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ORIGINAL STUDY

Tenosynovial giant cell tumor of the pes anserinus bursa with secondary involvement of a reconstructed autologous anterior cruciate ligament – A case report

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Tenosynovial giant cell tumor (TGCT) is defined by the World Health Organization (WHO) as a family of lesions most often arising from the synovium of joints, bursae and tendon sheaths. It is composed of synovial-like mononuclear cells, admixed with multinucleate giant cells, foam cells, siderophages and inflammatory cells *(1).* It can have various clinical manifestations, and is therefore subdivided in a diffuse and a localized/nodular subtype. Furthermore, the lesions can have an intra- or extra-articular location.

The purpose of this paper is to present the case of a 41-year-old male suffering from multifocal extra- and intra-articular TGCT of the right knee, with involvement of the pes anserinus bursa and an anterior cruciate ligament (ACL) autograft respectively. The ACL reconstruction was performed 11 years prior to the diagnosis of the TGCT, using tendons harvested from the pes anserinus.

Our case illustrates the risk of transferring TGCT from an extra- to intra-articular location during ACL reconstruction, when using tendons of a pes anserinus prone to develop this condition. To our knowledge, no similar case was published in the literature so far.

Keywords: Tenosynovial giant cell tumor; anterior cruciate ligament; ACL reconstruction; pes anserinus; case study; and pigmented villonodular synovitis.

INTRODUCTION

TGCT is a benign soft tissue proliferation arising from synovium. The most significant contribution to the understanding of these lesions was made by Jaffe et al. *(2)*, who regarded the synovial lining of a joint, tendon sheath or bursa as one anatomic unit, that can give rise to a heterogeneous group of lesions, that can thus have both intra- and/or extra-articular locations. The WHO divides TGCT into two main subtypes: a localized/nodular type and a diffuse type *(1).*

The etiology and histogenesis are still not completely understood: whereas TGCT used to be regarded as a reactive and inflammatory process, nowadays there is more evidence (e.g. by identification of clonal chromosomal aberrations) that the condition is of neoplastic nature *(3).*

Clinical symptoms mostly depend on the lesion's location. Intra-articular lesions often cause pain and swelling of the affected joint due to repetitive intra-articular bleeding (hemarthrosis), and sometimes articular dysfunction. Extra-articular lesions generally cause fewer symptoms, but can present with a soft-tissue mass or pain *(4).*

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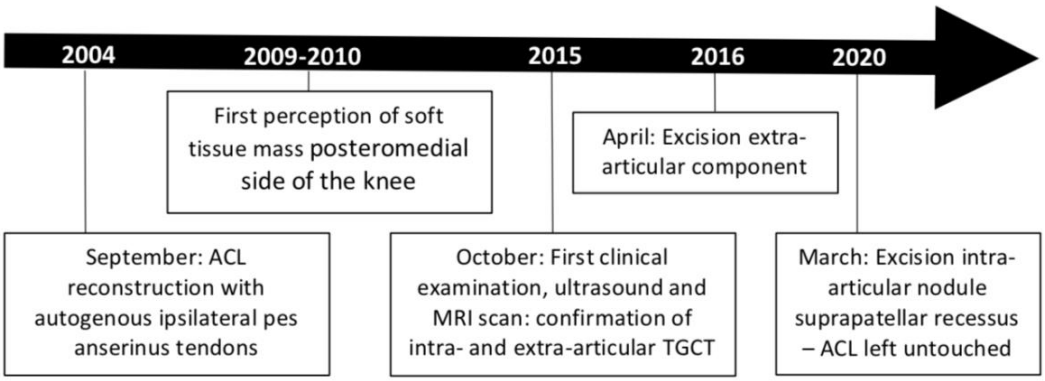

Figure 1. — Timeline with subsequent events.

On macroscopic examination, TGCT presents as a villous or nodular proliferation and is recognized by its red-brownish color, with distinct (ochre) yellow patches. Microscopically, it contains multi-nucleated giant cells and stroma. The macroscopic colors can be attributed to the presence of xanthoma cells/foamy histiocytes (in the yellow areas) and hemosiderin pigment (giving TGCT its characteristic "rust" color) *(1,4)*.

