Radiological and pathological characteristics of synovial hemangioma of the knee

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Abstract. Synovial hemangioma, a rare benign tumor that occurs most frequently in the knee in children and young adults, has four histological subtypes: Venous, arteriovenous, cavernous and capillary hemangiomas. Since the clinical presentation and radiological findings of synovial hemangioma are non-specific, there is frequently a long period between the onset and the diagnosis. The cases of nine patients, pathologically diagnosed with synovial hemangioma and surgically treated, were retrospectively analyzed. All nine patients had persistent knee pain. In addition, three patients also had a swollen knee with intra-articular hemorrhage. Plain radiography revealed intra-articular phleboliths in two patients. In seven patients, T1-weighted magnetic resonance imaging showed low signal intensity with small signal voids. On T2-weighted imaging, all patients showed high signal intensity containing small signal voids. All patients underwent surgical excision; there was no postoperative recurrence after the final operation, and the knee pain had disappeared at the final follow-up. From the pathological findings, the diagnoses were venous hemangioma, cavernous hemangioma and capillary hemangioma (three patients each).

Introduction

Synovial hemangioma is a rare benign tumor that occurs most frequently in the knee in children and young adults (1-3). There are four histological subtypes: venous, venous vascular malformation, cavernous, and capillary hemangiomas (4). Since the clinical presentation and radiological findings of synovial hemangiomas are nonspecific, there is often a long period from the onset to the diagnosis. In some cases, the diagnosis is made after osteoarthritis and cartilage damage have already occurred (5). After the diagnosis, an open or arthroscopic resection is performed in most cases, and good postoperative outcomes have been reported (6). Because of the rarity of synovial hemangiomas, most reports described a single clinical case, and they highlighted the rarity of this tumor. In this report, we describe a series of nine patients with synovial hemangiomas and the results of our retrospective analysis of the correlation between patients' pathological and radiological characteristics. Moreover, the usefulness of diffusion-weighted image (DWI) in the differentiation of synovial hemangioma from diffuse-type tenosynovial giant cell tumor (D-TSGCT) (formerly named pigmented villonodular synovitis) is also discussed.

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Key words: synovial hemangioma, knee, intra-articular tumor, magnetic resonance imaging, apparent diffusion coefficient, diffuse-type tenosynovial giant cell tumor

Patients and methods

After obtaining institutional review board approval, we identified the nine patients (five males and four females) who were each diagnose with synovial hemangioma of a knee and treated at Fukushima Medical University Hospital (Fukushima, Japan) or Fukushima Red Cross Hospital (Fukushima, Japan) during the period between January 1998 and December 2021. The patients' median age at surgery was 22 years (range 1-43 yrs). We retrospectively reviewed the patients' clinical presentations, radiological findings, surgical procedures, postoperative courses, and pathological findings.

Results

Clinical presentations and tumor locations. Table I summarizes the patients' clinical characteristics. All nine patients had persistent knee pain. Three patients also had a swollen knee with intra-articular hemorrhage. The duration of symptoms from their onset to the patient's initial visit varied, ranging from 1 day to 10 years. One patient had multiple lesions (Patient #6). Among the other eight cases, the synovial hemangioma was located in the suprapatellar bursa in two patients (#2 and #4), in the subcapsular fat body in four (Patients #1, #5, #7, and #9), and anterior to the femoral condyle localized in two (Patients #3 and #8).

Table I. Patients' clinical characteristics.

