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Tenosynovial Giant Cell Tumor (TSGCT) of the hip: MRI accuracy and results of surgical treatment

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Tenosynovial Giant Cell Tumor (TSGCT) or formerly pigmented villonodular synovitis (PVNS) is a rare nonmalignant tumor of the synovia seldom affecting the hip. MRI and surgical resection are the gold standards in its diagnosis and treatment. However, the accuracy of MRI is unknown, and only few reports on its surgical treatment results exist. The goal of the study was to investigate the MRI accuracy, results after surgical treatment, and natural history of untreated MRI-diagnosed hip TSGCT. Twenty-four consecutive patients with suspected TSGCT on hip MRI, between December 2006 and January 2018, were identified from our medical database. Six refused to participate. About 18 patients with a minimal follow-up of 18 months were enrolled. Charts were reviewed for histopathology results, specific treatment and recurrence. At the last follow-up, all patients had a clinical (Harris Hip Score [HHS]) and radiological examination (x-ray and MRI). Out of 18 patients with suspected TSGCT on MRI, with a mean age of 35y (range 17-52), 14 had surgical resection and 4 refused surgery 1 of whom had a CT-guided biopsy. Out of 15 cases with biopsies, in 10 TSGCT was confirmed. Three surgically-treated patients showed recurrence on MRI after 24, 31 and 43 months. Two non-treated patients showed progression after 18 and 116 months. At the last follow-up (65 m; range 18-159), the mean HHS with or without recurrence was 90 and 80pts (ns). Operative vs. non-operative treatment showed HHS of 86 and 90pts (ns). In the conservatively-treated group, HHS with and without progression was 98 and 82pts (ns), respectively. MRI-suspected TSGCT of the hip was confirmed with biopsy in two-thirds of the cases. Surgical treatment showed recurrence in more than one-third of the patients. Two out of four untreated patients showed progression of the TSGCT-suspected lesion.

Keywords: tenosynovial giant cell tumor; pigmented villonodular synovitis; MRI accuracy; tumor of the hip joint.

INTRODUCTION

PVNS or formerly TSGCT is a rare disorder affecting joints, bursae, or tendon sheaths[1]. Older studies reported an incidence of 1.8 per million[1,2] while newer data showed a higher incidence rate of 40 per million[3]. The typical age of patients with a TSGCT is 40-60 years with women being more often affected[2,3]. No other risk factors are mentioned in the literature.

Pigmented villonodular synovitis (PVNS) was first described in 1852 by Chassaignac as a nodular lesion of the synovial membrane that affected the tendon sheet of the finger[4]. The first histopathological description of PVNS was by Jaffe et al. in 1941[5]. They separated it into pigmented villonodular synovitis (PVNS), pigmented villo-nodular bursitis (PVNB) and pigmented villonodular tenosynovitis (PVNTS)[5,6]. World Health Organisation (WHO) then summarized PVNB and PVNTS as a giant cell tumor of the tendon sheath (GCTTS)[6]. Newer nomenclature describes both PVNS and GCTTS as tenosynovial giant cell tumors (TSGCT)[7]. TSGCT may be intra- or extra-articular and is classified by its macroscopic spreading as localized or diffuse type[7]. While the localized type is more common on small joints of the hand and foot, the diffuse type is more often seen at the larger joints such as the knee and hip while overall the knee is the most frequently affected joint in 75-80% and the hip the second-most at 10–15%[1,8,9]. Histologically, the diffuse and the localized types are essentially the same. Both show the typical giant cells, xanthoma-cells, and a rich mono-nuclear cell-stroma[2], but the diffuse type seems to be clinically more aggressive and shows a higher recurrence rate[3], which also explains the more difficult therapy when the whole synovia is affected. The exact pathomechanism is still unclear and both inflammatory[10] and neoplastic origins are discussed. However, the latest studies seem to favor neoplastic origin[11]. West et al. proposed that

