MULTIPLE OR METASTATIC OSTEOSARCOMA, IN YOUNG MALE PATIENT: CASE REPORT

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ABSTARCT
OSTEOSARCOMA AS A MLIGNANT TUMOR HAS A PRESERVED PROGNOSIS, WHICH DEPENDS ON THE EARLY, CORRECT DIAGNOSIS. SURGICAL AND ADJUVANT CHEMITHERAPY ALLOWS A GOOD PROGNOSIS.
A 17 YEAR OLD MALE PATIENT PRESENTED TO OUR INSTITUTION, WITH A RECENT APPEARED PALPABLE MASS OF PROXIMAL THIRD OF THE LEFT LEG. CLINICAL AND PARA CLINICAL INVESTIGATION PUT ON EVIDENCE AN OSTEOSARCOMA. THE PATEINT UNDERGOES TO CHEMOTHERAPY AND SURGICAL LIMP SPARING SURGERY WITH MODULAR PROSTHESIS RECONSTRUCTION. 6 MONTHS AFTER SURGERYA SLIGHT PAIN APPEARS ON THE RIGHT DISTAL THIGH AND CHEST, BIOPSY CT AND BONE SCINTIGRAPHY REVEALS OSTEOSARCOMA. THIS CASE UNDERLINES THE IMPORTANCE OF EARLY AND CORRECT DIAGNOSIS. FURTHER RESEARCHES ARE NEEDED IN ORDER TO KNOW THE EXACT MECHANISM OF THE APPEARNCE OF MULTIPLE METACHRONOUS OSTEOSARCOMA. OUR CASE REPORT PRESENTS A RARE APPEARNCE OF OSTEOSARCOMA, WITH A REDUCED NUMBER OF SIMILAR REPORTS IN THE REVIEWED LITERATURE.

KEY WORDS: OSTEOSARCOMA, LIMB-SPARING SURGERY, MODULAR PROSTHESIS

INTRODUCTION

Osteosarcoma is the second most common primary malignant bone tumor. It is characterized by malignant proliferation of bone cells, it can occur at any age. The incidence of primary high grade osteosarcoma is 1-3/1000000, per year; it occurs most frequently on the second decades of life. It can affect the entire skeleton; however most frequently affects the distal femur, proximal tibia and proximal humerus[3].

The etiology of osteosarcoma is still unclear; several theories have been described, which may lead to malignant proliferation of bone cells. Fuchs B. et al underline the presence of chemical agents like beryllium, viruses, irradiation as potential factors in the etiology of osteosarcoma. Recent studies show that hereditary diseases, like retinoblastoma, Rothmund-

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Thomson syndrome, Bloom syndrome, Li-Fraumeni syndrome increase the risk of developing osteosarcoma[4] [5].

The symptoms of primary osteosarcoma are not specific, it consist of progressive localized pain, which can be sever in late stages. The presence of night pain can be an important sign for the true diagnosis. The radiological aspects consist of osteolytic lesions, with bone production and destruction zone, with ill defined border. "Codman's triangle" as periostal reaction suggests the malignant character of the lesions[6] [7].

The surgical treatment of primary localized osteosarcoma consisted of the complete removal or amputation of the affected limb. Nowadays due to the advent of chemotherapy and the use of special modular prosthesis the aim in the treatment of osteosarcomas is limb-sparing surgery for a better life quality of patients[8].

In this paper we present a case of a patient with primary localized osteosarcoma, who underwent to limb-sparing surgery with special prosthesis implantation.

CASE REPORT

A 17 year old male patient presented to orthopedics institution in the University Emergency Hospital of Bucharest, with a palpable mass in the proximal third of the left leg, with any remarkable medical personal history in the past. The symptoms appeared approximately 8 months before presentation. The symptoms first consisted of slight pain with a progressive character. According the patient mentions pain often occurred at night in the last few weeks.

The clinical exam reveals a palpable mass of the proximal third of the antero-lateral region of the left leg, with an approximate 10/7 cm size. The tumor was fixed to the deeper tissues, the covering skin modification suggests the infiltrating and fast growing character of the tumor. (Figure 1.)

Figure 1. Aspect of the tumor at the clinical examination.

Figure 2. X-ray of the left leg showing ill determined osteolytic lesions with periostal reaction

[4] Fuchs B, Pritchard DJ. *Etiology of osteosarcoma*. Clin Orthop Relat Res. 2002; 397:40-52.
[5] Ottaviani G, Jaffe N. *The etiology of osteosarcoma*. Cancer Treat Res. 2009; 152:15-32.
[6] Jacobs P, *Radiological diagnosis of osteosarcoma*. Proc R Soc Med. 1976; 69(8): 543–546.
[7] Hopper KD, Moser RP, Haseman DB, et al. *Osteosarcomatosis*. Radiology 1990; 175:223–39.
[8] Amstutz HC. *Multiple osteogenic sarcomata: metastatic or multicentric: report of two cases and review of literature*. Cancer1969; 24: 923–931.

After clincal exam standard X-ray examination was performed (Figure 2.), in order to achieve better anatomical details of the tumor CT and MRI was performed, which confirms the malignant character of the tumor, without any pathological modification of the chest, abdomen, skull and pelvis (Figure 3.)

