CASE REPORT

Pain and fracture after anterior cruciate ligament reconstruction caused by giant cell tumour of the distal femur

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SUMMARY

The causes of pain after an anterior cruciate ligament (ACL) reconstruction are numerous and may have complex origins. We present an unusual case in which pain after an ACL reconstruction developed secondary to a giant cell tumour of the bone occurring around a fixation screw in the distal femur, with an associated fracture through the femoral tunnel of a previously well-functioning reconstruction. We discuss the aetiology and the treatment of a complex clinical scenario.

BACKGROUND

Orthopaedic implants have featured heavily in the literature over the last few years with the uncovering of problems related to certain metal on metal prostheses and subsequent concern regarding their oncogenic potential.¹⁻³ We present this unique case which reinforces the importance of thorough history and examination when faced with any

patient, and also review the literature on potential links between orthopaedic implants and the development of tumours.

CASE PRESENTATION

A fit and well, previously asymptomatic, 25-year-old man was seen in the fracture clinic. He had been running with his dog when his knee gave way and he fell. He had undergone a four stranded hamstring anterior cruciate ligament (ACL) reconstruction of that knee 7 years prior and had made a full recovery. A clinical diagnosis of patellar dislocation was made in accident and emergency, and an unsuccessful manipulation attempted under sedation. He was then sent home in a splint for follow-up in the fracture clinic.

INVESTIGATIONS

At presentation, he was unable to weight bear on the affected limb. The foot had normal

Figure 1 X-rays at presentation showing an aggressive lesion in the lateral femoral condyle with an associated pathological fracture. The fixation screws from the cruciate ligament reconstruction are also observed.

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Figure 2 Coronal and sagittal plane CT reconstructions of the distal femur showing a large lytic tumour with a permeative margin extending up to the subarticular surface of the joint.

neurovascular status. An X-ray of his right knee (anterioposterior and lateral views) demonstrated a pathological fracture of the lateral condyle of the right femur through a large aggressive lytic lesion (see figure 1).

A CT scan demonstrated an eccentric lytic lesion in the epiphysis of the lateral femoral condyle extending into the metaphysis (see figures 2 and 3). The lesion extended up to the subarticular region and was scalloping the cortex of the femur. There was no soft tissue mass or periosteal reaction. There was a small intra-articular step in the knee joint.

The patient was therefore referred to the North of England Bone and Soft Tissue Tumour Service for investigation, biopsy and further management. Biopsy confirmed giant cell tumour of bone and showed haemosiderin deposition indicating previous haemorrhage (figure 4). Blood tests excluded hypercalcaemia and the alternative diagnosis of brown tumour of hyperparathyroidism.

DIFFERENTIAL DIAGNOSIS

The clinical differential diagnosis included benign tumours such as aneurysmal bone cyst, malignant tumours such as telangiectatic osteosarcoma, osteolysis around the ACL screw and infection. Other conditions which share some or all of the histological features of a giant cell tumour of bone include brown tumour of hyperparathyroidism, aneurysmal bone cyst, chondroblastoma or giant cell-rich osteosarcoma.

TREATMENT

Our patient was a young, previously fit and well adult so it was felt that joint preservation was of paramount importance. He was therefore treated with aggressive curettage of the tumour, high-speed burring, argon beam diathermy and insertion of

polymethylmethacrylate bone cement. The screw was readily removed from the tumour. The reconstruction was protected with a contoured distal femoral locking plate to provide immediate stability and allow quicker return to function.⁴ Interestingly the ACL reconstruction appeared to be clinically functional after surgery.

Although surgery remains the mainstay of treatment for giant cell tumour of the bone, the availability of denosumab, a

Figure 3 CT reconstructions showing a comminuted fracture of the lateral femoral condyle.

monoclonal antibody that inhibits RANK ligand is opening up new approaches to treatment, particularly in unresectable or metastatic disease.⁵ RANK ligand is key in the recruitment and transformation of circulating monocytes into the osteoclasts that appear as the giant cells characteristic of this tumour.

OUTCOME AND FOLLOW-UP

Rehabilitation was uneventful and when last reviewed after 18 months the patient had minimal discomfort, was walking normally, had flexion from 0 to 110° with a stable knee. X-rays show a stable fixation (figure 5). He is currently considering a phased return to low-impact sports. He is at risk of developing early degenerative change in the knee.

DISCUSSION

The authors were unable to find any previous cases of giant cell tumour causing collapse and pathological fracture of the femoral tunnel following an ACL reconstruction. There is, however, one case of giant cell tumour of the proximal tibia occurring in a 52-year-old man 13 years after a bone-patella-bone ACL reconstruction.⁶ Unlike our case, the patient reported of increasing pain in the knee and the tumour was diagnosed prior to fracture. In our case there was no lesion present prior to the ACL reconstruction and follow-up X-rays demonstrated no identifiable lesion. The patient was completely asymptomatic prior to the fall.

