

Total Hip Arthroplasty on Unrecognized Bone Tumors: Dramatic Outcome from 11 Cases.

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Abstract

Background: Several mistakes in the diagnosis and treatment of bone tumors can be made, especially in non-specialized centers. Implanting conventional prostheses on an unrecognized bone tumor causes contamination of the entire region with dramatic consequences for prognosis. Purpose of our study was to try to understand which is the best way to deal with these patients. Does previous surgery affect prognosis? Does external hemipelvectomy achieve a better overall survival and local control than limb salvage surgery?

Hypothesis: Demolitive surgery, scarifying the involved limb ensures better local control of the disease and improves life expectancy.

Patients and Methods: We retrospectively evaluated all patients with bone sarcomas at the site of a total hip arthroplasty (THA) over the years 2000-2012. After reviewing the preoperative imaging and histological slides, 11 patients had a THA implanted on an unrecognized hip sarcoma. Diagnosis was chondrosarcoma in 10 patients and osteosarcoma in one. Five patients were immediately treated with external hemipelvectomy.

Results: Five of 11 patients (45%) died of disease at a mean time of 34 months (range 2-82 months), 4 are alive with disease and only 2 are continuously disease free. Six of eleven patients (55%) had a local recurrence at a mean time of 17 months (range 3-36 months); six of these patients had conservative treatment.

Conclusions: Although a very rare event, failure to recognize an occult malignant bone tumor during total hip arthroplasty associates with poor survival rate. Outcome after limb saving surgery is disappointing due to a high rate of local recurrences. According to our experience external hemipelvectomy provides better local control but this condition remains a dramatic event.

Keywords: Total hip arthroplasty; Chondrosarcoma; Pelvic tumors

Introduction

Primary sarcomas of the bone and soft tissues represent a very rare entity. Their incidence is considered generally not more than 1% of all malignant tumors. Specifically regarding the bone sarcomas, their incidence is of about 0.8-1 new cases/100000 inhabitants/year. As for other rare disease, diagnosis and treatment is complex, especially in non-specialized centers. Many errors in musculoskeletal oncology are due to a misdiagnosis: lack of detection of an abnormality that would suggest a neoplastic process, or attributing an abnormal clinical or radiological finding to a benign etiology.

In some cases, diagnosis of a bone tumor in the femoral head or acetabulum can be difficult and often mistaken with more common pathologies such as osteonecrosis or osteoarthritis [1]. Implanting a conventional prosthesis in a primary malignant bone tumor can cause contamination of the entire region thus compromising the patient's life.

Surgical management of pelvic tumors is a challenge for orthopedic surgeons [2] and even more so when previous surgery disseminated the tumor. Literature gives few indications on how to deal with such complex situations and consequently any recommendation and suggestion regarding the management of these patients is difficult.

The purpose of our study was to try to understand the safest approach to this event. Does previous surgery affect prognosis? Does external hemipelvectomy obtain better results in achieving increased

overall survival and better local control as opposed to limb salvage surgery?

Materials and Methods

Patients

We retrospectively studied the tumor files at our Institute of all patients admitted for a bone lesion at the site of a THA from 2000 to 2012. We identified 11 patients who were treated for the consequences of a THA implanted on an unrecognized bone tumor. Five were female and 6 male, with a mean age of 52 years (range 31-70 years). Of these we reviewed the preoperative X-rays and saw that, although sometimes very subtle, there was radiological evidence of the presence of tumor at the site of surgery (Figure 1). Ten patients were treated in another institution for the first surgery. Primary osteoarthritis was the indication in nine patients and two were treated for femoral neck fracture. Ten patients underwent a THA, one a hemiarthroplasty.

Diagnosis of the primary bone tumor was chondrosarcoma (CS) in 10 cases (two of these were CS dedifferentiated in high grade osteosarcoma) and radio induced osteosarcoma in one case. In 4 cases the tumor was located in the acetabulum, while in 7 in the femoral head. According to Enneking's classification [3], 9 patients were stage IIB (extra-compartmental localized disease) and two stage III (metastatic disease). Adjuvant chemotherapy was administered in four patients: in two with CS dedifferentiated in high grade OS, in one with OS, and in one with Gr 2-3 CS with lung metastases.



Figure 1: High grade osteosarcoma of the proximal femur a) Preoperative MRI in a 68-year-old patient presenting with pain in the right hip. Diagnosis of aseptic necrosis of the hip was supposed. b) THA was implanted. c) 1 Month later, MRI showed a soft tissue mass around the prostheses. d) Extra-articular resection was performed and reconstruction with allograft composite prosthesis. e) 3 Months later CT scan showed local recurrence. f) Hindquarter amputation was performed to treat the local recurrence.

