CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 14 (2015) 80–84

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Large prepatellar glomangioma: A case report

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ARTICLE INFO

Article history:
Received 17 April 2015
Received in revised form 29 June 2015
Accepted 4 July 2015
Available online 10 July 2015

Keywords:
Benign prepatellar glomangioma
Glomus tumor
Knee
Magnetic resonance imaging

ABSTRACT

INTRODUCTION: Glomangiomas are rare, benign tumors derived from the glomus body, typically presenting with the classic triad of pain, tenderness to palpation, and hypersensitivity to cold. Most commonly they present as a solitary lesion in the extremities, especially subungual, but they may present elsewhere.
PRESENTATION OF CASE: We describe the case of a large (64 mm × 59 mm × 41 mm) glomangioma on the anteroinferior aspect of a healthy 49 year old male's knee. Symptoms included constant throbbing pain with intermittent stabbing sensations localized to the mass. The mass was evaluated first by magnetic resonance (MR) imaging and then by histopathology following excision.
DISCUSSION: Although rare, clinical diagnosis of glomangiomas may be sufficient in typical cases, however in atypical cases, like the one discussed here, further evaluation is often necessary. Here MR findings were suggestive of a glomangioma with low to intermediate signal strength on T1 and mixed signal strength on T2. Intravenous gadolinium infusion demonstrated marked heterogeneous enhancement of the lesion, as well as serpiginous vascular malformations surrounding the lesion. Histopathology following excision confirmed a benign glomangioma depicting monomorphic small, round eosinophilic cells with minimal atypia which stained positive for smooth muscle actin, and negative for cytokeratin, S-100 and CK-34 via immunohistochemistry.
CONCLUSION: The following case report details an atypical presentation of a benign glomangioma anterior to the knee in a patient experiencing chronic minor trauma to the area. Diagnosis was suggested by clinical presentation and MR imaging, and was confirmed histologically.

1. Introduction

Surgeons are occasionally confronted with complex soft tissue tumors where the diagnosis may be in doubt. A carefully considered approach to the diagnosis and initial management is essential. Glomangiomas are rare, benign tumors derived from the glomus body. They have been reported to account for 1.6% of all soft tissue tumors in the extremities [1]. Glomus tumors typically arise in the extremities, especially subungual, but they may also be found in atypical locations. Surgical removal of glomangiomas is usually effective and curative. Unfortunately, diagnosis is often delayed. There are few reports of glomus tumors in the knee area, often presenting with knee pain (see Table 1). In the present study, we report an unusual case of a large prepatellar glomangioma.

2. Presentation of case

An otherwise healthy 49 year-old man presented with a progressively enlarging, painful mass on the anteroinferior aspect of his left knee for 1 year. Over the year the mass had gradually increased in size and become more painful. More recently, the patient described a constant throbbing pain with intermittent "stabbing" sensations and limited range of motion. The patient was a diesel mechanic and spent many hours on his knees. He reported multiple episodes of minor penetrating injuries to the area. On physical exam, there was limited range of motion at the left knee joint; a heterogeneous mass (60 mm × 50 mm × 50 mm) appeared on the anteroinferior aspect of the left knee, which was exquisitely tender with surrounding erythema and warmth (Fig. 1a,b). The mass also demonstrated small areas of ulceration (Fig. 1a). No regional lymphadenopathy was palpated.

Plain radiograph of the left knee joint showed a mass superficial to the patellar tendon with a nonspecific focus of calcification.

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http://dx.doi.org/10.1016/j.ijscr.2015.07.002

Table 1
Previously reported glomus tumors in the knee area: location, size, and patient attributes. Case report described presently included for reference (grey). *Mean (Range), NR = not reported.