Patient no.	Age, years	Sex	Symptom	Intra-articular hemorrhage	Symptom duration (until first visit)	Affected side	Location
1	1	F	Pain, swelling	+	3 months	Right	Infrapatella and suprapatellar fat-pad
2	11	F	Pain	-	3 months	Right	Suprapatellar bursa
3	14	M	Pain	-	1 month	Left	In front of the medial condyle of femur
4	17	F	Pain	-	0.5 months	Left	Suprapatellar bursa
5	22	M	Pain, Swelling	+	1 day	Right	Infra patella fat-pad
6	24	M	Pain	-	10 years	Left	Inter condylar fossa, infra patellar fat-pad and posterolateral capsule
7	28	M	Pain	-	1 year	Right	Suprapatella fat-pad
8	34	F	Pain	-	6 months	Left	In front of the medial condyle of femur
9	43	M	Pain, swelling	+	1 months	Left	Infrapatellar fat-pad

F, female; M, male.

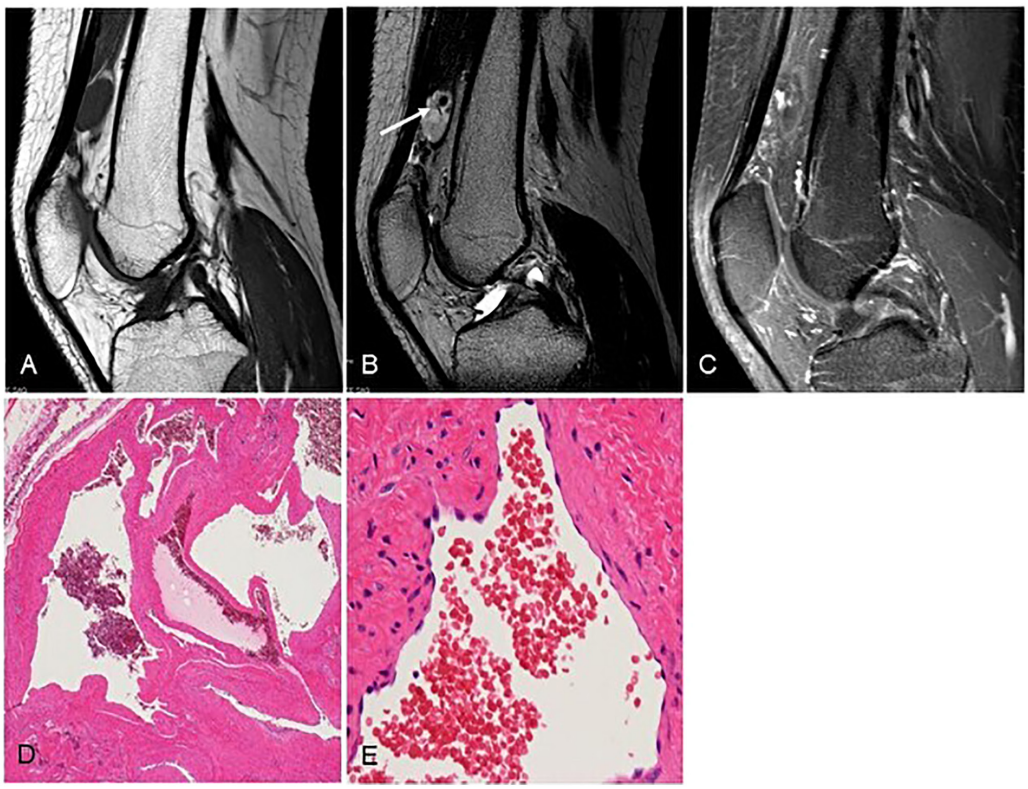

Figure 1. Venous synovial hemangioma (patient #4). On magnetic resonance imaging, (A) T1-WI (sagittal view) showed low-signal intensity without a signal void and (B) T2-WI (sagittal view) showed high signal intensity containing small signal void (arrow). (C) Gadolinium-enhanced T1-weighted fat-suppression imaging (sagittal view) showed heterogeneous staining of the tumor. Pathologically, the vascular smooth muscle is thickened and vascular lumen is dilated: (D) low-power field (magnification, x20), H-E staining; (E) high-power field (magnification, x200), H-E staining. Slow blood flow is prone to forming thrombus and phleboliths. H-E, hematoxylin and eosin.