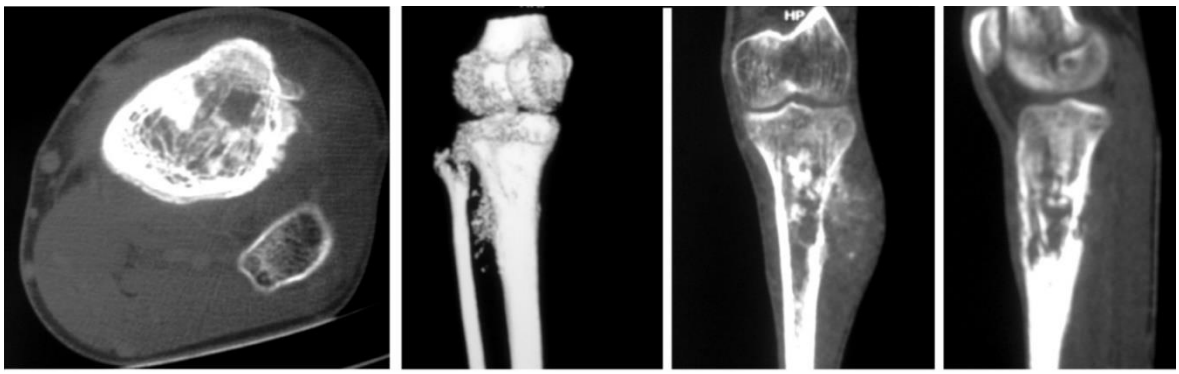

Figure 3. CT showing an expansive process of the proximal third of the left tibia with an approximately 70mm, with periostal reaction and muscular invasion.

During laboratory examination the following parameters were changed: PT: 78,4% (N:80-130); Alkaline phosphates 447 U/L (N:40-136 U/L); ESR: 39mm/h (N: 5-10mm/h). Other laboratory results were within normal ranges.

After incision biopsy the hystological result was high grade malignancy osteosarcoma. Due to the fast growing, highly malignant character of the tumor and in order to prevent secondary metastasizes the patient undergoest to limb-sparing surgery with modular prosthesis reconstruction.

The surgical procedure consisted of the resection of the proximal 1/3 of the tibia and of the proximal extremity of the fibula with an anterior approach of the knee and proximal leg. In order to reconstruct the extensor apparatus of the limb, the patellar tendon was des-inserted from the tibial tuberosity. (Figure 4)

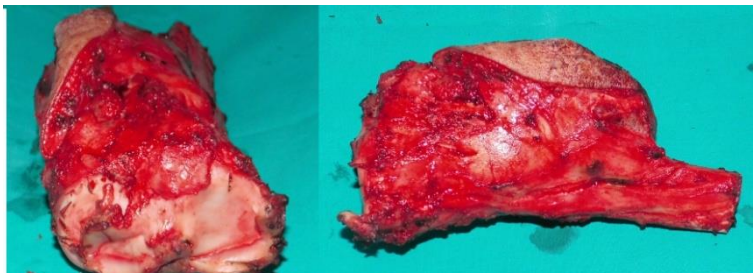

Figure 4. Tumoral piece after surgical removal of the tumor .

The preparation of the distal femur was performed with centromedullar guided distal cut. For reconstruction of the knee joint and the proximal third of tibia special modular prosthesis was used (Zimmer) The patellar tendon was reinserted in anatomical position on the tibial part of the prosthesis, with gastrocnemius covering flap. (Figure 5)

Figure 5. Modular prosthesis (A); distal femur cut (B); reconstruction process with covering flap(C)

The removed bony and soft tissue components of the tumor were sent for further histological examination. The results of the histological analysis confirm that the resection was performed in between oncological limits, without any malignant cells in the resection zone. Rehabilitation was begun on the second day after surgery with passive motions which was followed by active motion exercises. Postoperative evolution of the patient was good, partial weight bearing was achieved after 7 days of surgery. After 44 days of hospitalization the patient leaves our institution. (Figure 6)

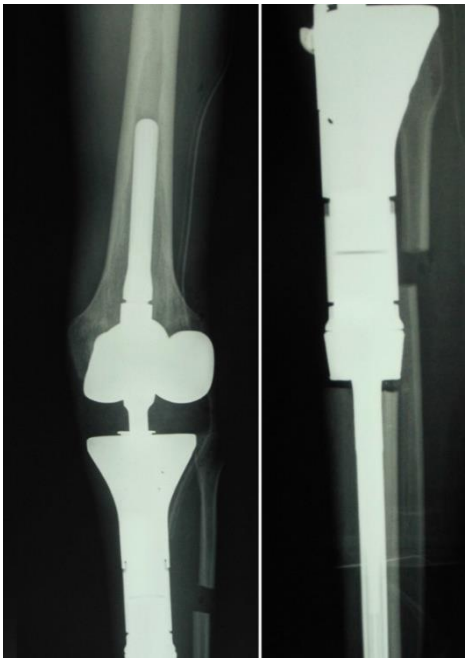

Figure 6. Postoperative X-ray of the reconstruction

6 Months after surgery the patient accuses slight dorsal chest pain and right distal thigh pain, symptoms, which appeared a few weeks ago without disturbing the daily life. Standard X-ray of the right lover limb reveals an osteolytical lesion of the distal third of the femur. Full body CT scan evidences a space occupying process in the right costo-vertebral space at T3-T4 level and also describes an osteolytic and osteoproductive process in the distal third of the femur, whith periostal reaction. Bone scintigraphy with Tc 99m shows hypercaptation in metabolic phase in the aforementioned thoracic and right femur region (Figure 7, 8)

Dispite of the adjuvant chemotherapy after surgical intervention, histological analysis of the newly appeared lesion confirms the osteosarcom of the right distal femur, and of the thorax.