Location	Size (mm)	Age (yrs)	Sex	Presenting Duration of Symptoms	Trauma History	Refs.
Medial aspect of knee	65 × 35 × 15	10	M	2 wks	Yes	[20]
Lateral femoral condyle	6 × 12 × 16	33	M	10 yrs	NR	[21]
Infrapatellar fat pad	8 × 5	42	F	1 yr	NR	[22]
Medial aspect of knee	50	73	M	3 yrs	NR	[23]
Prepatellar	15 × 11 × 20	75	M	30 yrs	NR	[19]
Medial aspect of knee	8 × 5	47	M	1 yr	No	[24]
Lateral aspect of knee	15 × 15 × 12	65	M	10 mo	No	
Anterior aspect of knee	4-5	60	M	4 yrs	NR	
Anterior aspect of knee	20 × 8 × 4	65	M	NR	No	
Popliteal possa	10 × 15 × 20	9	F	2.5 yrs	No	[25]
Anterior aspect of knee	10 × 10	69	M	5 yrs	Yes	[26]
Medial to tibial tuberosity	15 × 20	48	F	10 mo	Yes	
Present case: prepatellar	64 × 59 × 41	49	M	1 yr	Yes	–

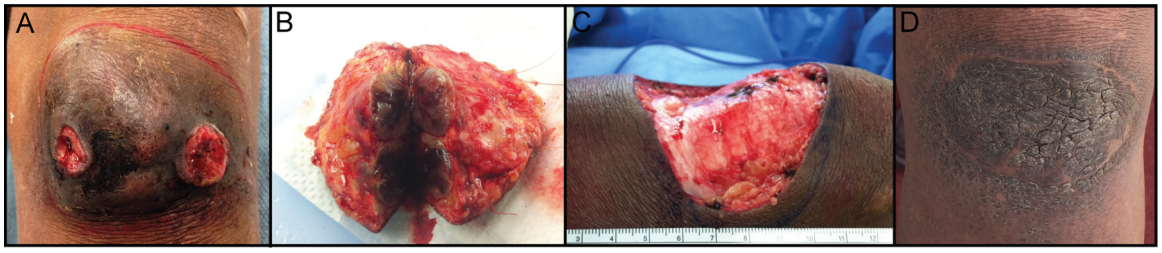

Fig. 1. A Large prepatellar mass following incisional biopsy. B. Excised gross specimen: gray/brown multinodular, encapsulated, and hemorrhagic mass measuring 55 ×43 × 27 mm with negative gross margins. C. Surgical defect following excision. Ruler demonstrates cm increments. D. Final aspect of the wound 2.5 years post-operative.

MR imaging (Siemens Symphony 1.5T) demonstrated a heterogeneous 64 mm × 59 mm × 41 mm mass, centered in the prepatellar subcutaneous fat, abutting the patellar tendon (Fig. 2). There was intermediate signal strength on T1 (Fig. 2), heterogeneous signal strength on T2 (Fig. 3), and marked signal enhancement with gadolinium (Fig. 4). Incisional biopsies at three sites of the mass showed reactive inflamed granulation tissue. The mass was excised using sharp dissection, and then vacuum assisted closure was performed.

Grossly the specimen demonstrated a gray/brown multinodular, encapsulated, and hemorrhagic mass measuring 55 mm × 43 mm × 27 mm with negative gross margins (Fig. 1). Histopathology demonstrated a monomorphic population of small, round, eosinophilic cells with minimal atypia (Fig. 5). Immunohistochemistry was positive for smooth muscle actin (Fig. 5) and negative for cytokeratin, S-100, and CK-34. This was consistent with a glomangioma. Once final histopathology confirmed the benign nature of the lesion the wound was closed with a split thickness skin graft. The graft was harvested from the left lateral thigh using a 0.018 inch dermatome with a 3 inch guard and placed unmeshed on the 80 × 100 mm wound. Numerous slit incisions were then made in the graft (pie crusting) to allow for fluid drainage. As of this writing the patient had remained symptom free for 2.5 years with no recurrence.

