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## Graft of allogeneic knee extensor mechanism in treatment of giant cell tumor of the patella

### Przeszczep allogenicznego aparatu wyprostnego stawu kolanowego w leczeniu guza olbrzymiokomórkowego rzepki

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## Summary

**Introduction:**

Cancer of the patella is a rare condition. Giant cell tumor of the patella is an extremely rare and a difficult to solve therapeutic problem. Depending on the degree of bone destruction, it may cause significant knee joint dysfunction. It is assumed that surgery is the treatment of choice.

**Material/Methods:**

This study presents an innovative treatment of a giant cell tumor of the patella in a 40-year-old male with significant dysfunction of the knee extensor mechanism. Complex therapy included neoadjuvant treatment using a human monoclonal antibody IgG<sub>2</sub> that binds to RANKL (Denosumab) and follow-up allogeneic grafting of knee extension mechanism.

**Results:**

Follow-up CTs showed correct positioning and healing of grafted knee extensor mechanism. Early functional outcome was perfect. Seven weeks after the surgery, reduction of pain, improvement of the contour of the joint, full active extension and flexion of 110° were noticed. Fifteen months after the surgery, the patient presented complete active extension and flexion of the knee joint, actively participated in professional life and CT tests did not reveal features of tumor recurrence.

**Conclusions:**

Treatment of isolated giant cell tumor of the patella with destruction of patella and joint dysfunction does not have a clear solution in the available literature. Such a condition may also exclude traditional procedures. In such cases, grafting of the whole extensor mechanism appears to be the appropriate treatment.

**Keywords:**

Giant cell tumor • GCT • Patellar tumor • Knee extensor mechanism reconstruction • Knee extensor mechanism grafting • Allograft of patella

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## INTRODUCTION

Cancer of the patella occurs rarely. Giant cell tumor of bone (GCTB), described by Cooper in 1818 [30], accounts for 1/3 of cases [20]. In 80% of cases they are benign lesions [26], but they may be locally malignant. About 10% of GCTB undergo malignant transformation at recurrence, with the risk of lung metastases at the level of 2–5% [14]. Histologically, GCTB is characterized by a great number of osteoclast-like giant cells and fusiform mononuclear cells [31]. Swelling, tenderness, redness, increased temperature, reduced mobility and pain may be present on physical examination. Radiologically, giant cell tumor causes bone destruction, geographic osteolytic lesions, thinning of cortex, pathologic fractures, and, rarely, sclerotic rims and periosteal reactions [3, 24]. Based on radiographic imaging, GCTB is classified according to the grading system introduced by Campanacci [26].

- Grade 1: well-defined, thinned rim; intact or slightly thinned, undistorted cortex.
- Grade 2: relatively well-defined margin but no rim; cortex and rim of a reactively altered bone are thin and moderately expanded but still present; lesions with a fracture present are classified as a separate subgroup.
- Grade 3: lesion with indistinct border; tumor infiltrates soft tissue.

Core biopsy and open biopsy with a collection of diagnostic material for histopathological examination are recommended in the diagnostic evaluation [8, 25, 30].

GCTB should be differentiated from aneurysmal cyst, brown tumor, chondroblastoma, osteoid osteoma, chondroma, osteochondroma, osteoblastoma, osseous hemangioma, simple bone cyst, osteosarcoma, chondrosarcoma, osseous lymphoma, and malignant fibrous histiocytoma [25, 26].

Giant cell tumor of the patella is a rarely occurring cancer. According to Campanacci, only 126 cases were described in the 20<sup>th</sup> century [7].

Due to the small number of described cases of giant cell tumor of the patella, there is no defined treatment [17].

Surgery is assumed to be the treatment of choice, with the percentage of recurrence varying between 10–50%, depending on the surgical technique [26].

The percentage of recurrence after curettage and subsequent widening of the compartment with high-speed burr ranges from 12–18% [1, 11]. The procedure is supplemented by pulsatile jet-lavage, which additionally cleans the tumor bed out of the cancer cells [9, 29]. Hydrogen peroxide, phenol cryotherapy and liquid nitrogen are used as adjuvant therapy [18, 22, 30]. After the tumor is removed, the tumor bed is filled with allogeneic bone grafts, bone cement (polymethyl methacrylate, PMMA) or bone substitute materials, often containing hydroxyapatite and tricalcium phosphate [26].

It should be stressed that the techniques of curettage involving the use of bone grafts lead to a higher risk of recurrence than in the cases when PMMA is applied [31].

Regardless of the surgical method, the core of effective treatment is proper mechanical removal of the tumor. It is only possible by uncovering the tumor by creating a wide bone window. The window needs to be big enough to provide access at every angle. Only that enables complete mechanical removal of the tumor [26].