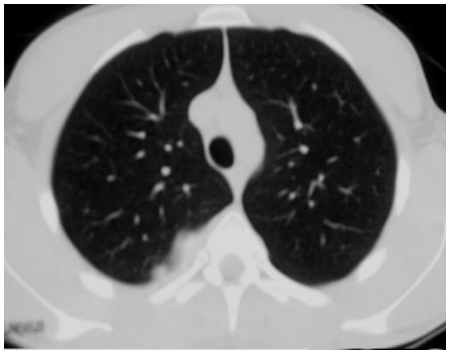

Figure 7. CT scan of the thorax, space occupying process in the right costo-vertebral space.

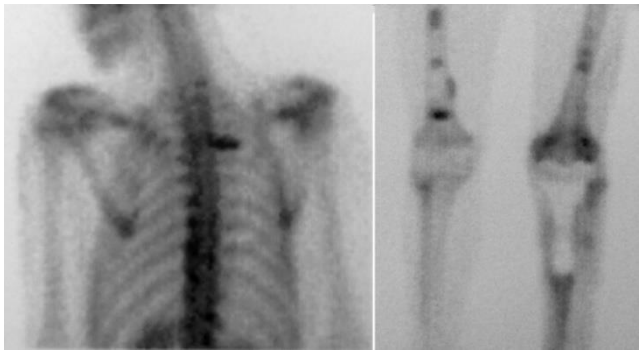

Figure 8. Bone scintigraphy showing hypercaptation, visible in thorax and in the right distal third of the femur.

DISCUSSION

Metachronous osteosarcoma is rare type of bone malignancy with poor prognosis. The pathology of metachronous osteosarcoma, is still unclear[9] [10]. initially, multiple primary malignancies were favored, due to the absence of metastatic routes and the presence of genetic mutations and hereditary diseases. Recent studies have proved that metastases can appear, with similar mechanism to prostatic cancer metastases, via Batson venous plexus[11]. Hatori et al in a recent study demonstrated lymphatic spread to the lungs as possible routes for metastases, which in the case of our patient could be the explanation of the appearance of the space occupying process on the right costovertebral space[12].

Surgical treatment with modular prosthesis reconstruction of ostesarcoma, allows limb salvage and do not affect the quality of life. The Surgical and chemotherapeutical combination in therapy has proven its efficacy. Despite of the good short term results after surgical and adjuvant chemotherapy, the appearance of the tumor in the thorax and the right femur worsens the prognosis[13] [14].

[9] Silverman G. *Multiple osteogenic sarcoma*. Arch Pathol 1936; 21 :88–95.

[10] Harris MB, Gieser P, Goorin AM, et al. *Treatment of metastatic osteosarcoma at diagnosis: a Pediatric Oncology Group study*. J Clin Oncol 1998;16 :3641–3648.

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[12] Hatori M, Ohtani H, Yamada N, Uzuki M, Kokubun S. *Synchronous multifocal osteosarcoma with lymphatic spread in the lung: an autopsy case report*. *Japanese Journal of Clinical Oncology*.2001; 31(11):562–566.

[13] Thompson RC Jr, Cheng EY, Clohisy DR, et al. *Results of treatment for metastatic osteosarcoma with neoadjuvant chemotherapy and surgery*. Clin Orthop 2002;397: 240–247.

This case underlines the importance of early and correct diagnosis. The mechanism of the newly appeared tumors during chemotherapy is unclear, possible explanation is the different chemosensitivty of the tumoral cells.

CONCLUSIONS

Early diagnosed, high grade localized osteosarcoma with surgical, so called limb-sparing, treatment with modular prosthetic reconstruction allows a better life quality. Surgical treatment and adjuvant chemotherapy makes this pathology to have a good prognostic index. Unfortunately with a still unclear mechanism, the appearance of metastases and metachronous osteosarcoma worsens the prognosis

Our case report presents a rare appearance of osteosarcoma, with a reduced number of similar reports in the reviewed literature.

[14] **Harris MB, Gieser P, Goorin AM, et al.** *Treatment of metastatic osteosarcoma at diagnosis: a Pediatric Oncology Group study*. J Clin Oncol 1998;16:3641–8.

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12. **Thompson RC Jr, Cheng EY, Clohisy DR, et al**. *Results of treatment for metastatic osteosarcoma with neoadjuvant chemotherapy and surgery*. Clin Orthop 2002;397: 240–247.