TSGCT is a tumoral process comprising mono- and multi-nuclear cells; translocation involving locus 1p 13 is detected in most TSGCTs, in a small proportion of cells (2-16%) with hyperexpression of CFS1. These tumor cells recruit macrophages bearing CFS1R receptors, differentiate into multinuclear cells and create the aggressive multinuclear "landscape" of TSGCT[7,11]. Despite its probably neoplastic process, metastatic disease is extremely rare[12]. More often than to a metastatic disease, an untreated TSGCT leads to cartilage degeneration and finally osteoarthritis[2]. Bony involvement with osteochondral lesions in 66-89% at the time of diagnosis is significantly more frequent in the hip than in the knee, where bony integrity and joint space are often preserved[13,14]. Nevertheless, reports about TSGCT in the hip are much rarer than in the knee. The largest reported series with TSGCT at the hip was published by Vastel et al. with 16 patients[15].

The clinical diagnosis and conventional radiography of a TSGCT are unspecific, and therefore the non-invasive diagnostic gold standard is the MRI[16]. Typical MRI findings are nodular synovial proliferation and low-signal intensity in T1 and T2, corresponding to hemosiderin deposits and revealing the "blooming artefact" resulting from the magnetic susceptibility of hemosiderin.

It is seen on T2* weighted gradient-echo sequences, a phenomenon not as appreciable in differential lesions such as synovial chondromatosis[17]. The invasive gold standard in the diagnostics of TSGCT is histology[2]. To our knowledge, there is one study about the value of the MRI in TSGCT of the knee[8], but no corresponding study on the hip.

The treatment of a TSGCT of the hip is either joint-preserving surgery with open or arthroscopic excision of a localized type or additionally total synovectomy in diffuse type[15,18] or open excision of the lesion, synovectomy. Finally, arthroplasty[9,15,19] is a surgical option in the presence of osteoarthritis.

Independent of the joint involved, excision of the lesion alone shows a higher recurrence rate than with an additional synovectomy[20]. Additionally, radiation after surgery is used by some authors[2,21] but with very low evidence on its effect on recurrence rate[22].

There is a lack of knowledge regarding the natural course of TSGCT.

The aim of this study was to investigate MRI accuracy, results after surgical treatment and natural history of untreated MRI-diagnosed TSGCT.

MATERIAL AND METHODS

Twenty-four consecutive patients with suspected TSGCT on MRI of the hip, between December 2006 and January 2018, were identified from our medical database. Six refused participation, and their data were not evaluated. About 18 patients with a minimal follow-up of 18 months were enrolled and seen for a follow-up appointment.

Chart review for Harris Hip Score (HHS) on the affected and the contralateral side was evaluated, and an X-ray AP pelvis and lateral cross-table view as well as an MRI of the affected hip were performed.

X-ray at the time of diagnosis and latest follow-up for evaluation of osteoarthritis according to Kellgren/Lawrence[23].

MR images at the time of diagnosis, suspecting the presence of TSGCT, and at follow-up were analyzed by a subspecialized MSK radiologist (L.F., 8 years of experience). At follow-up, MR imaging was performed with a 3 Tesla MRI (MAGNETOM Prisma, Siemens Healthcare). The imaging protocol included T1- and PD-weighted sequences in both the coronal and sagittal plane as well as an axial T2-weighted 3D DESS (double echo steady-state) sequence.

Lesions suspicious for TSGCT were assessed for subtype (diffuse vs. localized).

Kolmogorov-Smirnov test was employed to determine normally and abnormally-distributed data. Normally distributed data are displayed as mean and standard deviation, while abnormally-distributed data are displayed as median and interquartile range. Independent variables were compared with Mann-Whitney-U-Test, while Wilcoxon signed-rank test was used to compare two related samples. The analysis was conducted using IBM SPSS Statistics software version 23.0.0. The level of significance was set to $p < 0.05$.

RESULTS

The results are summarized in Table I.