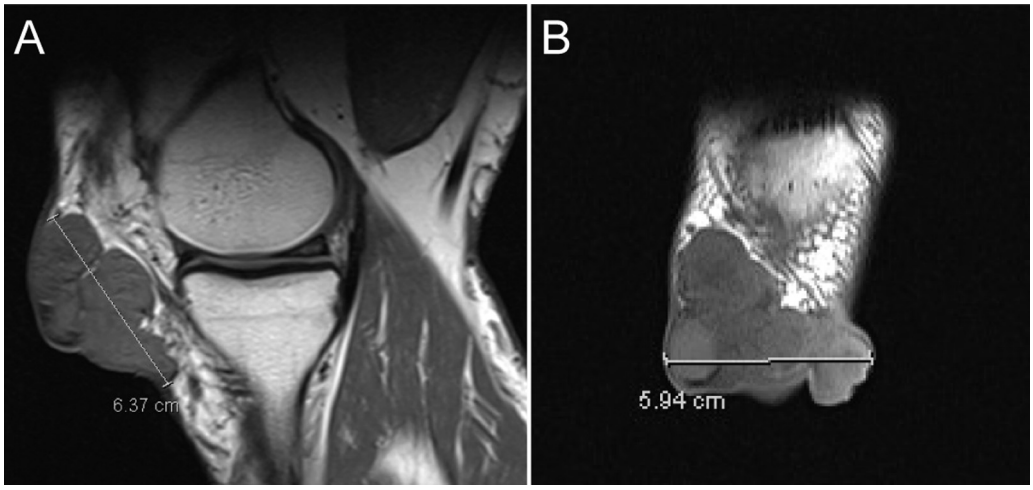

Fig. 2. T1-weighted MR images of a large soft tissue mass of the left subpatellar area (A) in the sagittal plane (TE: 18 ms, TR: 688 ms) and (B) in the coronal plane (TE: 11 ms, TR: 552 ms), measuring 64 mm craniocaudal × 59 mm transverse × 41 mm anterior-posterior mass. This soft tissue mass is lobulated and of low to intermediate strength on T1-weighted images. The lesion does not appear to invade the patellar tendon, bone or joint space.

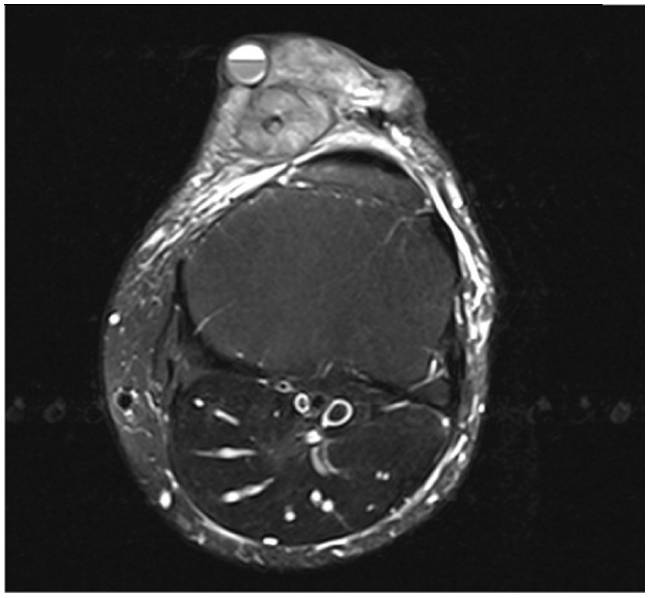

Fig. 3. T2-weighted MR image in the transverse plane (TE: 104 ms, TR: 6090 ms). The lobulated mass shows a semi cystic/fluid–fluid structure. The lesion is generally heterogeneous in signal strength. A moderate amount of subcutaneous edema is also present. The lesion is separated from the patella tendon by a thin fat plane.

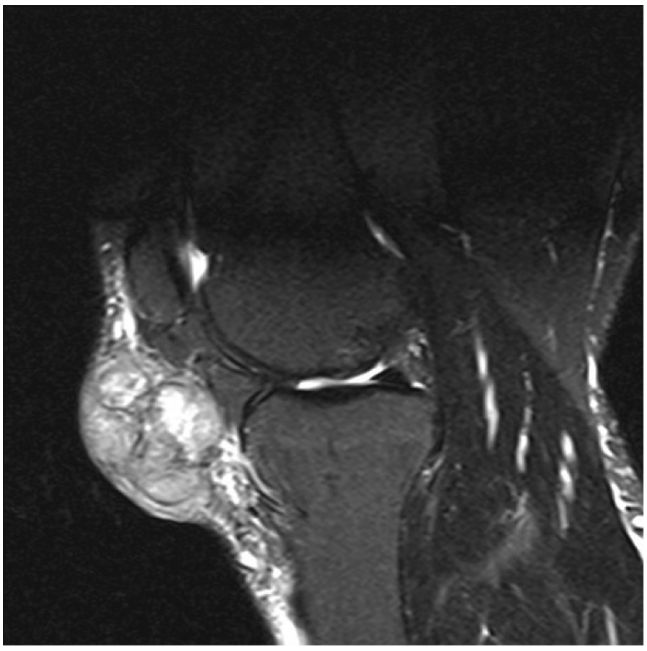

Fig. 4. T2-weighted MR image with intravenous gadolinium infusion demonstrates marked heterogeneous enhancement of the lesion (TE: 86 ms TR: 4240 ms). This finding in addition to the serpiginous vascular malformations surrounding the lesion is highly suggestive of a glomangioma. The enhancement contrasts with that seen in sarcomas where the outer margins of the lesions enhance to a greater degree.