Methods of assessment

Diagnosis was confirmed at our Institution by CT-guided biopsy in 4 patients (acetabulum localization) and in 7 by review of histological slides. Five patients were immediately treated with external hemipelvectomy and the other six with limb salvage surgery. In the 7 patients with localization in the proximal femur we performed 5 extra-articular resections and 2 amputations. We restored the bone defect by megaprosthesis in 3, by allograft prosthesis composite in one (McMinn-Link prostheses was used for the reconstruction) and by antibiotic spacer for THA infection in one. In all cases the femoral resection was performed below the stem.

In acetabular localizations (4 cases) we performed an external hemipelvectomy in 3 cases and a periacetabular resection and

reconstruction in one; massive bone allograft combined with stem cup prosthesis was used. Margins were wide in 8 cases (73%), marginal in 2 and intralesional in one. All patients were staged and discussed in a multidisciplinary setting and assessed by X-ray, MRI or CT scan of the pelvis, chest CT and Bone Scan during preoperative staging. Follow-up was conducted according to our protocols with chest CT scan and pelvic MRI or CT scan with contrast medium and X-ray of the prosthesis every 3 months for the first 2 years, every 4 months for the third year and then every 6 months.

Results

The reported results were recorded after a mean follow-up of 38 months and a minimum follow up of 6 months. Five of the 11 patients

(45%) died of disease (DOD) at a mean time of 27.4 months (range 6-40 months), 4 patients are alive with disease (AWD) at a mean time of 43 months (range 12-100 months) and only 2 patients are continuously free of disease (CDF) with a follow up of 16 and 68 months. Among the five patients who were immediately treated with external hemipelvectomy only one died of disease (20%), two are CDF and the other two are AWD (with lung metastases).

Six patients (55%) had local recurrence at a mean time of 15 months (range 3-36 months) and of these five had had conservative treatment. In the group of 6 patients treated conservatively, four died of disease (66.6%) and two are alive with disease. Among the patients who died, 4 had a local recurrence (80%). One patient treated with hemipelvectomy had a local recurrence. The three patients treated with inadequate margins had local recurrence.

Nine patients were free of metastases at diagnosis, 7 of these (78%) developed lung metastases at a mean time of 14 months (range 2-36 months). The patients who developed local recurrence also developed lung metastases, one of these also developed bone metastases. Two patients treated conservatively developed also bone metastasis. All these results are summarized in Table 1.

Discussion

Pelvic tumors are a big challenge for orthopedic oncologists. Wide surgical margins are often difficult to obtain and intralesional or debulking procedures have a higher rate of local recurrence [4] with a worse prognosis. In our study we evaluated 11 patients who were treated for standard THA implanted on an unknown tumor involving the acetabulum or proximal femur. This error jeopardizes surgical treatment, making local and systemic control of the tumor more difficult and thus compromising the oncologic outcome. As seen in our study, 5 patients (45%) died of disease at a mean time of 27.4 months, and only two are CDF, in all these cases the surgeon failed to see that tumor was present.

In our cohort, chondrosarcoma was the most frequent diagnosis and it is well known that surgical resection remains the main stay of treatment for this tumor [5]. Primary goal of surgery was complete removal of the tumor with limb function preservation whenever possible. The extent of tissue contamination in these cases is very high because of the acetabular and intramedullary reaming, and for the surgical approach itself. Moreover, the presence of the implant makes evaluation of residual disease or local recurrence difficult for inherent imaging artifacts. Adams et al. [6] described a series of 8 patients treated at a tumor center after an inadvertent internal fixation of a primary osseous sarcoma. Six of these subsequently underwent amputation and only two limb salvage procedures. Only four patients were alive at an average of 26.9 months. They concluded that inadvertent surgery for high grade bone sarcomas was associated with poor survival, in spite of the high rate of amputations. Puri et al. [7] evaluated 14 patients with prior intervention, either as an open biopsy or curettage/unplanned excision for a chondrosarcoma, documenting a poorer event free survival (EFS) but a similar rate of local recurrence. In this case though, tissue contamination is not the same as for patients who undergo major surgery such as joint replacement. Some of the reported survival rates [8,9] are comparable to primary treated sarcomas, but this mainly depends on the nature of the tumor rather than its surgical management.

Our study has the inherent limitation of being a retrospective evaluation. The small number of patients prevents us from statistically validating the observations. The decision for the type of surgical treatment was not only based on the surgeon's indications but also following the patient's preferences since some patients preferred conservative surgery preserving the limb rather than an external hemipelvectomy.

