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Masson's Hemangioma of Knee: A Rare Case Report

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Learning Point of the Article:

Orthopedicians should lens out rare clinical entities when patients present with musculoskeletal complaints of chronicity.

Abstract

Introduction: Intrasynovial hemangioma, a rare benign vascular lesion of joint cavities, is also called intrasynovial papillary endothelial hyperplasia or Masson's hemangioma (MH). These lesions are characterized by abnormal proliferation of endothelial cells with mild atypia with an organized thrombus. MH follows an individualized protocol for its management.

Case Report: A 22-year-old male gave a 10-year history of the diagnosed and operated (arthroscopically) case of synovial hemangioma of the left knee and presented with similar complaints. There was no history of trauma or infection over the left knee. The movements of the left knee were normal except with terminal restriction of movements. MRI of the left knee suggested intrasynovial low flow vascular malformation within the substance of the synovium. The patient underwent synovectomy of the hemangiomatous lesion in toto without any recurrence in the follow-up period for 8 months. Histopathology confirmed the diagnosis of intravascular papillary endothelial hyperplasia/MH.

Conclusion: Although intrasynovial hemangioma is a very rare clinical entity, Orthopedic surgeons should have a high index of suspicion for MH, when a long-standing pain and hemarthrosis of the knee joint is being encountered in the clinical practice.

Keywords: Hemangioma, Intrasynovial, MRI, Excision biopsy.

Introduction

Intrasynovial hemangioma is a rare benign vascular lesion of joint cavities [1, 2, 3]. They contribute 2–7% of all hemangiomas. They are also called intrasynovial papillary endothelial hyperplasia or Masson's hemangioma (MH) [4, 5, 6]. These lesions are characterized by abnormal proliferation of endothelial cells with mild atypia with an organized thrombus [7]. The diagnosis of MH is a challenging task as they mimic angiosarcoma and Kaposi's sarcoma. The combination of radiological, histopathological, and histomorphological modalities aid in clinching the appropriate diagnosis of the lesion [4, 8]. After obtaining informed and written consent from the patient, we quote a rare case of MH and its management in a nutshell manner in view of invoking fundamentals and enhancement of knowledge among all the orthopedic practitioners.

Case Report

A 22-year-old male gave a history of pain and swelling in the left knee before 10 years for which he got radiologically diagnosed as synovial hemangioma and arthroscopic debridement and biopsy was done. Now the patient resulted in similar swelling and pain in the left knee which was dull aching pain and vague. The pain was aggravated on movements and relieved partially by rest and medications. On examination, the left knee swelling was tender, euthermic with boggy consistency, and synovial hypertrophy. The patient had a painful terminal range of movements of his left

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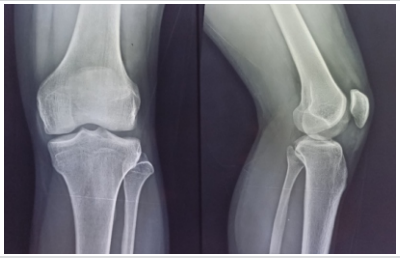


Figure 1: Plain radiograph of a left knee joint showing soft tissue swelling around left knee joint.

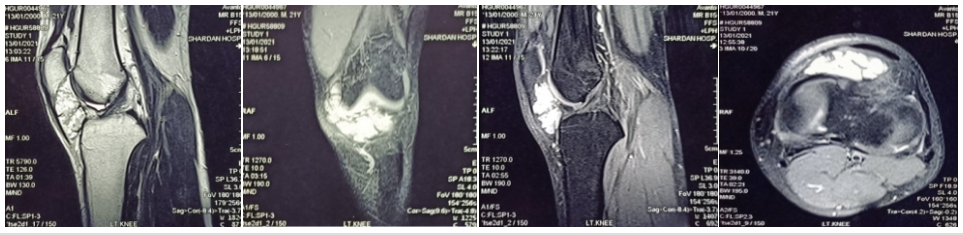


Figure 2: MRI of the left knee (a) T2W sagittal, (b) T2W coronal, (c) PDFS sagittal, and (d) PDFS axial sections showing hyperintense signals from the lesion.

knee. The plain radiograph of the left knee was shown in (Fig. 1). USG of the left knee revealed a well-defined, encapsulated mass of 4.2 × 1.9 cm^2 which was hypoechogenic in nature. Color doppler revealed the hypervascular nature of the lesion. MRI scan of the left knee was performed to understand the pathology, which revealed an encapsulated, well-defined, lobulated mass measuring 41 × 23 × 19 mm (approx.) in the substance of synovium lining of the left knee with hyperintense signals on T2W images and hypointense on T1W with a foci of gradient-echo images as shown in (Fig. 2a, 2b). PD-FS sequence shows hyperintense signals from the lesion in the anterior compartment of the left knee as shown in (Fig. 2c, 2d).

