Pelvic sarcoma surgery (literature review)

R. V. Malyk 1, Ya. O. Golovina 2, O. Ye. Vyrva 2

1 Kharkiv Medical Academy of Postgraduate Education of the Ministry of Health of Ukraine
2 Sytenko Institute of Spine and Joint Pathology National Academy of Medical Sciences of Ukraine, Kharkiv

Tumor lesions of the pelvic bones represent a difficult disease for management and to achieve good functional results. About 10–15 % of all primary malignant tumors are localized in the pelvic bones. Objective. To perform a literature review to identify key historical scientific and practical achievements that have influenced the stages of the development of pelvic sarcoma surgery. Methods. To study the publications at Google search engine, electronic databases PubMed, Google Scholar, archives of specialized journals and other sources of scientific and medical information. Results. The history of the development and improvement of the treatment pelvic bone tumor patients has more than 125 years. Important stages in the formation of this oncologic orthopedics area are improvement of anesthesia and resuscitation methods, study and understanding of mesenchymal tumors biology, modernization of neoplastic imaging methods, development of polychemotherapy (PCT) and radiotherapy protocols, the possibility of 3D-modeling of surgical interventions, creation of various artificial and biomaterials for bone defects replacement, critical view on oncological, functional outcomes and complications assessment. Today, the majority of patients with local pelvic bone sarcomas have the opportunity to perform limb salvage surgery. However, the issue of reconstruction of pelvic bone defects after massive resections remains incompletely understood. Reconstruction often provides a better functional result at a price of greater complications, and it influences the choice of the applied method. Conclusions. Despite of significant advances that was achieved since the first attempt of hemipelvectomy in 1891, a number of problematic issues in pelvic tumor surgery remains unresolved. Therefore, the search for a more adequate, less traumatic and functionally beneficial method of postresection pelvic bone defects reconstruction using biological materials and various types of custom-made/serial prosthetics keeps going, especially when the acetabulum is involved.

Key words. Pelvic tumor surgery, pelvic resection, hemipelvectomy
Introduction

Pelvis is a complex anatomical structure, and damage to its bones in tumor processes leads to many difficulties in treatment and obtaining good functional results, which significantly affects the quality of life. This is due to the peculiarities of the anatomical structure of its bones (in particular, the acetabulum), as well as the adjacent pelvic organs and vascular and nervous structures.

About 10–15 % of all primary malignant bone tumors are localized in the pelvic bones. The most common nosological forms of this severe disorder are chondrosarcoma, osteosarcoma, and Ewing's sarcoma. The ilium is affected in 65 % of cases, pubis in 20 %, and ischium and sacrum in 8 % each [1].

Given the complex anatomical structure of the pelvis, reconstruction of the pelvic ring after tumor removal is the most challenging task for surgeons. Particular difficulties are added by the desired restoration of the bearing capacity of the lower limb and the function of the hip joint in the presence of a post-resection defect of the acetabulum. Therefore, this localization of pelvic lesions is the most challenging and requires constant thorough study. The need for reconstruction requires a variety of restorative techniques and, accordingly, complications of their application [2]. Today, there is no universal model of reconstruction of the pelvis or its segments, and the existing methods do not ensure successful rehabilitation of all patients. This is related not only to tumor size, anatomical localization, histological type, radicality of resection, but also to a high risk of developing intra- and postoperative complications, which occur in 55–77 % of cases [3].

Purpose: to analyze the scientific literature to identify key historical scientific and practical achievements that influenced the stages of development of pelvic sarcoma surgery.

Material and methods

The study involved an assessment of publications from the Google search system, electronic databases PubMed, Google Scholar, archives of specialized journals and other sources of scientific and medical information.