Radiographs of affected joints may appear normal, but there can also be joint effusion, non-mineralized soft tissue swelling and signs of extrinsic cortical erosion. On MRI (Magnetic Resonance Imaging), the signal intensity is low on T1- as well as T2-weighted images, and even more so on gradient-echo images. This "blooming"-effect is a magnetic susceptibility artifact, caused by the presence of hemosiderin and is nearly pathognomonic of TGCT *(4)*.

We present the case of a 41-year-old male suffering from multifocal extra- and intra-articular TGCT of the right knee, with involvement of the pes anserinus bursa and an ACL autograft respectively.

To our knowledge, our case is the first extra-articular TGCT (of the medial hamstring insertion) to be reported of having led to involvement of an ACL autograft, that was reconstructed from the ipsilateral tendons of the semitendinosus and gracilis muscle.

CASE REPORT

In September 2004, an arthroscopic reconstruction was performed of the ACL in the right knee of a male patient, 30 years of age at the time. He ruptured this ACL 4 years prior to the surgery, playing soccer. An autogenous graft was used, harvesting the tendons of the semitendinosus and gracilis muscle from the pes anserinus of the ipsilateral knee. Intra-operatively, these tendons showed no macroscopic abnormalities. Postoperative follow-up was satisfactory: patient had no functional restrictions and clinical examination showed no swelling of the joint nor laxity of the ACL-graft.

A timeline of the subsequent events is given in Figure 1. In October 2015, patient was referred at age 41 to our orthopaedic oncology clinic, with a soft-tissue mass on the posteromedial side of the right knee and proximal lower leg. This mass was first noticed 6 years earlier and did originally give no complaints. However, the volume increased progressively and started to bother him during physical activities, especially with flexion of the knee.

An ultrasound scan showed a non-vascularized mass, with a maximal diameter of 7.5 cm. MRI confirmed the presence of a heterogeneous mass involving the area of the former pes anserinus and extending (possibly in a bursa) along the tendon of the semimembranosus muscle, which was – by its very low signal on T2-weighted images and its only minimal contrast enhancement – suggestive for a TGCT (Figure 2). Moreover, similar signal changes were noted along the ACL-graft in its course through the (widened) femoral and tibial tunnels (Figure 3) and a nodular lesion with the same characteristics was found in the suprapatellar recessus, at the exact location of the button for fixation of the ACL-graft

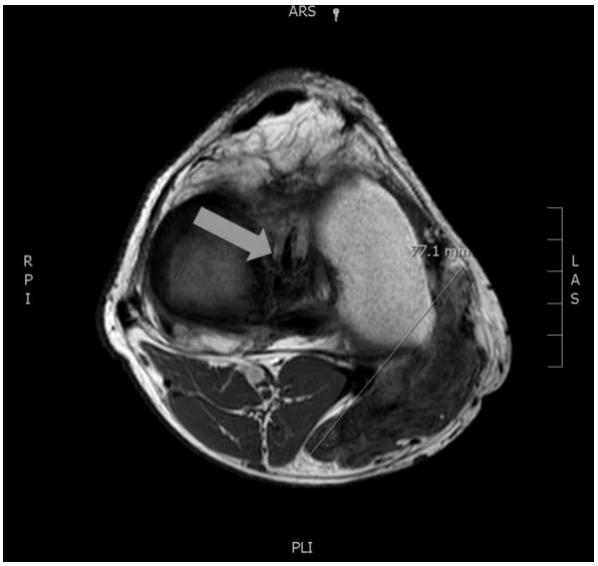

Figure 2. — Axial MRI (T1-weighted, with contrast enhancement), showing the pre-operative extra-articular mass on the posteromedial side of the knee, with a maximum diameter of 7.5cm. Note (arrow) the same low signal changes of the ACL graft in the intercondylar region. (October 2015).

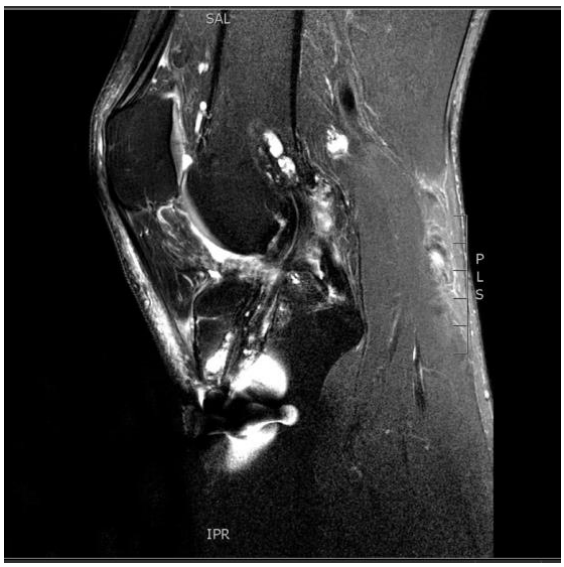

Figure 3. — Sagittal MRI (T2-weighted, with fat suppression), showing the course of the clearly affected ACL graft through the tibial and femoral tunnels, with metal artefacts surrounding the tibial staple. (October 2015).

