into the muscle and leads to the formation of an intramuscular ganglionic cyst.[10]

In about 30% of the cases, ganglionic cysts arise as isolated intramuscular ganglionic cysts.[11] Commonly intramuscular ganglionic cysts are reported in the gastrocnemius medial head, gastrocnemius lateral head, rectus femoris, biceps brachii, semimembranosus, peroneus longus, extensor digitorum of forearm, thenar muscle of hand, and flexor hallucis brevis. In the present case, the involved muscle was the extensor digitorum longus of leg.

There are three predominant theories for the origin of ganglia, according to which they arise as an outpouching or weakening of joint capsules, from a mesothelial embryonic rest, or because of mucin formation by the fibroblasts lining the joint surface or bursa.[12] The most commonly accepted theory is that they result from secondary myxoid degeneration of the connective tissue that is developed from defects in the joint capsules and tendon sheaths.[2] Brooks suggested that a ganglionic cyst may arise from extra-articular synovial remnants in adjacent joints at the time of joint formation.[13]

Various theories explaining the occurrence of rare intramuscular ganglionic cysts have been propounded. Some suggest that ganglionic cysts are formed by the sequestration of joint fluid from a defect on the surface of a tendon. These types of cysts are known as intramuscular dissecting ganglion cysts. Intramuscular ganglionic cysts are usually asymptomatic and are most often detected incidentally on radiological evaluation for some other condition.[14,15] Ganglionic cysts rarely originate from cartilage, nerves, muscles, or bones (intraosseous). They very rarely arise from the muscle without having a connection to the adjacent joint capsule or tendon sheath.

Ganglionic cysts are frequently found in the hand, wrist, and foot. On the dorsum of the wrist, ganglion cysts commonly occur on the radial side of the extensor digiti communis tendon, whereas on the volar side of the wrist, they are commonly found in the flexor carpi radialis and the abductor pollicis longus tendons.[16] The differential diagnosis of ganglionic cysts can be tendon tear, Baker's cyst, bursitis, tenosynovitis, abscess, myxoma, nerve sheath tumor, vascular lesions lipomas, and synovial sarcoma.

The size of intramuscular ganglionic cysts is usually <50 mm in their maximum diameter.[17] James et al., reported that the mean maximal dimension of the intramuscular ganglionic cysts in a case series of 10 cases was 26 mm, ranging from 15 to 40 mm.[18] Vayvada et al., reported a giant ganglionic cyst of the vastus lateralis muscle, measuring 110×30×20 mm.[17] Chang et al., reported a large intramuscular ganglion, measuring 100×209×91 mm, in the mid-portion of the flexor hallucis brevis of foot.[19]

Intramuscular ganglionic cysts tend to grow slowly over a period of months to years. They are usually small and rarely symptomatic. Majority of them are deep-seated but in some cases, their manifestation as subcutaneous swelling has been reported.[20] In the present case, the intramuscular ganglion was deep-seated in the lateral aspect of the leg and extended subcutaneously.

The pre-operative diagnosis of ganglionic cysts is usually based on radiological evaluation. Plain radiographs are often not helpful. MRI and ultrasonography are highly sensitive, specific, and effective methods for diagnosing ganglionic cysts.[1] Intramuscular ganglionic cysts are lobulated