Results and their discussion

The history of the development and improvement of the treatment of pelvic bone tumors spans more than 125 years and is associated with the progress and achievements of several related fields of medical science. The important stages of development that caused significant steps in the improvement of surgical treatment of patients with malignant tumors of the pelvis include:

- improvement of anesthesia and resuscitation methods;
- in-depth study and understanding of the biology of mesenchymal tissue tumors (sarcoma);
- improvement of methods of visualization of neoplastic processes, namely: introduction of computer tomography (CT) and magnetic resonance imaging (MRI) into practical medicine;
- development of modern polychemotherapy (PCT) and radiation therapy (RT) protocols;
- improvement of pelvic bone resections and pelvic organ surgery;
- the possibility of 3D modeling of surgical interventions;
- availability of various modern artificial and biological materials for replacing bone defects;
- critical assessment of treatment results and complications [3, 4].

At the current stage, most patients with local sarcomas of the pelvic bones have the opportunity to perform organ-sparing surgical interventions. However, the frequency of amputations/exarticulations in the treatment of pelvic bone tumors remains quite significant even now [3, 4].

At the current stage, most local sarcomas of the pelvic bones can be managed with organ-sparing surgical interventions. However, the frequency of amputations/exarticulations in the treatment of pelvic bone tumors remains quite significant even now [3, 4].

Historically, hemipelvectomy was first performed by Th. Bilroth in 1891, but with a fatal outcome due to hemorrhagic shock [5]. Subsequently, a successful operation, which was first mentioned in the English-language literature, was performed in 1900 by J. Hogarth-Pringle [6]. T. Kocher described removal of a pelvic tumor with limb preservation, and V. Putti cited a documented case of internal hemipelvectomy (1914) with a successful outcome [7, 8]. K. Speed popularized the term «hemipelvectomy» to describe a radical amputation through the pelvic bones and replaced it with the term hemipelvectomy. G. Gordon-Taylor used the term «hindquarter amputation» for this manipulation [9]. The modern term «internal hemipelvectomy», first given by F. Eilber in 1979 [10], is used to define a limb-sparing operation. At the same time, the operation without preservation of the limb is often called «external hemipelvectomy» in modern practice.

Studies at the beginning of the 20th century mostly described clinical cases or small groups of patients, paying particular attention to surgical anatomy and
access. The reported results of patient treatment, especially postoperative survival, were very poor [11]. In general, postoperative mortality remained excessively high throughout the first half of the 20th century. In particular, G. Gordon-Taylor reported the mortality due to hemipelvectomy in 56 % (31 out of 55 patients) in the case of treatment of pelvic sarcoma or bone tuberculosis [9, 12]. However, later the indicator decreased, and the same authors published data on 22 % mortality, explaining the significant improvement in postoperative survival of patients with significant experience and improvement of the work of anesthesiology and resuscitation services, which provided perioperative care [9].

In the middle of the last century, significant progress was made in understanding the processes of development of malignant tumors of mesenchymal tissue. It mainly concerned the definition of diagnostic categories of the tumor. For example, D. Dahlin and E. Henderson formulated the basic principles of chondrosarcoma treatment in 1956, which remain relevant today [13]:

– a necessary factor is obtaining a sufficient amount of material during a biopsy to establish a diagnosis;
– the operation is performed in such a way as to remove the biopsy site and the entire biopsy tract together with the main tumor and/or the limb without re-dissection;
– surgeons must not touch or damage the tumor, it is beyond their field of vision.

Also, D. Dahlin and E. Henderson determined that only 3.4 % of patients who received inadequate surgical treatment achieved a 10-year life expectancy. At the same time, the 10-year survival rate comprised 41% in patients treated according to the above principles. This study most clearly shows a more than 10-fold improvement in oncological survival in patients with pelvic bone tumors who receive proper treatment. Similar results in sarcoma of other bones and soft tissues finally confirmed the extreme importance of correct biopsy and removal of the tumor by en bloc resection [14, 15].