Radiological findings. On plain radiographs, intra-articular calcification consistent with phleboliths was observed in two patients. T1-weighted magnetic resonance imaging (MRI) showed low signal intensity with and without small signal voids in six patients and two patients, respectively, and high signal intensity without signal voids in the other patient (Patient #7). In all nine patients, T2-weighted imaging (WI) showed high signal intensity containing small signal voids

Table II. Radiological characteristics of the tumor.

Patient no.	Size, cm	Plain radiography	T1-weighted image	T2*-weighted image	Gadolinium-enhanced, T1-weighted fat-suppression image	DWI ADC-mapping
1	3.7x2.4x1.9	Phlebolith	Low intensity	High intensity, containing low intensity area of hemorrhage	-	-
2	3.0x2.8x1.5	Normal	Low intensity, containing small signal voids	-	-	-
3	7.2x4.1x0.9	Cortical erosion, phlebolith	Low intensity, containing small signal voids	-	Heterogeneous enhancement	-
4	3.1x2.1x1.1	Normal	Low intensity	-	Heterogeneous enhancement	-
5	1.5x0.8	Normal	Low intensity, containing small signal voids	-	-	-
6	Diffuse/multiple	Normal	Low intensity, containing small signal voids	-	-	-
7	3.8x3.3x1.1	Normal	High intensity, containing small signal voids	-	Heterogeneous enhancement	-
8	4x3.5x1.1	Normal	Low intensity, containing small signal voids	-	-	-
9	4.7x5.9x2.9	Normal	Low intensity, containing small signal voids	High intensity, containing low intensity area of hemorrhage	Heterogeneous enhancement	ADC value 2184

For all nine patients, the T2-weighted imaging result was high intensity, containing small signal voids. DWI, diffusion-weighted image; ADC, apparent diffusion coefficient.

and intra-tumoral septum comparable to joint fluid and fat. In the present cases, the synovial hemangiomas showed a small honeycomb pattern with a thin septum and a lobulated pattern. T2*-WI showed high intensity, containing low intensity area that could be post-hemorrhagic changes. Gadolinium enhancement was performed for four patients, with heterogeneous staining of the tumor (Figs. 1 and 2). The maximum tumor diameter ranged from 1.5 to 7.2 cm (Table II). MRI findings of D-TSGCT show nodular and extensive synovial proliferation, joint effusion, and bone erosion. Hemosiderin deposition in the proliferative synovial tissue results in point-like low signal on T1-weighted and T2-weighted images and extensive low-signal changes within the proliferative synovium. These may be differentiation from synovial hemangioma.

Surgical procedures and postoperative courses. The surgery was performed without a preoperative biopsy in all nine patients. The duration from the patient's initial visit to the surgery ranged from 1 to 9 months. Open resection (n=2 patients), arthroscopic resection (n=3), and conversion to open resection after arthroscopy (n=4) were performed. In one of the two patients who underwent an open resection (Patient #6) and in one of the patients who underwent arthroscopic resection (Patient #4), additional resections for local recurrence were performed.

The average postoperative follow-up period from the final surgery was 17.2 months, with the longest follow-up period being 60 months. After the final surgery, none of the patients developed postoperative recurrence, and their knee pain had disappeared at the final follow-up (Table III).

Pathological findings. Based on the pathological findings of surgical specimens, the diagnoses were venous hemangioma (Fig. 1), capillary hemangioma (Fig. 2), cavernous hemangioma

Table III. Surgical procedure, pathological diagnoses and oncological prognosis.