3. Discussion

A glomus tumor is a neoplasm composed of the perivascular smooth muscle cells responsible for thermoregulation in the dermis. These cells are associated with arteriovenous anastomoses involved in the release of heat based on the degree of dilation in the dermis. Glomus tumors are most frequently found in the skin, particularly associated with the extremities. In approximately 65% of cases, the tumor presents in the subungual position and most commonly is diagnosed between 20 and 40 years of age [2]. While glomus cells are typically a dermal finding, there are case reports of glomus tumors found in tissues that are not normally associated with these cells: oral cavity, kidney, liver, GI tract, lung, genitals [3–7]. Extradigital glomangiomas involving the knee have previously been reported in the subcutaneous tissue, infrapatellar fat pad, the popliteal fossa and lateral femoral condyle as detailed in Table 1.

The typical presentation of glomus tumors is a red-purple cutaneous nodule, usually with pain out of proportion to size. The average duration of symptoms is between 7 and 11 years [8]. They

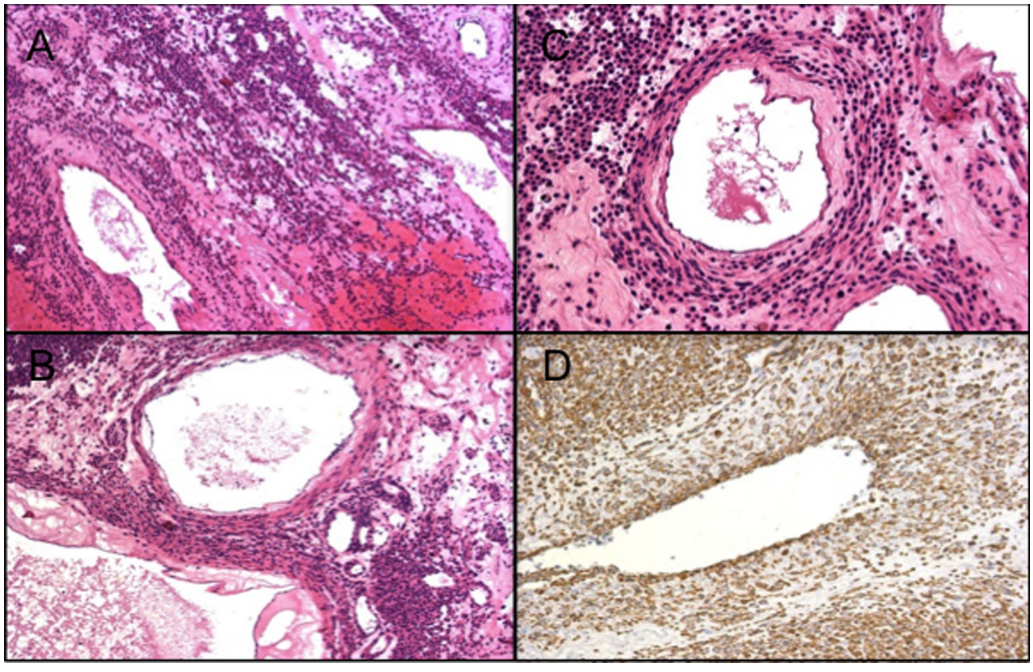

Fig. 5. A. Large dilated vessels with surrounding hemorrhage, normal endothelial cells and glomus cells. B. Large vessels with infiltrating glomus cells. C. Small rounded cells with eosinophilic cytoplasm encircling vessels indicates glomus cells. D. Smooth muscle actin confirmed by immunohistochemistry indicates glomus cells.

have a deep red–purple color and are soft to palpation. They are typically far smaller than our case, usually between 0.5 and 2 cm. Glomus tumors frequently demonstrate cold-sensitivity resulting from reflex vasodilatation; this may be attributed to the role of glomus cells in thermoregulation. Clinically, glomus tumors present infrequently and make up less than 2% of soft tissue tumors. In typical presentations, such as subungual, clinical diagnosis may be sufficient. Atypical presentations, however, may necessitate further evaluation, including MR imaging and histology.