In some tumors, especially large ones with extensive deformation, as well as those destroying areas around joints, wide local resection is the treatment of choice. The percentage of recurrence decreases to 0% after wide local resection [15, 19, 25, 30, 32]. Resective surgery, depending on the extent, may cause problems with tissue, anatomical and functional deficits and may require follow-up reconstructive and plastic surgeries [13, 19, 26, 28].

Therefore, prostheses, including megaprotheses, and techniques of biological reconstruction, such as bone grafts, cartilage transplants, vascularized transplantation as well as methods of bone deficiency regeneration with the use of external stabilizers (e.g. Ilizarov apparatus or Taylor Spatial Frame) are used [31].

In recent years, neoadjuvant therapy has been used more often. Anti-RANKL bisphosphonates (Pamidronate and Zoledronic acid) therapy (Denosumab) and preop-

erative embolization are applicable [26]. They are used primarily when surgical treatment is insufficient, difficult or impossible due to the location of the tumor [31].

## MATERIALS AND METHODS

In this study we present a case involving the destruction of the right patella as a result of a giant cell tumor, as well as innovative surgical procedures.

A 40-year-old male after a twisting injury of the right knee joint sought care at an orthopaedic emergency room. The X-ray revealed a slightly altered bone structure of the patella. The patient was informed of the need for further diagnostic evaluation and was treated in the typical manner for a twisting injury of the knee joint. Due to a temporary improvement and the disappearance of complaints, the patient did not return for a follow-up, which significantly delayed the diagnosis. When treatment was taken over by our institution, radiographic examination revealed destruction of the patella. Significantly expanded joint contour, increased temperature and limited range of motion ( $-5^{\circ}$ - $90^{\circ}$ ) were observed.

As part of the presurgical evaluation of the affected knee, three MRI studies and one CT were performed. These showed a large mass within the patella infiltrating almost its whole volume. Only a small portion of the patella adjacent to the patellar base remained intact. Although the distended nature of the tumor resulted in moderate patellar enlargement with thinning of the patellar cortex, there was no tumor infiltration into the surrounding tissues, nor any tumor expansion into the joint cavity. The articular surface of the patella was secondarily distorted by the tumor mass. The mass showed a heterogeneous intermediate T1WI signal intensity with mixed heterogeneous hypo- and hyperintense T2WI areas and a marked heterogeneous contrast

enhancement. Also, the patellar tendon and quadriceps ligament showed moderately increased T1WI and T2WI SI with contrast enhancement, which most likely was reactive in nature and partially decreased as confirmed in a series of presurgical imaging studies. There was no structural distortion of the patellar ligament and quadriceps tendon. The other structures of the knee had no significant findings (Fig. 1).

Open biopsy of the right patella was performed with a frontal median incision. The histopathological examination revealed a giant cell tumor without malignancy features.

After oncologic consultation, the patient was eligible for therapy involving a human IgG<sub>2</sub> monoclonal antibody that binds to RANKL (Denosumab). Three doses of 120 mg each were injected at 4-week intervals. Improvement of joint range of motion ( $0^{\circ}$ - $120^{\circ}$ ) was obtained as well as reduction of swelling and pain.

Meanwhile, solid allogeneic graft consisting of quadriceps tendon, patella, patellar ligament and tibial tuberosities was obtained (Fig. 2).

Imaging examination (MRI) was once again performed. Apart from partial regression of reactive changes within adjacent tendinous-ligamentous structures, during the diagnostic-observational period of the last two MRI studies, reduction of the tumor mass was observed.

After obtaining informed consent, the patient was eligible for grafting of patella with knee extensor mechanism.

The surgery was performed using the anterior medial approach. The affected patella with patellar ligament and tibial tuberosity were removed as well as the dis-