Patient no.	Duration from initial visit to surgery, months	Procedure	Pathological sub-type	Follow-up period, months	Prognosis
1	45	Open resection after arthroscopy	Cavernous hemangioma	11	NED
2	5	Arthroscopic resection	Capillary hemangioma	12	NED
3	9	Open resection	Cavernous hemangioma	1	NED
4	2	1st surgery: Arthroscopic resection 2nd surgery: Open resection	Venous hemangioma	9	NED[a]
5	3	Arthroscopic resection	Capillary hemangioma	6	NED
6	1	1st surgery: Open resection 2nd surgery: Arthroscopic resection	Venous hemangioma	41	NED[a]
7	3	Open resection after arthroscopy	Cavernous hemangioma	13	NED
8	7	Open resection	Venous hemangioma	2	NED
9	2	Open resection after arthroscopy	Capillary hemangioma	60	NED

[a]NED after the resection of local recurrence. NED, no evidence of disease.

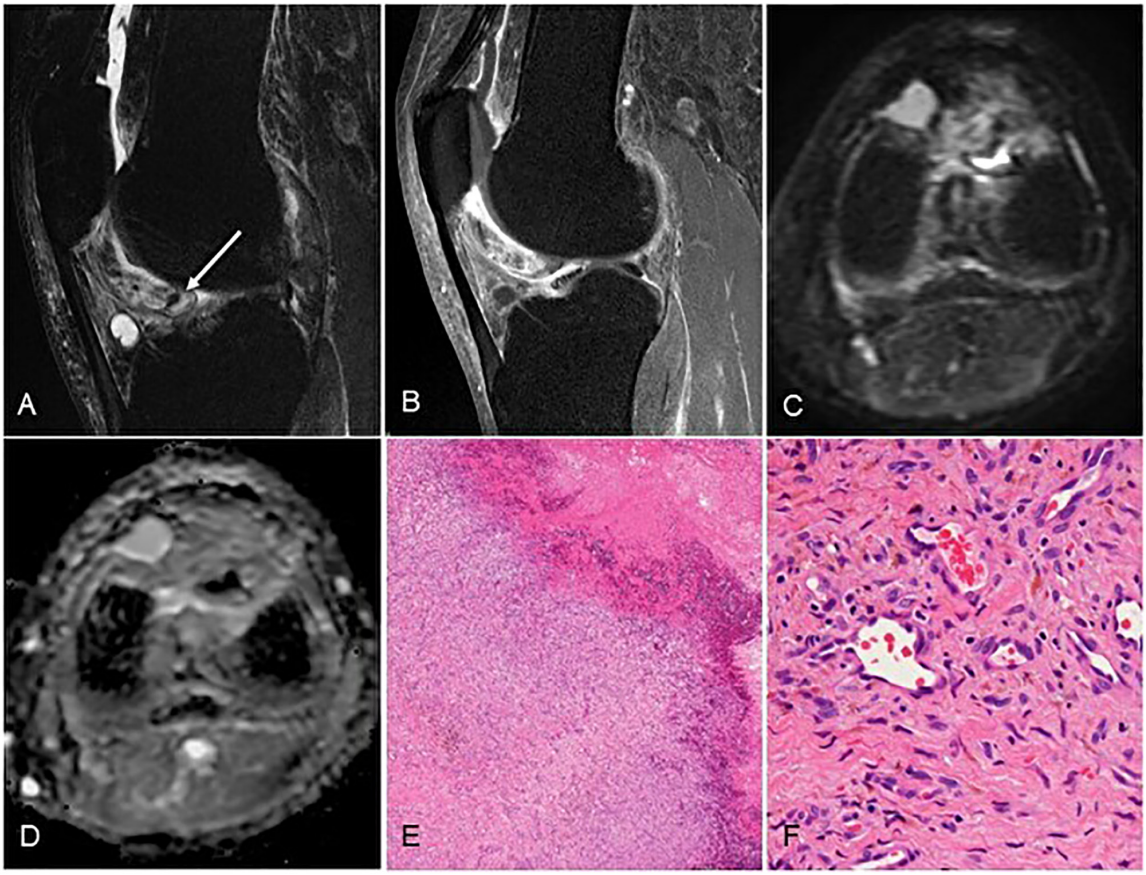

Figure 2. Capillary hemangioma (patient #9). (A) On magnetic resonance imaging, T2-weighed fat-suppression imaging showed high signal intensity containing small signal void (arrow). (B) Gadolinium enhancement showed heterogeneous staining of the tumor. (C) DWI showed low-signal intensity in the tumor and (D) high apparent diffusion coefficient (ADC) value of 2,184/mm2/sec. Microscopic findings show small vascular lumen, which was less likely to form thrombus or phleboliths, and this tumor was difficult to diagnose without microscopic confirmation: (E) low-power field (magnification, x20), H-E staining; (F) high-power field, (magnification, x200), H-E staining. H-E, hematoxylin and eosin.

(Fig. 3), in three patients each. Venous hemangiomas have thickened vascular smooth muscle and a dilated vascular lumen. Because of slow blood flow, they are prone to forming thrombi and phleboliths. Capillary hemangiomas have a small vascular lumen and are less likely to form thrombus or phleboliths. Microscopy should be used for diagnosis. In cavernous