In 1960–1970 Academician O. O. Korzh, Professor M. I. Kulish and specialists of Professor M. I. Sytenko Institute (Kharkiv) developed a number of unique surgical interventions in tumors of pelvis and hip joint area with the use of alloplasty of bone and bone-joint defects. The existing radical-preserving and restorative operations on the bones of the pelvis, performed for various diseases, including benign and malignant neoplasms, have been improved. Kharkiv scientists have developed original approaches allowing surgical interventions on various parts of the pelvic bones, covering clinical and radiological manifestations of pelvic bone diseases and differential diagnosis of various nosological units and presenting features of surgical technique and analgesia in bone oncopathology. Academician O. O. Korzh has defined the basic principles of surgery for malignant bone tumors, which are relevant to this day and are the key to successful treatment of both pelvic bone sarcoma and tumors of the musculoskeletal system in general. Scientists of Professor M. I. Sytenko Institute for the first time in Ukraine have provided an evaluation of the effectiveness and conducted an analysis of errors and complications of surgical treatment of pelvic diseases [16–18].

W. Enneking became an outstanding innovator in pelvic and hip tumor surgery. He initiated the development of the modern era of pelvic sarcoma treatment, generalized, popularized and unified the principles of operative organ-sparing treatment of pelvic neoplasms [19]. The experience he accumulated helped to study the biology of tumors and formulate the principles of treatment, as well as to spread them among the surgical community, increased the role of the surgical stage in the complex treatment of malignant tumors of the pelvic area [3].

Diagnostic methods have also undergone their evolution on the example of diagnosing tumors and tumor-like diseases of the pelvic bones. Visualization of tumors of this area is still not an easy task, despite the variety of diagnostic methods. The first surgical interventions to remove tumors of the pelvic bones were performed on the basis of the data of conventional single-plane radiography, as well as the results of physical and surgical examinations. Later, simple spiral tomography was used for better visualization of bone structures in combination with intravenous angiography and bowel contrast with barium sulfate [20]. Bone osteoscintigraphy was also used, but this method lacked spatial resolution [3].

Diagnostic methods have also undergone their evolution on the example of diagnosing tumors and tumor-like diseases of the pelvic bones. Visualization of tumors of this area is still not an easy task, despite the variety of diagnostic methods. The first surgical interventions to remove tumors of the pelvic bones were performed on the basis of the data of conventional single-plane radiography, as well as the results of physical and surgical examinations. Later, simple spiral tomography was used for better visualization of bone structures in combination with intravenous angiography and bowel contrast with barium sul-
Bone osteoscintigraphy was also used, but this method lacked spatial resolution [3]. The lack of adequate visualization of the pathological process often leads to incorrect selection of the biopsy site, which causes false or incomplete diagnosis and, as a result, a poor result. In 1978 W. Enneking published a series of clinical studies involving patients treated between 1957 and 1977, drawing attention to the fact that pelvic bone resections in a third of patients were performed oncologically inadequately due to the imperfection of diagnostic measures. Tumor recurrence was observed in 100% of patients with tumor-contaminated resection margins [21].

The development of spiral computed tomography (SCT) in the 1970s significantly improved the diagnosis of pelvic bone tumors [22, 23]. CT gave surgeons two main advantages: first, a significant improvement in determining the anatomical location of the spread of bone and soft tissue sarcoma in the pelvis, the presence and size of extraosseous component of the tumor, as well as damage to the pelvic organs. Second, CT of the chest compared to X-ray or X-ray tomography of the lungs provided high accuracy in the detection of lung metastases. This advantage contributed to a more balanced approach to radical surgical treatment in patients with detected visceral (pulmonary) metastases [3].