The mean age overall was 35y with range 17-52, while it was 32y with range 17-47 in the surgical group. Overall, 10 (56%) were women and 8 (44%) were men, while they were 10 (71%) and 4 (29%), respectively, in the surgical group.

About 14 of the 18 patients underwent surgery, while three refused it or further diagnosis, and one had a CT-guided biopsy (Fig.1). Of the 14 surgically treated patients, seven underwent arthroscopic surgery, four a surgical hip dislocation, two open surgery by an anterior minimal invasive approach and one a total hip

Table I. — Distribution of the 18 patients with a suspected TSGCT in the MRI

	case	Sex	Age (y)	fu (mts)	Initial MRI	Type of TSGCT on MRI	Recurrence/ Progression	HHS initial (pts)	HHS fu (pts)	Radia-tion	Histo TSGGT yes/no	Arthritis Kellgren/ Lawrence
	Surgical treatment											
A	1	m	35	42	n*	local	no	71	100	no	no	0
	2	f	30	64	iv*	local	no	na	88	yes	yes	0
	3	f	34	159	a*	diffuse	no	96	99	no	no	4
	4	m	17	86	a*	local	no	95	73	no	no	2
	5	f	25	59	iv plus*	local	no	61	97	yes	yes	0
	6	f	19	43	iv plus*	local	yes	76	84	no	yes	0
	7	m	41	117	a*	local	no	na	100	no	no	0
B	8	f	27	23	iv plus*	local	no	88	100	no	yes	0
	9	m	27	93	a*	diffuse	yes	60	53	no	yes	2
	10	f	23	31	iv*	local	yes	96	96	no	yes	0
	11	f	30	18	iv*	local	no	96	96	no	yes	0
C	12	f	47	135	a*	local	no	49	63	no	yes	0
	13	f	44	33	iv*	local	no	na	74	no	yes	0
D	14	f	47	42	a*	diffuse	no	53	86	no	no	2
	Conservative treatment											
E	15	m	51	116	iv*	local	yes	x	78	no	yes	0
F	16	m	30	25	a*	local	no	x	96	no	no	0
	17	m	52	57	a*	local	no	x	86	no	no	0
	18	m	46	18	a*	local	yes	x	100	no	no	0

A= arthroscopy, B= surgical hip dislocation, C= open surgery direct anterior or posterior approach, D= Total Hip Arthroplasty (THA), E= CT-Biopsy, F= no further diagnostic. X= no baseline HHS available, patient nr. 9 received THA 30mts after initial surgery. *MRI-Protocol: n= native, a=Arthro-MRI, iv= intravenous contrast medium, iv plus= Arthro-MRI and intravenous contrast medium.

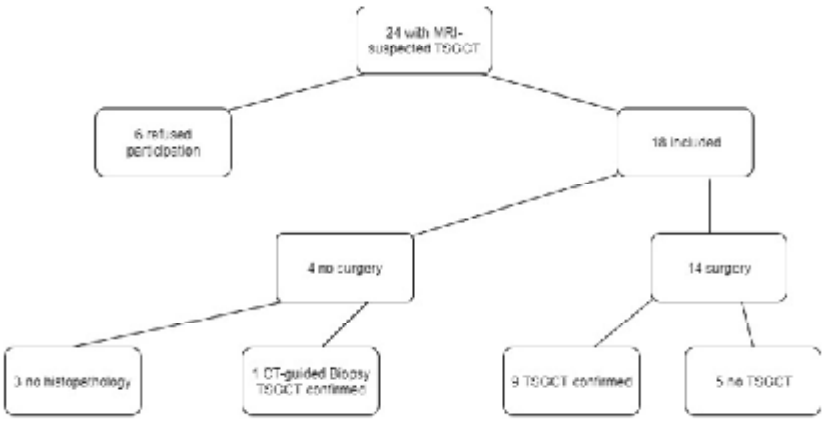

Fig. 1. — Shows 24 consecutive patients with TSGCT-suspected lesion in the MRI between 2006-2018.

arthroplasty (THA). Only two of the surgical patients received hip radiation in the postoperative course. Of the suspected TSGCT in the MRI, 13 were classified as localized type and three as diffuse type, while two were localized extraarticularly posterior to the greater trochanter between the external rotators and the M. gluteus maximus as shown in Figure 2 and 3.