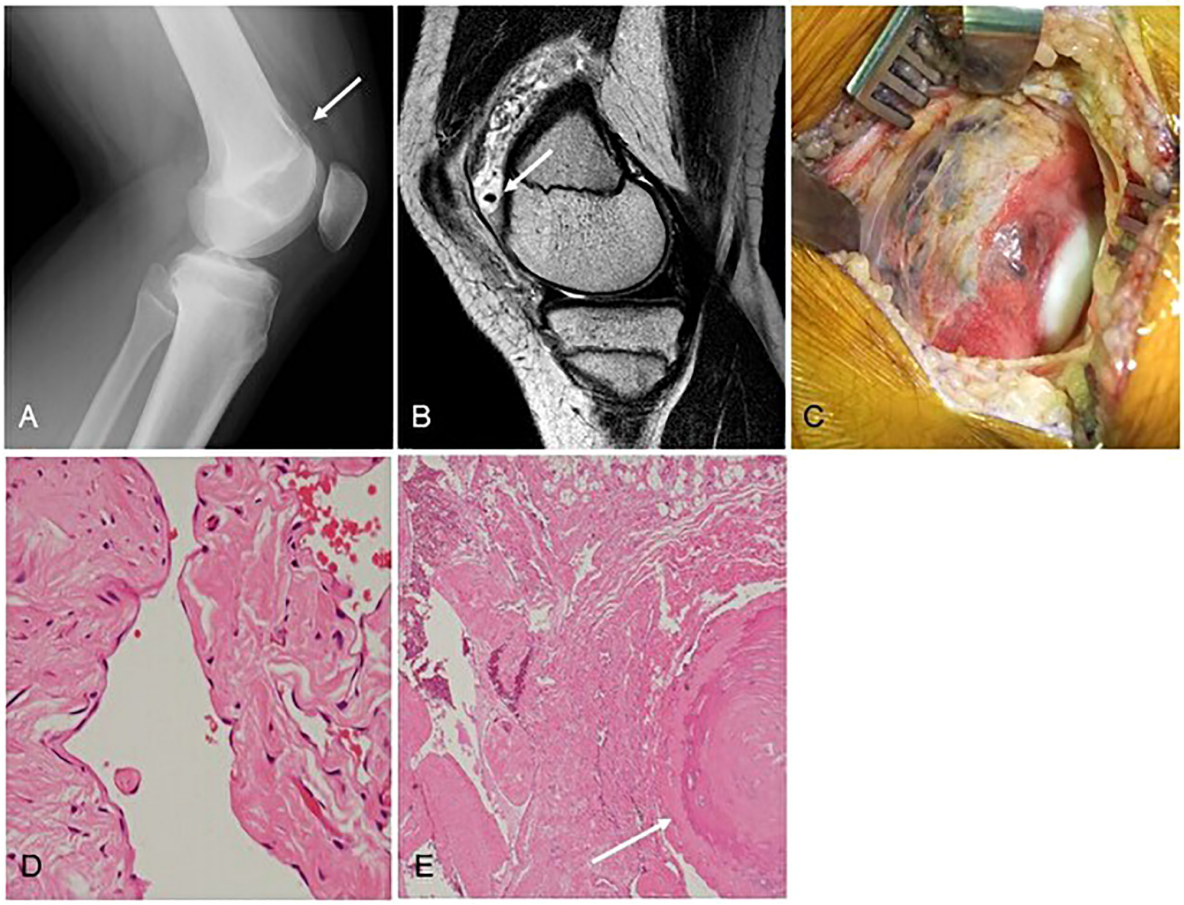

Figure 3. Representative case (patient #3, a 14-year-old boy). (A) Intra-articular calcification consistent with phlebolith (arrow). (B) Magnetic resonance imaging demonstrating a mass with high signal intensity containing a small signal void on T2-weighted images (sagittal plane). (C) Intraoperative gross findings revealing a dark red tumor covered by the synovium on the surface of lateral condyle of the femur. (D) Microscopic findings of the surgical specimen showing expanded blood lumens lacking vascular smooth muscle (high-power field, magnification x200). (E) Formation of thromboses and phleboliths caused by slow blood flow was easily identified (arrows) (low-power field, magnification x20).

hemangiomas unlike the venous hemangiomas, vascular smooth muscle is absent. The size of the intra-tumoral vascular lumen differs depending on the subtype of hemangioma.

Representative case (Patient #3). A 14-year-old boy presented with a 1-month history of continuous left knee pain. Plain radiographs of his left knee joint showed an intra-articular calcification consistent with phlebolith (Fig. 3A). MRI revealed a mass with high signal intensity containing a small signal void on T2-WI (sagittal plane) (Fig. 3B). From the radiological findings, we initially diagnosed a synovial hemangioma of the knee. Since the patient's left knee pain did not improve, we decided to perform the surgical resection 9 months after the initial visit. Because of the size of the tumor, open resection was selected. Intraoperative gross findings showed a dark red tumor covered by the synovium on the surface of lateral condyle of the