MRI is a powerful method for diagnosing the condition of soft tissues [24], but CT remains the leading one. The latter makes it possible to comprehensively assess the features of tumor damage to the bone and adjacent soft tissues. Accurate data about a bone tumor, in particular: features of cortical damage, the spread of invasive tumor growth through the bone marrow canal, etc., help to carry out a differential diagnosis of a neoplasm. In the presence of an extraosseous component of the tumor in soft tissues, CT has certain advantages over MRI, as it makes it possible to evaluate the mineralization features of the soft tissue neoplasm using radiological specific density according to the Hounsfield scale [25]. Mineralization in a tumor outside the bone may result from ossification or calcification. A limitation of MRI is that the signal from calcium-containing tissues is variable in intensity [26, 27]. Radiological density, estimated in Hounsfield units, is a unified tool for tissue differentiation on CT scans [28]. In addition, modern software for CT diagnostic procedures allows for 3D modeling and visualization of neoplasms. It is also important to take into account the features of blood supply and topographic relationships of anatomical structures of the segment of the limb affected by the tumor. Spiral computed tomography angiography (SCTA) is fully capable of addressing these issues. Accurate data on the presence, number and location of the vessels feeding the tumor, topography of the main vascular structures of the limb and tumor (adjacency and vascular invasion) make it possible to plan the surgical stage of patient treatment, namely: to determine the type and scope of the intervention (organ-sparing surgical measures or crippling operations), possible intraoperative features (ligation of vessels feeding the tumor, angioplasty), etc. [29].

The most common primary malignant tumors of the pelvic bones are chondrosarcoma, osteosarcoma, and Ewing's sarcoma. Chondrosarcoma, specifically its central variant, remains resistant to any known type of adjuvant treatment. And the prognosis largely depends on the oncological stage of the disease, invasion into soft tissues and ablascity of tumor resection for patients with localized tumors of the pelvic bones [30].

The main revolutionary achievements in oncology were the development of chemotherapy, which significantly improved the prognosis for patients with osteosarcoma and Ewing's sarcoma. Before the era of adjuvant chemotherapy, the survival rate for clinically localized central osteosarcoma was less than 15% [14]. At the same time, specific survival rates for patients with osteosarcoma of the pelvic bones before the start of chemotherapy have not been registered. These tumors are known to have an even worse prognosis than extremity tumors, and it is reasonable to assume that long-term recurrence-free survival was not often achieved in these patients [3].

The advent of chemotherapy based on doxorubicin immediately and significantly increased the survival of patients with osteosarcoma [31]. These advances have provided a significant potential for survival in patients with high-grade axial skeletal sarcomas and opened a possibility for surgical treatment of diseases usually considered fatal and incurable [3].

At the same time, significant progress was made in the treatment of patients with Ewing's sarcoma [32], in the understanding of the use of radiation therapy in the case of damage to the pelvic bones [33].

The development of surgery for tumors of the pelvic bones took place in parallel with the progress in the surgery of the pelvic organs in the case of their diseases [34]. This made it possible to identify subgroups of patients with locally widespread forms of malignant neoplasms of internal organs and musculoskeletal disorders without distant metastases. Typical examples are locally advanced primary or recurrent colorectal cancer affecting the sacrum or...
Impairments of the locomotor system in malignant tumors of visceral organs have traditionally been a marker of their inoperability. However, over time, the in-depth study of tumor biology and the improvement of pelvic bone resection techniques made it possible to carry out extended operations on the organs of the small pelvis with the involvement of musculoskeletal structures from the mid-1980s [35, 36].

It has been subsequently proven that it is quite possible to obtain satisfactory oncological and functional results and a good rate of recurrence-free survival even in significant lesions, which greatly affects the quality of life [37]. As practice has shown, in almost all pelvic tumors, the cleanliness of the resection edges is a key factor that determines the final outcome of the treatment, emphasizing the role of wide ablastic resection [3].

Until the 1970s, almost all pelvic bone tumors involving critical parts of the pelvis were treated surgically with the help of inter-abdominal amputation. Nowadays, thanks to significant achievements in the field of bioengineering, methods of imaging abnormal processes and auxiliary (including adjuvant) methods of treatment, organ-sparing resection of pelvic bones with the use of various types of reconstruction is considered a reasonable option that allows obtaining a satisfactory oncological prognosis for patients [38]. The treatment plan for preoperative management, the type of resection, and subsequent methods of reconstruction of defects of bone structures should be determined taking into account the anatomical location and size of the tumor, the age, concomitant diseases of the patients, and the general prognosis, taking into account the stage of cancer and the degree of tumor differentiation [39–41].