Patient	Age	Diagnosis	Months before referral to our clinic	Stage	Treatment	Margin	Mets at diagnosis	Months to LR	Second treatment	Status	Months FU	Mets at follow up	Other Treatment
1	66	Myxoid CS Gr 3	36	IIB	Hemipelvectomy	W	No	---	--	CDF	16	No	No
2	64	Central CS Gr 2-3	24	III	Hemipelvectomy	W	Lung	---	CHTP	AWD	48	---	CHTP
3	31	Central CS Gr 2-3	3	IIB	Conservative	M	No	36	Hemipelvectomy	DOD	40	Lung	No
4	39	CS Gr 2-3	4	IIB	Hemipelvectomy	W	No	---	---	CDF	68	No	No
5	32	Myxoid CS Gr 2-3	6	IIB	Conservative	W	No	6	Conservative	DOD	32	Lung+ bone	CHTP + T
6	40	CS grade 2-3	17	IIB	Conservative	W	No	19	Hemipelvectomy	DOD	20	Lung	T
7	64	CS dedif in OS Gr4	3	III	Hemipelvectomy	W	Lung	---	CHTP	AWD	12	---	CHTP
8	68	Gr 4 OS	0.5	IIB	Conservative	M	No	3	Hemipelvectomy + CHTP	AWD	12	Lung	CHTP
9	32	Clear cell CS	168	IIB	Conservative	I	No	24	Hemipelvectomy	AWD	100	Lung + Bone	RxTp
10	63	Central CS Gr 2	2	IIB	Conservative	W	No	---	---	DOD	39	Lung	CHTP + T
11	70	CS dedif in Os Gr3	37	IIB	Hemipelvectomy	W	No	4	CHTP	DOD	6	Lung	CHTP

Table 1: Patient data, treatment and results at follow-up
W=Wide; M=Marginal; I=intralesional; CHTP=Chemotherapy; CDF=Continuously disease free; AWD=Alive with disease; DOD=Dead of disease; LR=Local recurrence; RxTp=Radiotherapy; T=Thoracotomy

Limb salvage after intralesional unplanned surgery is controversial. According to Enneking [10] the definitive management of sarcoma patients who underwent prior surgery involves removal of all tumor and potentially contaminated tissue at the local site. Contamination of surrounding tissues makes tumor resection with free margins technically difficult. Contaminations after such surgery is comparable to a pathological fracture, and for many authors, sarcomas accompanying pathological fractures are considered to be a relative indication for amputation [8]. Local recurrence rate in our study was 55% (6/11 patients). Five of these patients were treated with limb salvage surgery but only one patient, treated immediately with external-hemipelvectomy, had a local recurrence. Secondary amputation for local recurrence after limb salvage procedures was performed in 4 of 6 patients. In these patients the recurrence was very severe and re-excision was not feasible. Studies in literature confirm that previous unplanned surgery is associated with a higher rate of local recurrence [6-8,11,12]. Dae-Geun et al. [8] evaluated 25 patients treated with unplanned intralesional procedures, 22 had limb salvage surgery and of these 4 had a local recurrence. Three cases had an amputation with no local recurrence. They concluded that attempts for limb salvage in malignancies treated intralesionally is accompanied with a higher rate of local recurrence and extensive operative fixations represent a relative contraindication to limb salvage procedures. This finding was also confirmed in our study since patients treated with amputation had less local recurrences than those treated with limb salvage.

Nine patients were free of metastases at diagnosis, 7 of these (78%) developed lung metastases at a mean time of 14 months (range 2-36 months). Patients who developed local recurrences also developed lung metastases. Different authors sustain that improper manipulation of a malignant tumor increases the risk of metastases [6]. Wang et al. [12] reported 50% of lung metastases in patients who received unplanned treatment for osteosarcoma, this percentage is similar for patients with appropriate treatment. This can be explained by the fact that this group of patients received more aggressive chemotherapy. In our study the rate of metastases was much higher because of the nature of the tumor. It is well known that chondrosarcoma, the most frequent diagnosis in our series, is resistant to chemotherapy, and this is why adequate surgical treatment is mandatory for the oncological outcome.

Extensive surgery performed at the site of an unrecognized malignant tumor compromises surgical resection by spreading the tumor in the adjacent tissues. The initial diagnostic work-up is an important aspect to avoid such mistakes. In 11 cases of our study the surgeon failed to recognize that tumor was present, and only in one case the surgeon recognized that there was a tumor present but it was probably misdiagnosed as a benign lesion (case number 9: a clear cell chondrosarcoma [13] initially diagnosed as chondromyxoid fibroma). Adequate attention to patient history is fundamental for a correct diagnosis as can be seen with patient number 8. Although the patient's clinical history stated prior radiation therapy for uterine cancer, the surgeon did not consider the possibility of a pathological fracture from a radio induced sarcoma [14]. Another fundamental step to avoid such errors is to perform a complete radiographic evaluation for patients in whom typical changes of osteoarthritis are not present. These patients should be assessed with MRI or even CT scan, and if there is still a doubt, a biopsy should be performed prior to the hip reconstruction. A pathological examination of the femoral head should always be performed or even a frozen section if an unexpected mass is discovered during the surgery.

Conclusion

In this small series of patients it is not possible to give definitive recommendations. The presence of any clinical or radiological sign of pathologic bone should raise the clinical suspicion of a bone tumor. In this case an accurate study (CT-scan; MRI or even a biopsy) should be previously performed. Local recurrence after conservative surgery is very high due to local contamination. Following to our experience, independently to the method of treatment the prognosis in this group of patients still remains poor.

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