femur (Fig. 3C), and the tumor was completely resected. The microscopic findings of the surgical specimen showed expanded blood lumens lacking vascular smooth muscle (Fig. 3D). The formation of thromboses and phleboliths caused by slow blood flow was easily identified (Fig. 3E). The tumor was diagnosed as a cavernous SH. At 1 month after the operation, the patient's symptom had completely resolved.

Discussion

Synovial hemangioma has accounted for 0.07% of all soft tissue tumors and 0.78% of resected hemangiomas (7). Although it can occur in a variety of joints, the most common location is the knee (1). Synovial hemangioma occurs most frequently in children and young adults (1-3), with male predominance. Since there is no specific clinical presentation, there is often a long period between the onset and the diagnosis (8). The most common symptom is long-term persistent knee pain; other symptoms include a limited range of motion, joint swelling, and nontraumatic intra-articular hematoma. However, except in phases of exacerbation, the presence of a synovial hemangioma does not limit activities of daily living, and patients with this benign tumor rarely visit a medical facility.

The clinical findings of synovial hemangioma are similar to those of D-TSGCT and hemophilic arthropathy. Both may cause sudden and non-traumatic intra-articular hemorrhage, and a delayed diagnosis can thus lead to osteoarthritis and joint function disuse (9). There are two case reports of synovial hemangioma in which >40 years passed from the onset of the disease until the final diagnosis (10,11). In another report, a 67-year-old patient was diagnosed with a synovial

