Intra-articular soft-tissue sarcoma of the knee: Is extra-articular resection and tumor endoprosthetic reconstruction the solution? A retrospective report on eight cases

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Abstract

Intra-articular sarcoma of the knee joint is a very rare condition. Extra-articular resection and reconstruction with a tumor prosthesis is usually performed. This report describes the results with this rare surgical procedure. This retrospective study evaluated the clinical and functional results after extra-articular resection of the knee joint in eight patients with soft-tissue sarcomas of the knee that were reconstructed using a tumor endoprosthesis. Five of the eight patients (62.5%) ultimately had to undergo amputation, mainly due to periprosthetic infection. In addition, two patients experienced periprosthetic fractures. The mean Musculoskeletal Tumor Society score was 18 (range 10-22), as function was impaired due to a weak extensor mechanism. These results suggest that in patients with intra-articular soft-tissue sarcomas, limb salvage procedures with tumor prostheses after extra-articular resection are associated with very high complication rates. In most cases, long-term limb salvage was not possible. When limb salvage is successful, function is also poor due to a weak extensor mechanism in the knee joint. The indication for this procedure should therefore be considered critically.

Introduction

In most cases, intra-articular tumors of the knee joint consist of benign tenosynovial giant cell tumors or chondromatosis.[1] In contrast, intra-articular sarcomas are extremely rare.[1,2] Synovial sarcomas, extraskeletal myxoid chondrosarcomas, or undifferentiated pleomorphic sarcomas are mainly found.[1] Extra-articular resection is recommended, due to tumor contamination in the joint. Apart from amputation or rotationplasty, reconstruction after an extra-articular knee resection is usually achieved using endoprosthetic reconstruction and restoration of the extensor mechanism.[2-4]

In these patients, the knee is resected *en bloc* without opening the joint, with vertical splitting of the patella or complete *en bloc* resection with patellectomy.[3] As a consequence, more extensive muscle resection, especially of the extensor apparatus of the knee, is unavoidable during extra-articular resection − resulting in poorer muscle coverage and functional results, and possibly greater mechanical demands on the stem-bone interface and the joint mechanism.[1]

In most cases, extra-articular joint resection is indicated due to a direct intra-articular extension of a tumor of the distal femur or proximal tibia, either through direct tumor growth, erosion, or pathologic fracture, or due to iatrogenic contamination during biopsy of a bone sarcoma.[4] Herzog *et al*. reported about an extraarticular resection in a patient with a clear cell sarcoma of the anterior cruciate ligament without mentioning the long-term results.[2] To the best of our knowledge, there have been no larger reports describing the results after extra-articular resection in patients with primary soft-tissue sarcomas of the knee joint. This report questions whether this surgical procedure is justified, in view of the oncologic and functional results.

Materials and Methods

A retrospective review identified eight patients (four females, four males; median age 47 years, range 13–66) who had been treated with extra-articular knee resection for a soft-tissue sarcoma between 1992 and 2015 (Table 1). Some patients were included in a study of our group published in 2013 without a special emphasis of the subgroup of patients with a primary soft tissue sarcoma of the knee.[3] The diagnoses were as follows: synovial sarcoma (n=5), leiomyosarcoma grade III (n=1), myxofibrosarcoma grade III (n=1) and undifferentiated pleomorphic sarcoma (n=1). None of the patients had any metastases at the initial presentation. Three patients had previously undergone arthroscopic R1 resections without macroscopic evidence of residual disease, and three patients had had local recurrences after a previous intralesional open resection (two times n=2, three times n=1).

The remaining two patients received an open biopsy of the intraarticular tumor. Three patients had received radiotherapy and four had had chemotherapy. Radiotherapy (60-66 Gy) was applicated in two patients with recurrence after the first intralesional surgery and in one patient after extraarticular resection of the primary tumor in our department. Chemotherapy was applicated in the two children both neoadjuvant and adjuvant according to the CWS protocol. The two adult patients received an individual chemotherapy with ifosfamid and doxorubicin (adjuvant n=1, neoadjuvant and adjuvant n=1).

Extra-articular resection was carried out, using vertical splitting of the patella in the frontal plane and detaching the suprapatellar synovial pouch and infrapatellar fat pad from the patella and quadriceps tendon, including the proximal tibiofibular joint, due to communication with the tibiofemoral joint[5]. All scars from previous surgeries including from arthroscopies were resected *en-bloc* with the resection specimen. The distal femur was resected approximately 7-8 cm above the joint space in order to prevent opening of the joint capsule. *En bloc*

resection of the joint with complete patellectomy was carried out in one child, due to a very thin patella that made vertical splitting impossible. Histopathological examination of the specimens showed that a wide margin had been achieved in all patients.