Classification of pelvic bone resections according to Enneking and Dunham, dividing them into four types, has been the most common and effective way of dividing the surgical volume of operations in malignant tumors [21]. Type I resection involves the removal of the pubic bone, type II — the periacetabular area, type III — the pubic and ischial bones, type IV — the lateral mass of the sacrum (Figure).

It is possible to create multiple combinations of these types of resections that include more than one site and are classified by a combination of individual types of tumor removal (e. g., type II–III resection).

Resection of the largest part, or rather of all pelvic bone structures (type I–II–III), is called hemipelvectomcy [43–45]. In involvement of the proximal part of the femur in the resection of the pelvic bones, it is designated as type H, and is further divided into three types: H1 — removal of the femoral head, H2 — the part of the head and neck of the bone, H3 — the proximal part [46]. When the tumor affects not only the posterior parts of the pubic bone and the sacrum, but also extends to the lower parts of the lumbar spine, the operation is called an extended interpubic-abdominal amputation. This procedure involves resection of the pubic bone and sacrum, amputation of the lower limb and part of the lower lumbar spine. All details about surgical tactics, extent of resection, and methods of reconstruction of pelvic bone defects should be established based on the classification of W. Enneking and W. Dunham [21].

A key aspect of the development of surgery in general and scientific progress is the work of sharing experience and critical evaluation of results. Accordingly, in parallel with the advances of surgery for sarcoma of bones and soft tissues of the pelvis, professional organizations were created with the aim of improving treatment methods and analysis of the results of their use in patients with malignant musculoskeletal neoplasms [3]. Here are the well-known organizations in this field and the years of their foundation:

– Musculoskeletal Tumor Society (MSTS, 1977);
– International Society of Limb Salvage (ISOLS, 1981);
– European Musculoskeletal Oncology Society (EMSOS, 1987);
Connective Tissue Oncology Society (CTOS, 1995);

These specialized professional organizations continue to actively develop both the practical aspects of bone and soft tissue tumor treatment, as well as the study of tumor biology, including pelvic localization. A vivid example of their activity was the creation of a system for evaluating treatment outcomes, the work on which began in 1981 at the ISOLS symposium. As a result of the cooperation of specialists, the Musculoskeletal Tumor Society Score (MSTS) scale was created, which is still actively used in the surgery of musculoskeletal tumors [47].

Modern methods of visualization of the abnormal process make it possible to reliably determine the extent of the tumor lesion and to determine the general oncological status in patients with malignant tumors of the pelvic bones. In addition, it is currently possible to perform organ-sparing resections of these bones in most patients. The commonly accepted nomenclature for amputative resections is either «external hemipelvectom» or «interventral-abdominal amputation». Organ-sparing pelvic bone resections are called «internal hemipelvectom» and are classified according to the degree of involvement of the pubic bone, acetabulum or pubic area in the tumor process [21]. Clinical outcome, as before, is most often assessed using the MSTS scale [47].

Over time, in different parts of the world, different approaches and features of management of patients with malignant neoplasms of pelvic bones were formed. Initially, experts were focused only on tumor removal; reconstructive techniques were used quite rarely due to significant technical difficulties [10]. Recent studies demonstrate the significant viability of this approach and it remains a relevant surgical option in modern practice [48]. However, many research surgeons have found significantly better functional results in the case of restoration of femoral-sacral continuity by anatomical reconstruction after pelvic bone resections [49]. Significant difficulties arise during operations in removal of a portion of the acetabulum. A number of different approaches have been (and continue to be) used for these patients. In particular, cemented hip arthroplasty has been extremely frequent, being used more often in patients with periacetabular metastatic lesions, where the bone defect is usually smaller than in resections for primary malignancies [50]. Hip arthrodesis was also often performed, but it was technically difficult, often complicated by the development of pseudarthrosis and, as a result, extremely satisfactory functional results [51]. In the world experience of anatomical reconstructions, massive pelvic allografts or processed (autoclaved) autografts were used [52]. These operations were also technically complex and had a high complication rate.