Out of 15 cases with biopsies (14 surgeries and 1 CT-guided biopsy), in 10, TSGCT was histologically confirmed. In the non-confirmed cases, the synovia of the biopsy showed only unspecific signs of inflammation.

Out of ten histologically confirmed TSGCT, nine underwent surgery.

Three showed recurrence on MRI (24, 31 and 43 months postoperatively), one after arthroscopy and two after surgical hip dislocation.

One of the patients demonstrating a recurrence received THA 30 months after the surgical hip dislocation because of residual pain, relapse of the TSGCT and osteoarthritis. At the final follow-up, five years after the THA, the MRI shows no recurrence of the TSGCT.

Out of the four patients without surgery, two showed progression: one with the positive CT-guided biopsy after 15 months and the second on the follow-up MRI after 18 months.

Baseline HHS was available for 11/18 patients with a median of 76 (IQR 60-76; range 49-98). At

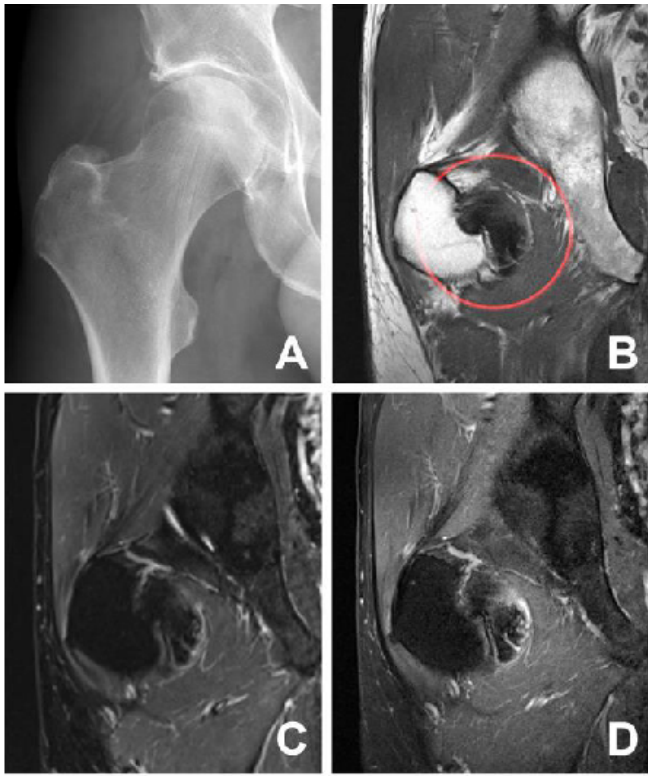

Fig. 2. — 52-year-old male patient with biopsy-proven focal TSGCT medial to the greater trochanter. Anteroposterior x-ray (A) shows mild contour irregularity of the medial aspect of the lesser trochanter. Coronal T1 (B), STIR (C) and post-contrast T1 fat sat (D) reveal a lobulated hypointense mass with heterogeneous peripheral enhancement and erosion of the adjacent lesser trochanter. The hypointense signal is related to hemosiderin deposition within the mass.