Defect reconstruction was performed using a Modular Universal Tumor And Revision System (MUTARS®; Implantcast, Buxtehude, Germany) tumor endoprosthesis. In five patients a Mutars KRI component was used for femoral reconstruction. The prosthesis was silver-coated in five patients.[6] The proximal tibia defect was reconstructed with wedges, which were cemented with the underlying spongy bone (Figure 1). A proximal tibia replacement was carried out in patient no. 4.

The median reconstruction length was 11.5 cm (range 7.5-20.5 cm). Five patients with a KRI component received a femoral revision stem (Mutars RS). Cemented femoral stem fixation was carried out in two patients. A rotating-hinge prosthesis was used in seven patients (polyethylene, n=1; metal-on-metal mechanism, n=6) and a fixed-hinged mechanism was used in one expandable prosthesis (MUTARSᴼ Xpand).

Patellar resurfacing was performed in two cases. One (n=5) or two (n=2) gastrocnemius flaps were used to provide adequate muscle coverage of the prosthesis and to reinforce the reconstruction of the extensor mechanism. In addition, a reattachment tube was used for extensor mechanism reconstruction in four patients. Three patients received a mesh graft after gastrocnemius flap placement. Postoperatively, all patients received intravenous cephalosporin for 3 days, followed by oral therapy until wound healing. In patients who were treated with a gastrocnemius flap, knee flexion was restricted in a knee immobilizer for 4 weeks.

The end point of the study was the patient's death, secondary amputation due to local recurrence, or prosthetic failure. Periprosthetic infection was diagnosed in accordance with the Centers for Disease Control (CDC) criteria. Functional results were assessed in accordance with the Musculoskeletal Tumor Society (MSTS) functional score in five patients who did not undergo early amputation.[7] Knee flexion and a possible extension gap were noted in three patients.

Results

Oncological results

Six patients (75%) had no evidence of disease at the final follow-up examination (median 80 months, range 3–91). One patient died of disease (lung metastases) 97 months postoperatively. One patient is alive with disease 47 months postoperatively, with inguinal lymph-node metastases. With regard to local tumor control, none of the patients developed any local recurrences.

Number of revision operations and limb survival

A total of seven patients (87.5%) underwent at least one revision operation (six revisions n=1, three n=1, one n=5). Five of the eight patients (62.5%) ultimately had to undergo amputation in median 24 months after prosthesis implantation (range, 1-36) (Table 1).

Complications

Three patients developed a patella alta due to extensor mechanism insufficiency.

Figure 1. Postoperative anteroposterior and lateral radiographs after extra-articular resection of the knee joint and reconstruction with a distal femur replacement (patient 2).

Table 1. Patients' characteristics.

Patient	Age	Diagnosis	CTX	RTX	Previous intralesional surgeries	Recurrence before extraarticular resection	Complications	Time of first complication*	Amputation	Time of amputation*	Stage of disease	Follow-up
1	57	Leiomyosarcoma	No	No	No	No	Periprosthetic fracture	32	No	/	DOD	96
2	31	Synovial sarcoma	Yes	Yes	Yes	Yes	Infection	14	Yes	36	NED	36
3	66	Synovial sarcoma	No	No	Yes	No	Compartment syndrome	0	Yes	1	AWD	1
4	43	Synovial sarcoma	No	No	Yes	No	Insufficient quadriceps tendon	0	No	/	NED	83
5	51	Myxofibrosarcoma	Yes	No	Yes	Yes	Insufficient quadriceps tendon Periprosthetic fracture	0 65	No	/	NED	91
6	15	Synovial sarcoma	Yes	Yes	Yes	No	Insufficient quadriceps tendon Infection	0 21	Yes	24	NED	24
7	61	Synovial sarcoma	No	Yes	Yes	Yes	Infection	1	Yes	1	NED	1
8	13	Sarcoma (NOS)	Yes	No	No	No	Infection	28	Yes	28	NED	28

*Months after primary surgery. NOS: not otherwise specified, CTX: chemotherapy, RTX: radiation therapy, DOD: dead of disease, NED: no evidence of disease, AWD: alive with disease.