Under spinal anesthesia, surgical exploration of the left knee was done and performed excision biopsy of the mass along with the synovium as shown in (Fig. 3a, 3b). Grossly, the synovium of the left knee was admixed with the vascular tumor measuring 5.8 × 3.2 cm. The cut surface of the mass revealed numerous vascular channels with areas of thrombus and hemorrhages. The histopathological examination of the mass revealed fibrous tissue along with the papillary proliferation of vascular channels and organized thrombus as shown in (Fig. 4a). Few cells are dilated and lined by plump endothelial cells which show mild atypia. The histopathology section in (Fig. 4b) shows the papillary proliferation of blood vessels along with deposition of fibrin. Immunohistochemical studies showed positive staining for α-SMA and Ki-67 markers as shown in (Fig. 5a, 5b). The aforementioned findings were suggestive of MH. During the follow-up of 8 months, the patient showed a full range of movements over the left knee without any recurrence of the lesion.

Discussion

In 1923, Masson's coined the lesion as "vegetant intravascular hemangioendothelioma" in a patient with ulcerated irreducible hemorrhoids [5, 7, 9]. MH, called intravascular papillary endothelial hyperplasia (IPEH) [6, 10], synovial hemangioma [3], reactive papillary endothelial hyperplasias [8], or intravascular angiomatosis [11, 12], is a rare benign soft tissue lesion of vascular origin which is characterized by hamartomatous proliferation of endothelial cells [7]. Intrasynovial hemangioma contribute for 2–7% of all hemangioams. Hashimoto et al. reported three types of IPEH namely (a) primary IPEH (de novo pattern) (56%), (b) secondary IPEH (secondary to pre-existing vascular lesions) (40%), and (c) extravascular IPEH (secondary to post-traumatic hematoma) (4%) [13]. Although MH lesions are non-neoplastic, they mimic benign or malignant lesions of vascular origin such as pyogenic granuloma, angiosarcoma, and Kaposi's sarcoma [5, 14, 15]. MH lesions are more commonly seen in females around 30 years of age. They are frequently found in extremities, head and neck, and face but IPEH/MH occurrence in abdominal viscera were reported in the literature [5, 16].

It is hypothesized that IPEH/MH develops when organized thrombus as a result of vascular injury, mediate proliferation of endothelial cellular lining of the vascular channel [17, 18, 19]. There is a temporal association between basic fibroblastic growth factor and the development of IPEH/MH lesion [17, 20]. The molecular pathogenesis in IPEH/MH formation is by the release of endothelial b-FGF by macrophages in the surrounding environment producing a positive feedback loop mechanism of the proliferation of endothelial cells [6, 21].

Various studies have stated that the diagnosis of synovial hemangioma is delayed due to the presence of non-specific

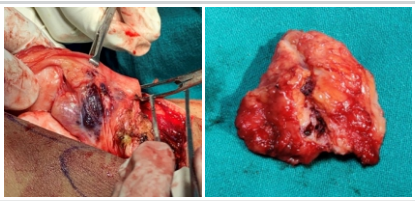


Figure 3: (a) Intraoperative image showing hemangiomatous lesion adhered to the synovium of the knee and (b) excised synovial specimen with hemangiomatous lesion.

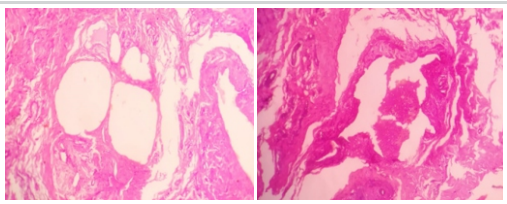


Figure 4: Histopathology of the excisional biopsy shows (a) fibrous tissue along with a papillary proliferation of blood vessels, lined by plump endothelial cells, and (b) organized thrombus with fibrin deposition.