Glomus tumors may be subcategorized as solid type, glomangiomas, and glomangiomyoma [9]. Most common is the solid type, in which the glomus cells predominate, appearing in solid cell sheets. The solid type is found in 75% of cases. The glomangioma variant is associated with greater vascularity, and can be found in 20% of resected lesions. Lastly, the glomangiomyoma subset, in which both vascularity and smooth muscle cells are prominent, occurs in only about 5% of glomus tumors. While glomus tumors are primarily individual tumors, there is a tendency for multiplicity in 10% of presentations.

MR imaging can be used to identify the characteristics of glomus tumors prior to excision. Lesions have characteristic intermediate to low T1-weighted signal intensity and high T2-weighted signal intensity. Gadolinium enhancement is intense and diffuse, a characteristic identified in nearly all glomus tumors [10–12]. While glomus tumors demonstrate classic features with T1 and T2-weighting as well as contrast enhancement, prior literature has demonstrated a glomus tumor located in the tip of an index finger that lacked the classic MR features [13]. Furthermore, while MR imaging may be used to identify many characteristics of soft tissue tumors, the differentiation of benign from malignant lesions is not absolute [2,12]. Imaging findings may then be coupled with the size, location, borders, adjacent tissue involvement, and internal characteristics of the lesion to inform a differential diagnosis and surgical planning [11]. The images in this case could not definitively classify the lesion as either benign or malignant.

Glomus tumors, including solid type, glomangioma and glomangiomyoma, may be confirmed histologically. Microscopically, solid type glomus tumors consist of solid sheets of glomus cells. Glomangiomas consist of vascular malformations with large, often dilated vascular channels and surrounding nests of glomus cells. Glomangiomyoma demonstrate glomus cells, vascular channels, as well as smooth muscle cell differentiation. Glomus cells, thought to serve as thermoregulatory receptors, are small uniform cells with prominent eosinophilic cytoplasm and centrally located, rounded nuclei. They are positive for smooth muscle actin, myosin, vimentin, and desmin, all supporting a likely smooth muscle origin. Glomangiomas are typically less circumscribed than their better-known counterpart, solid type glomus tumors, which lack such prominent vessels. Mitotic figures are rare in both entities, which also lack zonal necrosis and significant cellular pleomorphism. Glomus tumors are primarily benign neoplasms which are usually "cured" by local excision with little risk of recurrence [14,15].

While glomus tumors are primarily a benign pathology, there is a 1% likelihood of malignancy. While this is rare, there are case reports of metastasis [16]. Features that increase the likelihood of malignancy include size greater than 2 cm, deep location, abnormal mitotic figures, mitotic rate greater than 5 per high-power field, and moderate to high nuclear grade. If all of these criteria are met, the risk of metastasis has been found to be 25%; however incomplete criteria cannot be used to confirm nor deny malignant potential [17]. Should malignancy be determined, there are three patterns of development: locally infiltrative glomus tumors, glomangiosarcoma arising from benign glomus tumor, or glomangiocarcinoma arising *de novo*. Management of a glomus tumor is primarily excisional. Sclerotherapy using sodium tetradecyl sulphate has been reported to be effective in patients with multiple hereditary glomangiomas but excision is the treatment of choice [18]. There is a recurrence rate of approximately 10%, [19] generally associated with incomplete excision. While there are examples of using Mohs surgery in the literature for cosmetically sensitive areas, the typical procedure is wide local excision. The typically small size makes primary closure more feasible than in our case, in which the large area of excision necessitated use of a split-thickness skin graft.

4. Conclusion

We report a case of a large glomangioma in rare location, anterior to the knee, presenting with sharp pain and a history of chronic minor trauma to the area. The diagnosis was suggested by clinical presentation and MR imaging, and the final diagnosis was confirmed by histological evaluation.

Conflict of interest

None

Funding

None

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request

Author contributions

Richard Bell, Melissa Maxey, Katherine Mastriani and Fernando Navarro participated in the diagnosis and treatment of the present case. Richard Bell, Chase Houghton, Katherine Mastriani, Melissa Maxey, and Fernando Navarro drafted, critically revised, and approved the manuscript.

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