Wound healing disturbances occurred in four patients (50%). Three patients with superficial skin necrosis were treated nonsurgically, whereas one patient with extensive muscle necrosis underwent amputation 1 month postoperatively. In the other three patients with wound healing disturbances, a periprosthetic infection occurred.

None of the patients suffered aseptic loosening. No structural failure of the prostheses occurred (breakage of the prosthetic body or stem, failure of the bushings). Periprosthetic fractures occurred in two patients (25%) 32 and 65 months postoperatively. One of these patients, who had a tibial fracture, was treated with a longer proximal tibia replacement; the other, who had a combined femoral and tibial fracture, was treated with implantation of longer cemented stems.

Periprosthetic infection was observed in four patients (50%) a median of 18 months postoperatively (range 1-28 months). All of the patients developed fistulas, and the causative organism was identified in three patients. The silver-coated prostheses became infected in two of the five cases (40%) and the uncoated prostheses in two of the three cases (66.7%). All of the patients who were treated with additional radiotherapy (n=3) developed infections.

Two patients were treated with primary amputation due to poor soft-tissue conditions, and two patients received a cement spacer after prosthetic explantation. However, although the first of these patients was not treated with reimplantation due to poor soft-tissue conditions and had to undergo amputation, the other had a prosthesis reimplanted. An above-knee amputation was ultimately unavoidable due to repeat infection.

Functional results

The MSTS functional score showed a median of 18 (range 10-22), representing 60% of normal function. Two of the three patients in whom limb salvage was achieved used an orthosis for stance phase stabilization and had a patella alta with an active extension gap >50°.

Discussion

Knee joint involvement in sarcoma patients is usually caused by ingrowth of a bone sarcoma in the distal femur or proximal tibia into the joint space, or due to a pathologic transarticular fracture with subsequent joint hematoma.[1,3] Intra-articular sarcomas of the knee are very uncommon,

whereas benign tumors are much more frequent (*e.g.*, chondromas and giant cell tumors of the tendon sheath).[1] Due to the rarity of malignant intra-articular sarcomas, physicians who are unaware of the possibility of a sarcoma may carry out arthroscopic intralesional resections. In the present study, three patients were treated with arthroscopy because the tumor has been assessed as benign. In addition, three patients were treated for local recurrences after inadequate open resection.

There have been no studies in the literature explicitly investigating the surgical treatment of sarcomas originating in the knee. Chebib *et al.* reported that seven out of 15 patients with intra-articular knee sarcomas (46.7%) required primary amputation.[1] The authors did not report the type of surgical excision used in the limb salvage group. The aim of the present study was to evaluate whether extra-articular knee resection is an oncologically safe and technically feasible option in this very rare group of patients. Despite the small numbers of patients included, which make statistical analysis impossible, we believe that the clearly negative clinical and functional results make publication of the findings obligatory. With regard to the oncologic results, Chebib *et al.* reported that nine patients (60%) were free of disease at the latest follow-up examination (range 12-150 months).[1] In the present study, 75% of the patients are free of disease even though a limb salvage procedure was attempted. Importantly, none of the patients experienced local recurrences, after wide resection in all cases. We therefore consider that extra-articular resection is usually possible, with vertical splitting of the patella, as described by Hardes *et al.*[3] However, due to the resection of large parts of the knee extension apparatus and poor soft-tissue coverage despite the use of one or two gastrocnemius flaps, this procedure is associated with a very high rate of complications. The most important point here is the infection rate of 50%. All three patients who received adjuvant radiotherapy developed infections. Overall, all of the patients had more or less poor soft-tissue conditions and had to undergo amputation either primarily or after unsuccessful attempts at secondary implant exchanges. Another patient who had extensive muscle necroses also had to undergo amputation, so that extremity-preserving treatment was not possible in the short or long term in a total of 62.5% of the patients. The other three patients required ortheses, with markedly reduced walking ranges overall. Periprosthetic fractures

occurred in two of these patients as a result of increased susceptibility to falling.

Conclusions

The high rate of secondary amputations and the poor functional results among the patients who have so far received extremity-preserving surgery for primary soft-tissue sarcomas of the knee mean that the critical question needs to be raised of whether primary amputation and exoprosthetic treatment might have spared the patients burdensome follow-up operations with generally only a slight likelihood of long-term preservation of the extremity. It must also be critically noted that modern microprocessor-guided exoprostheses in patients with above-knee amputations provide very good functional results and would in all probability surpass the functional results achieved in the patients described here.

References

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