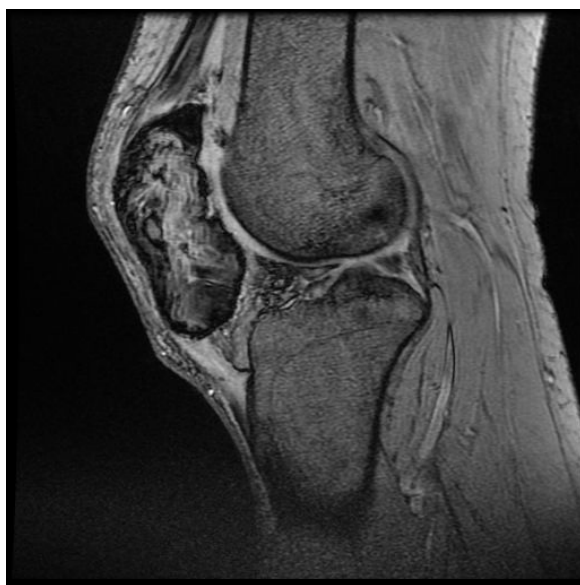


Fig. 1. Presurgical MRI evaluation of the affected knee

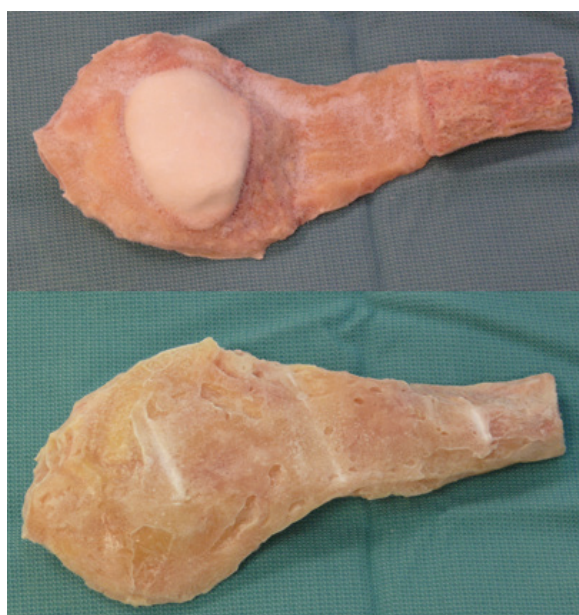


Fig. 2. Allogeneic graft

tal part of quadriceps tendon (Fig. 3). When fitted, the solid allogeneic graft was fixed with the use of the press-fit method. The distal part (tibial tuberosity) was cross-stabilized with two 3.5mm Synthes cortex screws with washers. Two Biomet Ti Screw titanium anchors each 3mm in diameter were screwed into the base of the patella which was sewn with the distal part of the patient's quadriceps tendon, in knee extension with tension. The obtained connection was additionally strengthened with long-term absorbable suture, using the Krakow suturing technique. Patellar retinacula were reconstructed using two 2.9mm JuggerKnot anchors, one on each side. Frontal articular capsule was sewn to the grafted capsule with a continuous suture using long-term absorbable suture material (Fig. 4).

## RESULTS

Correct positioning of the grafted patella and a satisfying alignment of the graft were observed in postoperative radiographic examination (Fig. 5). The result of histopathological examination confirmed a giant cell tumor without malignancy features.

The first follow-up CT (without contrast administration) performed 4 weeks postoperatively showed the proper healing of tuberosity graft into native tibial bone with adequate partial bone union. Patellar ligament as well as the quadriceps femoris tendon were taut. No implant

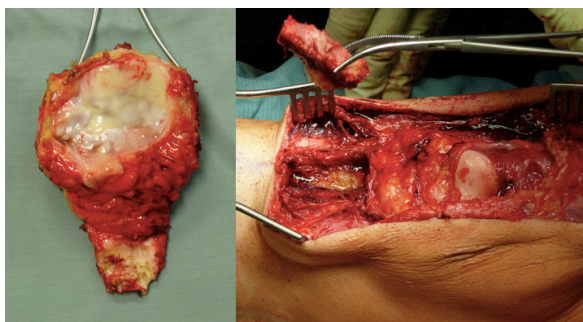


Fig. 3. Resection of the tumor

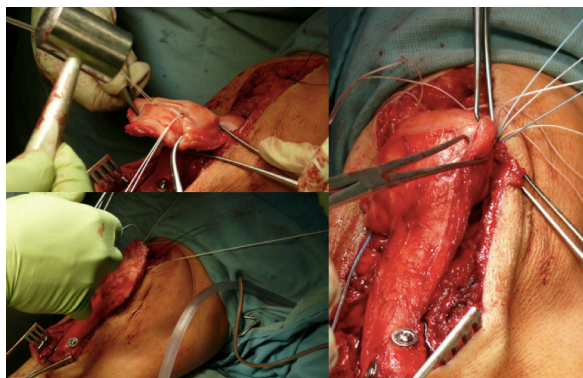


Fig. 4. Proximal stabilization and patellar retinacula reconstruction



Fig. 5. Positioning of the graft in postoperative radiographic examination

loosening was found. The density of the patellar bone was slightly uniformly increased, which most likely corresponded to changes caused by presurgical allograft bone preparation. The patellofemoral joint congruency was intact. A small effusion within the suprapatellar recessus of the joint cavity was seen (Fig. 6).

The second follow-up CT (without contrast administration) performed 15 months postoperatively showed bone union of tuberosity graft and native tibial bone. Patellar ligament as well as the quadriceps femoris tendon were taut. No implant loosening was found. The patellofemoral joint congruency was intact (Fig. 7).

Functional outcome was assessed regularly. Seven weeks after the surgery, the patient presented complete active extension of the knee joint and active flexion at 110° (Fig. 8). Fifteen months after the surgery, the patient presented complete active range of motion, actively participated in professional life and CT tests did not reveal features of tumor recurrence (Fig. 9).