(Figure 4 and 5).

As symptoms were progressive and the volume of the tumor was increasing, the extra-articular mass was marginally excised in April 2016, without opening the joint. The mass proved to be stuck to the semimembranosus tendon. The pathology report confirmed the tissue to be compatible with TGCT.

During surgery for (keloid) scar revision in March 2020, the nodule in the suprapatellar recessus was resected through a small direct arthrotomy, upon the patient's request. Again, the pathology report confirmed the diagnosis of TGCT.

On the latest pre-operative MRI (March 2020), this nodule was non-progressive compared to the imaging in 2015. There were no signs of local

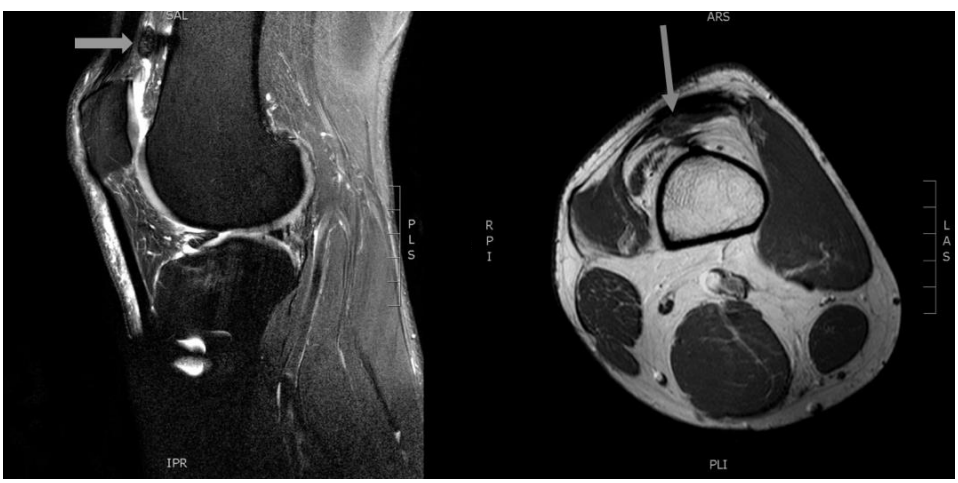

Figure 4. — Sagittal and axial MRI (T2-weighted, with fat suppression and T1-weighted, with contrast enhancement respectively), with the arrow indicating the PVNS nodule in the suprapatellar recessus. Note the subtle metal artefact of the underlying button. (October 2015).

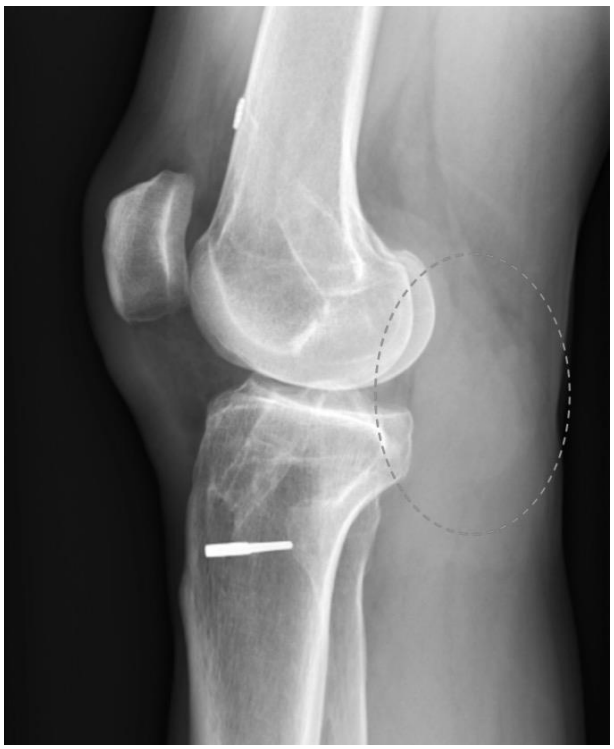

Figure 5. — Lateral radiograph, showing the tibial staple and femoral button. The non-mineralized shadow of the soft tissue mass is delineated by a dotted line. (January 2016).

recurrence along the semimembranosus tendon and the affected ACL-graft showed no progressive disease.