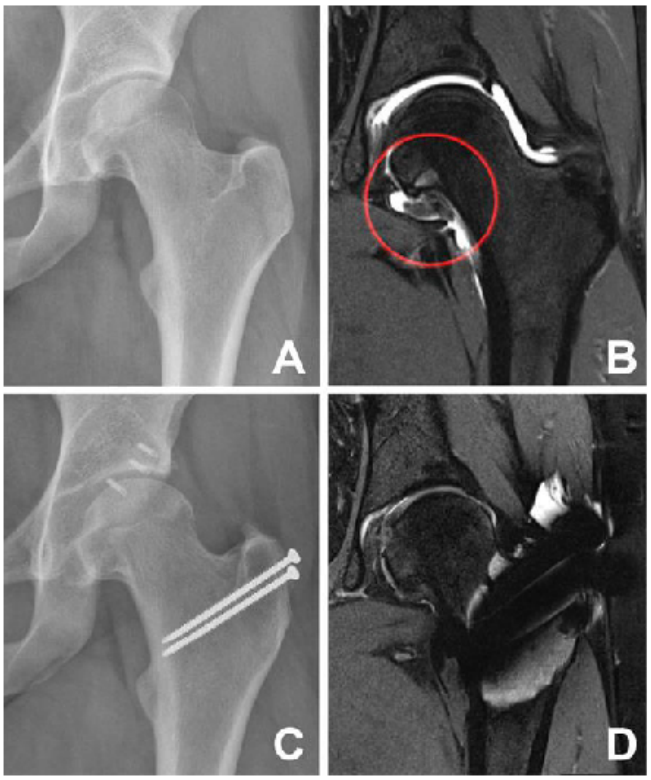

Figure 3. — 25-year-old female patient with femoroacetabular impingement and concomitant biopsy-proven focal TSGCT inferior to the femoral head. Preoperative ap view (A) and MR arthrography (B) show a hypointense mass with erosion of the adjacent femoral head. Postoperative x-ray (C) and MRI (D) after tumor resection, trochanteric osteotomy, acetabular trimming and osteochondroplasty show partial bony healing of the inferior femoral head.

the latest follow-up (65m; range 18-159), the mean HHS with or without recurrence was 90 pts and 80 pts (p=0.91), respectively. Patients with confirmed TSGCT versus non-confirmed TSGCT showed no significant difference (p=0.9) for the HHS with 83 pts versus 92 pts, respectively.

DISCUSSION

In our cohort, MRI-suspected TSGCT of the hip was histopathologically confirmed in 66% of the cases, which is lower than in the study of Barile et al. who reported about 84% of the MRI-suspected cases being confirmed at the knee joint[8]. Nevertheless, despite this unsatisfactory accuracy, MRI remains the gold standard for non-invasive diagnostics for detecting a TSGCT[24,25]. In view of the fact that most symptomatic patients in the MRI-suspected TSGCT in the hip undergo surgery, they should be informed about the one-third false-positive MRI diagnosis of TSGCT. Therefore, watchful waiting may be considered. In our cohort, out of the four patients with suspicion of TSGCT but on non-operative treatment, one TSGCT was histologically confirmed by biopsy. At the last follow up, in two of these four patients, a progression of the lesion was seen on MRI. However, all of these three patients showed very good clinical results at the follow-up, independent of progression or not.

The recurrence rate of 33% after surgical treatment in our cohort is in accordance with the current literature of TSGCT of the hip and knee[3,13,18,21] which described the rate of XY up to 30%. However, contrary to other studies, we had fewer cases (n=3) in which, on the MRI, a diffuse type of TSGCT was suspected and finally confirmed by biopsy (n=1). Therefore, it remains unclear if the diffuse type may be associated with a more aggressive course of the disease and with a higher recurrence rate when compared to the localized type of TSGCT.

The clinical results of this study are comparable with other studies[9,13] which showed similar HHS of 91 and 94 points, respectively. Moreover, the overall

clinical outcome was good irrespective of recurrence or progression of the lesion in non-operated patients after a minimum follow-up of 18 months.

CONCLUSION

MRI-suspected TSGCT of the hip was confirmed with biopsy in two-thirds of the cases. Surgical treatment showed recurrence in more than one-third of the patients. Two out of four untreated patients showed progression of the TSGCT-suspected lesion.

Conflict of Interest: None.

Funding: None.

Ethical approval: The responsible Ethical committee of Zurich (2018-01468) approved the acquisition of the data.

Informed consent: Patients were only included when informed consent was available

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