hemangioma after experiencing severe osteoarthritis; the patient finally underwent a total knee arthroplasty (12). As a characteristic physical examination finding, cutaneous hemangiomas have been reported to be present in approx. 40% of patients with synovial hemangiomas in the knee (1), but in the present series there were no coexisting cutaneous lesions. A reported average duration from the onset of symptoms until the diagnosis of synovial hemangioma was 4.5 years (13). We were able to diagnose the disease earlier, with an average duration of 4.18 months from onset to diagnosis. When clinicians are diagnosing knee pain and/or intra-articular hemorrhage in young patients, the possibility of synovial hemangioma should be considered. Synovial hemangiomas are expected to have good surgical outcomes with resection, but if left undiagnosed for a long period of time, they can lead to osteoarthritis and functional disability of the knee. Preoperative imaging and intraoperative arthroscopic findings should confirm the extent of the tumor and if adequate resection is possible, arthroscopic resection should be used. If the tumor is difficult to resect due to its location or size, the approach should be switched to open. The present patient series includes a 1-year-old patient, and surgery is recommended as soon as the diagnosis and perioperative management are possible.

Plain radiography is usually the first radiological examination conducted in patients who are suspected to have knee pain and/or intra-articular hemorrhage. Plain radiography of a synovial hemangioma typically shows no abnormality, but periosteal reactions, osteolysis, osteopenia, intra-articular phleboliths, osteoarthritis, or soft tissue swelling may be observed (1,2,14-18). In the present patient series, we identified cortical erosion in one case and intra-articular phleboliths in two cases, but these findings were not specific for synovial hemangioma and could not be used as diagnostic evidence. As in other soft tissue tumors, MRI thus plays a more important role than plain radiography.

Compared to muscle tissue, an MRI evaluation of a synovial hemangioma reveals low to iso-intensity on T1-WI and high intensity on T2-WI, but no characteristic findings are seen on fat-suppression imaging (19,20). In the present patients, the synovial hemangiomas showed a small honeycomb pattern with a thin septum and a lobulated pattern. Small signal voids were also observed within the tumor in all nine patients. Contrast-enhanced MRI was performed in four cases, and gadolinium contrast showed heterogeneous enhancement only of the septum and tumor margins, indicating a cystic-like structure. In addition, a fibrous septum separated the tortuous vascular component, which has been reported as one of the specific findings of synovial hemangioma (21,22).

The size of the intra-tumoral vascular lumen differs depending on the subtype of hemangioma. In a larger vascular lumen, the blood flow becomes slower, and the contrast will be partial. It was reported that large venous synovial hemangiomas showed high signal intensity on T1-WI due to slow blood flow (20). Since synovial hemangiomas are blood-rich tumors and are usually heterogeneously enhanced on gadolinium contrast, the use of which helps distinguish these tumors from cystic synovial hyperplasia (23), intra-articular hematoma, and joint effusion (5,9,24,25). However, it has been claimed that synovial hemangioma was correctly diagnosed preoperatively in only 22% of cases (9,14). The usefulness of contrast MRI had been supported for confirming the localization and the extension of this tumor and detecting local recurrence in the postoperative follow-up (5).

The pathological diagnosis of synovial hemangioma is classified into the venous, arteriovenous, cavernous, and capillary subtypes (4). Venous synovial hemangiomas have thick vascular smooth muscle and a dilated vascular lumen. Cavernous synovial hemangiomas also have a dilated lumen, but unlike the venous subtype, they are differentiated by the absence of smooth muscle in the vessel wall. In both of these subtypes, the dilated lumen slows the blood flow and increases the formation of a thrombus and/or phleboliths compared to the capillary subtype, which has a smaller lumen. In the present patient series, the presence of phleboliths was confirmed by plain radiography and MRI in only two cases, both of which were the cavernous subtype (Fig. 2). On the other hand, capillary subtypes do not form a thrombus or phleboliths, because of their narrow lumen. It is thus difficult to diagnose capillary subtypes as hemangiomas without microscopic confirmation.

