Case report

Bilateral internal hemipelvectomy for osteosarcoma in a pediatric patient previously treated for rhabdomyosarcoma

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A B S T R A C T
The surgical treatment of malignant bone tumors involving the pelvis represents a great challenge in terms of local control. Internal hemipelvectomy is a major surgical procedure that involves the resection of the entire hemipelvis or of a portion of the hemipelvis with preservation of the ipsilateral extremity. The need for a bilateral internal hemipelvectomy is an extraordinary situation. We describe the case of an 11-year-old girl with a primary diagnosis of rhabdomyosarcoma of the bladder at the age of two years who subsequently developed a right pelvis osteosarcoma at the age of six years and a left pelvis osteosarcoma at the age of nine years. She ultimately underwent sequential bilateral internal hemipelvectomies and she postoperatively ambulates without an assist device.

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1. Introduction

Internal hemipelvectomy involves local resection of all or part of the hemipelvis while preserving the ipsilateral lower extremity [1]. It is indicated for management of tumors involving the pelvis that require resection but do not involve the major neurovascular structures of the lower extremity, so that a functional extremity can be preserved [2,3]. Enneking and Dunham described a classification system including type I (resection of the ilium), type II (resection of periacetabular region) and type III (resection of ischiopubic region) [4]. Reconstruction may be done by prosthetic means, biological means, or by combination of endoprosthetics and bone grafts [5,6]. Also, a non-reconstructive procedure referred to as hip transposition has been described [7]. Our patient underwent sequential internal hemipelvectomies and a non-reconstructive approach was taken. Two years after her last internal hemipelvectomy, the patient ambulates without an assist device.

2. Clinical report

An African American female presented at 22 months of age with an embryonal rhabdomyosarcoma of the bladder that required surgical resection, chemotherapy with vincristine, actinomycin D and cyclophosphamide, and radiation therapy with a total dose of 41.4 Gy. Thirty months after completion of therapy, a biopsy of a new lesion in the right iliac wing/acetabulum was diagnostic for chondroblastic osteosarcoma. After neoadjuvant chemotherapy, she underwent local control with a type I-II-III right internal hemipelvectomy. Following the diagnosis of a second malignancy,
the patient was tested for p53 mutations, but no germline mutation was identified. Twenty-nine months after completion of therapy, a biopsy of a new lesion in the left ischium was diagnostic for chondroblastic osteosarcoma, which was thought to represent a third primary malignancy. There was no evidence of metastatic disease. After neoadjuvant chemotherapy, she underwent surgical resection with a type II-III left internal hemipelvectomy. She is able to ambulate with a Trendelenburg gait without pain and good emotional acceptance. She uses crutches for long distances, but otherwise walks without assistance, including climbing stairs and riding a bike. Current imaging studies (Figs. 1 and 2) show the resected proximal femur and iliac bone on the right side and pseudo-acetabulum created by the residual iliac bone covering the intact femoral head on the left side. The medial pubic bones remain intact and provide structural support to internal pelvic structures.

3. Discussion

Compared to other anatomical sites, surgical treatment of pelvic sarcomas shows higher rates of local recurrence and complications and a lower functional outcome [1]. Reconstruction methods are challenging and have to focus additionally on the growing skeleton in the pediatric patients [2,3]. Some of these patients are long-term survivors of irradiated pelvic tumors during childhood or adolescence [8]. Radiotherapy is considered to increase the risk of subsequent bone sarcoma 100-fold following treatment in some pediatric populations [9]. In our patient, given the long interval since her prior osteosarcoma and the fact that this was a solitary lesion, the suspicion of a new primary tumor was high. Although our patient was tested for p53 mutations and no germline mutation was identified, the aggregation of two or more forms of cancer in the same family should be investigated to rule out genetic abnormalities [10]. A variety of different means of pelvic reconstruction after tumor resection have been described including prosthetic and allograft. Complication rates associated with these procedures range from 55 to 77% of cases and are related to soft tissue coverage, wound healing, loosening of the implant and, most frequently, deep infection [5,6]. Also, a non-reconstructive procedure called hip transposition has been described and is considered an excellent surgical procedure for achieving wide margins in periacetabular tumors with good functional outcomes. Gebert et al. [7] reported 62 patients who underwent a hip transposition after periacetabular resection and concluded that this technique is an excellent surgical procedure for achieving wide margins in periacetabular tumors with good functional outcome. Some authors suggest that a hip transposition may provide a better option than other methods in view of long-term functional rehabilitation with less morbidity due to good tissue fibrosis and assistive orthotic devices providing the necessary stability and compensation for limb shortening. A larger leg-length discrepancy is a disadvantage associated with hip transposition. The symmetry and portion of the superior and inferior pubic bones provides structural support to the pelvic floor and associated structures, thus limiting the risk of incisional hernias and bladder displacement.

New technologies have been utilized in the management of this challenging tumor location including the construction of individual pelvic models and the intraoperative navigation technology. Pelvic models are made using available CT-data, becoming helpful tools to planning the manufacture of partial pelvic replacements and ensuring optimal osseous resection of the involved bone [11,12]. Perioperative CT computer navigation is helpful to obtain minimal bone resection while optimizing oncological margins. Patient-specific instruments have been developed as a preoperative resection strategy that can be replicated intraoperatively [13]. We have described a pediatric case of metachronous bilateral pelvic osteosarcoma that occurred four and seven years, respectively after treatment of bladder rhabdomyosarcoma. The need for a bilateral internal hemipelvectomy and the good functional outcomes make this case to be unique, although the 2-years follow-up of this patient makes it difficult to predict the long-term functional outcomes.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

References