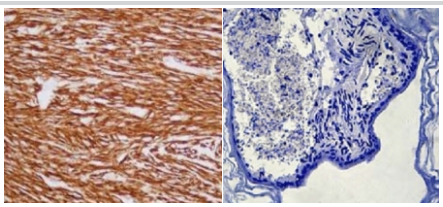


Figure 5: Immunohistochemistry section show positive staining for (a) α-SMA and (b) Ki-67.

symptoms. About 60% of synovial hemangioma occurs in the knee joint usually in the anterior compartment. These hemangiomas present as non-specific synovitis, synovial chondromatosis, giant cell tumor of the tendon sheath, fibrolipoma, lipoma arborescens, or localized villonodular synovitis of the affected knee. The diagnosis of IPEH plays a major diagnostic glitch. A radiograph of the affected knee joint shows no pathological findings other than soft tissue swelling around the knee joint. MRI assists in clenching the diagnosis and aid in planning the management. Sasho et al. studied MRI features of MH/IPEH who described the lesion as iso- or hypointense signals on T1W images and hyperintense signals on T2W images. With gadolinium injection, heterogeneous enhancement of the lesion can be visualized. In our case, MRI of the left knee showed a hyperintense signal from T2W and PD-FS images from the lesion in the anterior compartment of the left knee which was encapsulated, well defined lobulated mass measuring 41 × 23 × 19 mm (approx.) in the substance of synovium lining of the left knee.

The diagnosis of IPEH/MH lesions poses a great challenge among pathologists. The biological behavior of IPEH/MH must be differentiated from other lesions of vascular etiology. IPEH/MH is characterized by (a) intraluminal lesion with the intravascular origin, (b) organized thrombus, (c) minimal necrosis with lack of pleomorphism and mitotic figures, and (d) rarely extravascular hematoma [5, 22, 23]. The histomorphological features were consistent with the lack of pleomorphism and mitotic figures, possess mild endothelial cellular atypia, and intraluminal lesion with organized thrombus, which were suggestive of IPEH. Immunohistochemical markers aid in a more precise diagnosis of MH. Akdur et al. demonstrated positive staining for CD-31, -34, and -105, factor 8 and 13a, type 4 collagen, and α-smooth muscle actin in IPEH lesions [17]. In a series of IPEH lesion of the hands, Patai et al. reported positivity for WT1, CD-31, α-SMA, and ERG and negativity for GLUT-1 receptors [24]. In our case, we observed positive staining for ki-67 and α-SMA markers.

The management of IPEH/MH depends on the tumor size, extent, behavior, resectability, and the functional need of the individual [25, 26]. They range from intrasynovial instillation of sclerosing agents (sodium tetradecyl sulfate [3% sterol], sodium morrhuate, monoethanolamine oleate, and sodium psylliate) [27], holmium ablation [28], Nd:YAG laser [29], cauterization [30], freezing [30], radiotherapy, embolization [30], and open or arthroscopic synovectomy [31, 32]. In our case, we performed open synovectomy to provide a better functional outcome. The literature stated that inadequate excision poses a recurrence rate of 20–60% [33, 34, 35, 36]. No recurrence was reported in our case in the last 8 months of follow-up.

Conclusion

Although intrasynovial hemangioma is a very rare clinical entity, Orthopedic surgeons should have a high index of suspicion for MH, when a long-standing pain and hemarthrosis of the knee joint is being encountered in the clinical practice. MRI along with the histopathological examination of the biopsied lesion aid in the prompt diagnosis of the MH. Complete excision of MH/IPEH prevents the occurrence of recurrence in the follow-up period.

Clinical Message

Orthopedicians should include rare entities under the mindful list of probable diagnosis when patients present with a history of chronic pain in the knee joint with or without hemarthrosis in order to plan best treatment as per patient's profile and deliver optimum care to subjugate associated complications compromising functional quality of patient's life.

Declaration of patient consent : The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient's parents have given their consent for patient images and other clinical information to be reported in the journal. The patient's parents understand that his names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil **Source of support:** None

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