Table 1: Reported cases of intramuscular ganglion

S. No.	Author	Year	Age (years)/ Sex	Involved muscle	Side	Size of cyst	Communication with adjacent joint	Management	Remark
1	Mori et al.[21]	1993	62/F	Gastrocnemius medial head	Right	20×30×80 mm	No	Excision	
2	Rohrich and Rich.[22]	1994	39/F	Biceps brachii	Left	15×20 mm	No	Excision	Evolved from an embryological arrest
3	Chiou et al.[23]	1994	10/M	Thenar muscle	Right	30×10 mm	Yes	Excision	Rare in children
4	Beggs et al.[16]	1998	57/F	Gastrocnemius medial head	Left	20 mm	No	Excision	
5	Vayvada et al.[17]	2003	16/F	Vastus lateralis	Left	110×30×20 mm	No	Excision	Giant intramuscular ganglion
6	James et al.[18]	2007	27–68/ 7M, 3F	8 Gastrocnemius medial heads, 2 Gastrocnemius lateral head	5 Right 5 Left	15–40 mm	No	Excision	Case series of 10 cases
7	Park et al.[20]	2009	55/F	Gastrocnemius medial head	NA	NA	NA	Excision	
8	Kim et al.[24]	2009	43/F	Semimembranosus	Right	17×15×13 mm	No	Excision	Recurrence seen
9	Park et al.[22]	2010	60/M	Gastrocnemius	Right	NA	No	Excision	
10	Nicholson and Freedman.[14]	2012	53/F	Gastrocnemius medial head	Right	87×22×18 mm	No	Excision	
11	Kim et al.[11]	2013	17/M	Quadriceps femoris	Right	34×22×14 mm	No	Excision	
12	Najjar and Nasser.[12]	2015	46/M	Extensor digitorum muscle of the forearm	Left	50×26×15 mm	No	Wide excision	
13	Özen et al.[25]	2016	37/M	Peroneus longus	Left	83×35 mm	No	Wide excision	Peroneal neuropathic symptoms
15	Kang et al.[26]	2017	30/M	Peroneus longus and peroneus brevis	Right	50×70×30 mm and 40××10×30 mm	No	Excision	Multiloculated Mimicking peroneal compartment syndrome
16	Fan et al.[27]	2018	61/F 30/F	Gastrocnemius medial head Gastrocnemius lateral head	Right Right	40×30×30 mm 40×10×10 mm	No Yes	Excision Arthroscopic synovectomy	Two case reports Arising from pigmented villonodular synovitis
17	Madi et al.[28]	2018	52/M	Gastrocnemius medial head	Left	83×37×30 mm	No	Excision	A complex tear noted in the posterior horn of medial meniscus
18	Zachariah et al.[29]	2018	42/F	Gastrocnemius medial head	Right	72×40×30 mm	No	Excision	
19	Chang et al.[19]	2020	50/F	Flexor hallucis brevis	Right	100×209×91 mm	No	Conservative	
20	Singh et al.[30]	2021	12/M	Rectus femoris	Left	41×25 mm	No	Excision	
21	Present case	2024	60/M	Extensor digitorum longus of leg	Left	68×41×48 mm	No	Excision	Recurrence seen after 3 months

unilocular or multilocular swellings with well-defined septations ("bunch of grapes appearance") exhibiting homogeneous low signal on T1-weighted images and high signal on T2-weighted images with contrast enhancement at borders. Although ganglionic cysts can be identified using ultrasonography, this investigation is usually insufficient to establish intricate details regarding the probable origin of the lesion. MRI provides a better anatomical overview and is often necessary to study the relation of the cyst with the joint and adjacent structures, providing valuable insights into its origin. Intramuscular ganglionic cysts around the knee joint are much more uncommon, with only a few cases, mostly in the medial and lateral heads of the gastrocnemius muscles and in semimembranosus muscles, reported in the literature (Table 1).

In the majority of instances, ganglionic cysts can be managed conservatively. James et al., reported a series of 10 cases, all of which were incidental findings on MRI and were managed conservatively.[18] Aspiration is not recommended for ganglionic cysts as they have a high chance of recurrence. Recently Ju et al., described an ultrasound-guided aspiration and steroid injection therapy for symptomatic lower extremity ganglionic cyst, which included a single case of ganglion involving the posterior aspect of the knee.[31] Surgical excision is the gold standard for the management of large painful ganglionic cyst. Nicholson and Freedman reported a case of intramuscular dissection of a large ganglionic cyst in the gastrocnemius muscle that warranted surgical excision.[14] Han et al., reported a case of intramuscular ganglion, presenting as a swelling in the popliteal fossa that was increasing in size and was painful on palpation, and required surgical excision.[32] In the lower extremity, ganglionic cysts recur in approximately 10% of cases after surgical excision.[33,34]

In the present case, the ganglionic cyst recurred 3 months after surgical excision. The patient had a history of previous surgery for the same swelling 6 months earlier, with recurrence after 2 months of surgery. Therefore, even though some authors have reported ultrasound-guided aspiration of ganglionic cysts as a potential alternative to surgery, care should be taken in performing it on lesions in atypical locations.[23]

CONCLUSION

Intramuscular ganglionic cysts involving skeletal muscle usually present as an extension from a nearby joint. It is proposed that a tear in the muscle sheath leads to dissection of fluid into the muscle and the formation of intramuscular ganglionic cyst. Intramuscular ganglionic cysts are commonly reported in the gastrocnemius medial and lateral heads, rectus femoris, biceps brachii, semimembranosus, peroneus longus, extensor digitorum of the forearm, thenar muscle of hand, and flexor hallucis brevis. In the present case, the involved muscle was the extensor digitorum longus of leg. In the majority of cases, ganglionic cysts can be managed conservatively. Surgical excision is the gold standard for the management of large intramuscular ganglionic cysts. In the lower extremity, intramuscular ganglionic cysts recur after surgical excision in approximately 10% of the cases.

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DECLARATION

The authors confirm that written informed consent was obtained from the patient.

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