## DISCUSSION

Giant cell tumor of the patella is rare and constitutes a difficult therapeutic problem. Depending on the degree of bone destruction, it may cause significant knee joint dysfunction. It also causes pain, alteration in the contour and increased joint temperature. At early stages of the disease, with no magnification and bone destruction, the treatment of choice is intralesional resection and filling of the tumor bed with bone grafts and bone graft supplements. The method most often used to remove the tumor and clean the tumor bed is curettage with high-speed burr. Some authors recommend follow-up use of phenol, hydrogen peroxide or cryotherapy [23]. Typically, the bed is filled with bone cement. Allograft bones from a tissue bank are also used as supplementary material. When



**Fig. 6.** First follow-up CT

properly prepared and inserted into the tumor bed, they undergo spontaneous remodelling [23]. Increasingly popular bone substitutes, most often containing hydroxyapatite and calcium sulphate, are the solution for patients that do not tolerate allogeneic transplants or in the case of sepsis.

The procedure just described is typical and the most often used. Unfortunately, however, it cannot be applied in complicated cases of significant bone destruction. In such cases, wide local resection with follow-up reconstructive surgery may provide a better functional effect than cleaning the lesion [27]. Alternative patellectomy, as a mutilating surgery with poor functional outcomes, should be treated as a last resort. Similarly, patellar resurfacing surgeries, which due to the great frequency of implant loosening, involve a great number of com-



**Fig. 7.** Second follow-up CT

plications. Arthrodesis, however, causes serious dysfunction of the locomotor system. It is crucial to note that the methods just mentioned are not accepted by patients in the majority of cases.

One remaining option to consider is grafting the patella or the whole extensor mechanism. Due to the frequency of unsatisfying results of reconstructive surgeries of patellar ligament, especially of damaged and degenerated ones, grafting of patella with the use of fixation implants may be thought to carry a high degree of risk. For this reason, grafting of the whole extensor mechanism for the treatment of giant cell tumor of the patella appears to be the appropriate treatment. Such a solution, due to the extreme rarity of the problem, is occasionally used in extensor mechanism injuries that are complications of alloplasty and revision surgeries as well as knee joint injuries [6]. The literature provides a few reports from clinical outcomes of these procedures [2, 4, 5, 10, 16, 21].



**Fig. 8.** Knee range of motion seven weeks after the surgery



Fig. 9. Knee range of motion fifteen months after the surgery

Experience shows that grafting of the whole extensor mechanism is the best method to treat such complications [12]. Unfortunately, these studies are applied to small groups of patients and do not offer long observation periods [2, 4, 5, 10, 16, 21].

The issue concerning the treatment of isolated giant cell tumor of the patella with destruction of patella and joint dysfunction does not have a clear solution in the available literature. In the 20<sup>th</sup> century only 126 cases were described [7]. The rare occurrence of the condition does not facilitate defining procedure algorithms at particular stages of the disease. Given the encouraging outcomes of grafting of whole knee extensor mechanism in complications of injuries, alloplasty and revision surgeries, clinical outcomes with other injuries may be expected to be similar. With no reasonable possibility of treatment of destruction of the patella in the course of giant cell tumor, grafting of whole knee extensor mechanism is justified.

In 2009, Malhotra et al. presented results of such surgery for the treatment of aggressive giant cell tumor of the patella. A 3-year observation period indicated positive results of the surgery [17].

Available literature does not provide any other case of successful grafting of the whole extensor mechanism in the treatment of giant cell tumor of the patella. The case study we present is also the first report of associating

neoadjuvant treatment with the use of a human monoclonal antibody IgG<sub>2</sub> that binds to RANKL (Denosumab) with grafting of knee extensor mechanism.

Early functional outcome was perfect. Reduction of pain, improvement of the contour of the joint, full active extension and flexion of 110° provided the patient with the possibility of returning to the activities of both everyday and professional life. Fifteen months after the surgery, patient presented complete active range of motion, actively participated in professional life and CT tests did not reveal features of tumor recurrence.

## CONCLUSIONS

1. The issue concerning the treatment of an isolated giant cell tumor of the patella with destruction of the patella and joint dysfunction has no clear solution and presents a management problem.
2. Classical treatment methods for a giant cell tumor are often insufficient in cases of significant destruction of the patella and dysfunction of the knee joint.
3. Applying allogeneic grafting of knee extensor mechanism is a promising solution for the treatment of a giant cell tumor of the patella with a significant destruction of patella and dysfunction of knee joint.
4. Applying a human monoclonal antibody IgG<sub>2</sub> that binds to RANKL (Denosumab) appears to be a good method for supplementing the treatment of giant cell tumor of the patella.

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