The patient remains asymptomatic and fully functional, with the knee being stable at the latest follow-up in April 2020. A written informed consent was obtained from the patient.

DISCUSSION

In an attempt to explain the findings in our case, it became clear that using the correct terminology is paramount, when talking about the various manifestations of TGCT (Table I).

The most significant contribution to the understanding of these lesions was made by Jaffe et al., who regarded the synovium of the joint, the tendon sheath and the bursa as one anatomic unit, that can give rise to a common family of lesions, with intra- and/or extra-articular location *(2).* Nowadays, the WHO divides TGCT into two main subtypes: a localized/nodular type (also known as giant cell tumor of tendon sheath) and a diffuse type (the so-called pigmented villonodular synovitis or PVNS) *(1).*

The most frequent and well-known presentation of TGCT is PVNS, the diffuse intra-articular form. It is predominantly affecting large weight-bearing joints, with a predilection (80%) for the knee joint. Less frequently but still regularly encountered, is the localized/nodular extra-articular form, mostly involving the digits (the palmar side of fingers and toes in particular) *(5).*

A search through current literature revealed that also the diffuse extra-articular form (pigmented villonodular bursitis) *(6,7)* and the localized/nodular intra-articular form of the disease exist *(5,8-11)* but both these forms are very rare and described in the literature mainly as case reports.

Several articles describe some sort of link between TGCT and the ACL, but they all highlight different aspects: an intra-articular lesion of the nodular subtype arising from the ACL *(5,8,9)* or the ligamentum mucosum *(11),* the bilateral presence of a nodular subtype lesion in association with native ACL failure *(10)*, a knee with prior ACL reconstruction with an allograft *(12)* or an artificial ligament *(13)* getting affected by PVNS, and the extra-articular reconstruction of a ruptured ACL in case of pre-existing PVNS *(14)*.

Table I.

Tenosynovial GCT	Diffuse	Localized/Nodular
Intra-articular	PVNSynovitis Most commonly (80%) knee	Tenosynovial GCT e.g. ACL
Extra-articular	PVNBursitis e.g. pes anserinus bursa	Tenosynovial GCT Most commonly fingers/toes

No reports were found, however, of the very particular situation in our case, with selective affection of an ACL autograft reconstructed from tendons, with concomitant PVNBursitis at their donor site.

We only came across one report in the literature on the simultaneous presence of an intra- and extra-articular lesion at the level of the knee *(15)*. Whereas these authors stress the co-incidence that both lesions seemed to be anatomically completely unrelated, we believe that the intra-articular lesion in our case, was secondary and clearly subject to the same pathogenic condition that affected the extra-articular location in the first place.

CONCLUSION

In our case of an affected ACL graft, we don't believe that it is a diffuse PVNS of the knee joint that has affected the reconstructed ACL, as the rest of the joint seems to be unaffected on MRI. However, there is a second lesion right at the exit point of the femoral tunnel, but its presence can be simply explained by the surgical technique with a button.

A localized/nodular intra-articular form of TGCT is very unlikely as well, as our patient doesn't have a pedunculated lesion arising from the ACL, but a very diffuse infiltration of the whole autograft, with widening of the tibial and femoral bone tunnels.

Furthermore, both suggestions would still not explain the synchronous extra-articular localization in the pes anserinus bursa.

We are convinced that there was already a propensity for the diffuse extra-articular form in the pes anserinus bursa at the time of graft harvesting, as this provides us with a logical explanation for the later development of a PVNBursitis in the m. semimembranosus bursa as well as involvement of the ACL graft, reconstructed from the m. gracilis and m. semitendinosus tendons. Furthermore, the pathogenesis of the prefemoral second intra-articular nodule - as a focal deposit at the end of the femoral tunnel – is explained.

To our knowledge, no similar case has been described in the literature, so we are the first to suggest that PVNBursitis of the pes anserinus can lead to secondary involvement of an ACL autograft, when the graft is reconstructed from the pes anserinus tendons.

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