There are case reports of malignant tumours presenting after ACL reconstruction: Efe *et al*⁷ reported a case of malignant fibrous histiocytoma of the distal femur 40 months following ACL reconstruction. This was treated with neoadjuvant chemotherapy, wide tumour resection and distal femur reconstruction using a silver-coated non-cemented tumour knee joint prosthesis. Sirveaux *et al*⁸ also reported a pleomorphic malignant fibrous histiocytoma of the 'medial soft tissue' of the operated knee joint in a 19-year-old male patient 6 years after ACL reconstruction. Interestingly at the time of surgery suture and metal particles were identified in the sarcoma, hence the authors suspected a connection to the initial ACL reconstruction. We did not find any metal debris in this tumour specimen. Caron *et al*⁹ also reported a leiomyosarcoma in the distal femur of a patient 12 years after ACL reconstruction.

There is also literature documenting the development of sarcomas next to orthopaedic implants including total hip

Figure 5 Postoperative X-ray showing reconstruction using contoured locking plate and methylmethacrylate cement.

replacements, intramedullary nails and osteotomy staples; however, the rarity of these cases makes cause and effect hypotheses debatable.¹⁰ Other factors that may predispose to the formation of giant cell tumour of bone include hypoxia¹¹ and it is possible that the presence of a screw or ligament graft in the distal femoral metaphysis was responsible for generating a locally hypoxic environment, but this is speculative.

We present the first reported case of pathological fracture through the femoral bone tunnel secondary to giant cell tumour of bone following an ACL reconstruction. While it is difficult to demonstrate a direct relationship between the two, it is rational to document these cases in the literature in order to contribute to the monitoring of any potential association between tumour development and orthopaedic implants. Furthermore, it is important to be aware that pain after an ACL reconstruction may have an unexpected cause.

Figure 4 H&E photomicrograph showing numerous multinucleated osteoclast-like giant cells on a background of mononuclear stromal cells.

Learning points

- ▶ Sudden onset of pain and deformity in a limb requires an X-ray on presentation or postmanipulation.
- ▶ Reporting of any cases involving pathological lesions around implants is of paramount importance to ensure they are retrievable in the literature, to further our understanding of any potential associations.
- ▶ Any new pathological lesion in the musculoskeletal system must be investigated appropriately and possibly discussed with the local tumour services.

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REFERENCES

- 1 Noonan B, Chung KS. A practical review of the mechanisms of pain and pain management following ACL reconstruction. *Orthopedics* 2006;29:999–1005.
- 2 Mäkelä KT, Visuri T, Pulkkinen P, *et al*. Risk of cancer with metal-on-metal hip replacements: population based study. *BMJ* 2012;345:e4646.
- 3 Wagner P, Olsson H, Ranstam J, *et al*. Metal-on-metal joint bearings and hematopoietic malignancy. *Acta Orthop* 2012;83:553–8.
- 4 Ayerza MA, Aponte-Tinco LA, Farfalli GL, *et al*. Joint preservation after extensive curettage of knee giant cell tumours. *Clin Orthop Relat Res* 2009;467:2845–51.
- 5 Chawla S, Henshaw R, Seeger L, *et al*. Safety and efficacy of denosumab for adults and skeletally mature adolescents with giant cell tumour of bone: interim analysis of an open-label, parallel-group, phase 2 study. *Lancet Oncol* 2013;14:901–8.
- 6 Fitzsimmons SE, Chinitz N, Glashow J. Giant cell tumour at tibial screw site after anterior cruciate ligament reconstruction. *Am J Orthop* 2010;39:E54–6.
- 7 Efe T, Heyse TJ, Schofer MD, *et al*. Malignant fibrous histiocytoma of the distal femur after an arthroscopic anterior cruciate ligament reconstruction: A case report and review of the literature. *BMC Cancer* 2010;10:264.
- 8 Sirveaux F, Hummer N, Roche O, *et al*. Pleomorphic malignant fibrous histiocytoma at the site of an arthroscopic reconstruction of the anterior cruciate ligament. A case report. *J Bone and joint Surg [Am]* 2005;87:404–9.
- 9 Caron JJ, Pambuccain SE, Steen JT, *et al*. Leiomyosarcoma of the distal femur after anterior cruciate ligament reconstruction. *Clin Orthop Relat Res* 2004;214–17.
- 10 Keel SB, Jaffe KA, Petur Nielsen G, *et al*. Orthopaedic implant-related sarcoma: a study of twelve cases. *Mod Pathol* 2001;14:969–77.
- 11 Knowles HJ, Athanasou NA. Hypoxia-inducible factor is expressed in giant cell tumour of bone and mediates paracrine effects of hypoxia on monocyte-osteoclast differentiation via induction of VEGF. *J Pathol* 2008;215:56–66.

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