An important differential diagnosis for synovial hemangioma is D-TSGCT. Because of these two tumors' similarity in age of onset and clinical presentation, D-TSGCT can be difficult to distinguish in clinical practice. We noted that all nine of the present patients' cases showed high signal intensity on T2-WI, similar to circumferential fatty tissue and joint fluid. D-TSGCTs usually show low-signal changes on T2-WI due to hemosiderin deposition (26), whereas synovial hemangiomas show high-signal changes on T2-WI (23,27,28). This radiological difference in MRI between D-TSGCT and synovial hemangioma is thought to be useful. However, T2-WI shows various signal changes according to the presence of intra-articular hemorrhage or fatty degeneration, leading to potential diagnostic difficulty for synovial hemangioma on MRI.

In the present series, diffusion-weighted imaging (DWI) was performed in one of the capillary subtype cases. The DWI showed low signal intensity in the tumor and the high apparent diffusion coefficient (ADC) value of 2,184/mm^2/sec, excluding the area with high signal intensity that was thought to be post-hemorrhage changes (Fig. 3). Our review of eight cases of D-TSGCT of the knee that were treated at our institute between 2006 and 2021 revealed that the MRI findings revealed high signal intensity in DWI and the low mean ADC value of 770/mm^2/sec. Usually, in the case of a solid or malignant tumor, cell proliferation causes an increase in intracellular structures and a narrowing of the stroma, which restricts the movement of water molecules, resulting in a low ADC value. D-TSGCT is a synovial tumor with a high proliferation of tumor cells and restricted movement of water molecules. Conversely, synovial hemangioma is an angioproliferative disease in the subsynovial layer, and thus the movement of water molecules in the lumen and stroma is not as restricted as in D-TSGCT, and its ADC may be higher than that in D-TSGCT. The present case series raises the possibility that DWI and ADC values may be important differentiators between synovial hemangioma and D-TSGCT. On the other hand, the importance of enhanced MRI in diagnosing synovial hemangioma was not observed in this series.

Despite their similar clinical findings, synovial hemangiomas and D-TSGCT have different postoperative outcomes. Although two of the nine patients in our present series had postoperative recurrence because of an uncomplete resection, synovial hemangiomas have been reported to have a postoperative recurrence rate of 3.4% and a symptom persistence rate of 11.4% (29). From our experience both careful preoperative radiological examination and intraoperative attention may reduce the risk of recurrence. D-TSGCT has a high recurrence rate of 25-46% (30-32), even when open resection is performed. When D-TSGCT is strongly suspected from the MRI findings, surgeons should anticipate circumferential invasion and perform a more aggressive resection of the tumor. During the surgery, the identification of the differences in gross findings between synovial hemangioma and D-TSGCT is also useful in the final decision regarding the aggressiveness of resection.

A limitation of the present study is that the imaging conditions used for the MRI examinations were not standardized, and DWI and ADC values were measured in only one case. When we encounter patients with intra-articular tumors including synovial hemangioma and D-TSGCT in the future, we will obtain MRI findings under uniform conditions for the purpose of increasing the number of cases for an additional review.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Authors' contributions

TS and MH participated in the design of study, interpreted clinical data and drafted the manuscript. YK and HY participated in the design of the clinical part of this study and provided clinical data. OH analyzed and contributed to the interpretation of the radiological findings. SY analyzed and contributed to the interpretation of the pathological findings. SK participated in the design of study and supervised the project. MH and SK confirm the authenticity of all the raw data. All authors read and approved the final manuscript.

Ethics approval and consent to participate

This retrospective chart review study involving human participants was in accordance with the ethical standards of the institutional and national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. This study was approved by the Ethical Review Committee of Fukushima Medical University (approval no. 2022-36).

Patient consent for publication

This study is retrospective study. Patients were not required to give informed consent to the study because the analysis used anonymous clinical data that were obtained after each patient agreed to treatment by written consent. We applied Opt-out method to obtain consent on this study. The Opt-out was approved by the Ethical Review Committee of Fukushima Medical University.

Competing interests

The authors declare that they have no competing interests.

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