As one of the types of bioreconstruction after resection of the acetabulum in the case of malignant neoplasms at the State Institution «Professor M. I. Sytenko Institute of Spine and Joint Pathology of the National Academy of Sciences of Ukraine» in 2018 by a group of scientists under the leadership of Professor O. E. Vyrva developed a technique for replacing the post-resection defect of the periacetabular part of the pelvis (patent No. 144210, Ukraine).

The authors proposed forming a bone block in the area of the removed acetabular zone with mobile fixation of the femoral head by using the synthetic material attachment tube and bone alloimplants in the form of chips. Over time, even in significant defects of bone tissue, the roof of the acetabulum is formed, which enables the patient to fully load the lower limb and obtain a certain amount of movement in the hip joint [53].

Saddle-shaped endoprostheses designed for revision hip surgery in cases of significant bone loss due to aseptic instability of the acetabular component of the endoprostheses or periprosthetic infection have been used to repair the hip joint after tumor resection to allow for reconstruction using modular endoprosthesis [54]. However, significant limitations of this method were subsequently revealed, so its use in clinical practice is purely individual [55]. Modern methods of reconstruction of the periacetabular zone involve installation of modular endoprostheses, individual prostheses and porous tantalum implants manufactured serially or individually [56, 57]. Each of these methods has certain advantages and disadvantages, which depend on the type of resection, the size of the bone defect, as well as the experience of the medical institution and preferences of the surgeon. The use of intraoperative navigation or pre-made individual instruments allows for accurate resections for anatomical replacement of pelvic bone defects [3].

It stands to mention that not all pelvic bone defects can be reconstructed after tumor removal. However, restoration of the pubic bone area after its resection has been described [58]. At the same time, most reports indicate that it is advisable to perform reconstruction of only soft tissues in such resections. Opinions of experts regarding the need to reproduce the removed supraacetabular part of the zygomatic cyst are also
ambiguous. Some authors are in favor of abandoning reconstruction (to minimize complications and reduce the manifestations of the Trendelenburg symptom due to medialization of the hip) [59], others have shown good results after its implementation [60].

Despite the successes in many areas of organ-sparing pelvic surgery, the role of external hemipelvectomy/inter-abdominal amputation (IAA) in clinical practice remains quite significant [61]. Currently, indications for its implementation are considered to be as follows:

– a low level of functional outcome after ablasic organ-sparing resection of the tumor is predicted. This primarily occurs when two or three critical functional elements that ensure the function of the lower limb (sciatic nerve, femoral vascular bundle and acetabulum) must be resected for radical tumor removal. This principle was defined in 1989 by M. O’Connor and F. Sim [41];

– the mass formed as a result of the resection of the soft tissue defect is so large that the wound cannot be effectively closed without the use of an amputation piece. With the development of plastic surgery for large defects, this situation is becoming less likely;

– oncological indications in patients with tumor recurrence after organ-sparing pelvic bone resection. Although many authors note the low level of functional results after IAA, the use of modern methods of exoprosthetics allows many patients to move without additional support [62].

It is extremely difficult to make a decision about amputation of the lower limb or organ-sparing surgery, since surgeons have to analyze the possibility of conducting ablasic wide resection and at the same time evaluate the technical possibility with the oncological feasibility of preserving the lower limb. When the doctor decides on limb-sparing surgery, the obtained functional results after the surgery should always be higher than those given by the IAA [3].

Oncological stages of pelvic bone sarcoma have recently been changed. An assessment by the American Joint Commission on Cancer (AJCC) showed a less favorable prognosis in the case of sarcoma located in the bones of the axial skeleton. That is why the latest eighth edition of the AJCC Staging Manual includes anatomic location (with specific criteria for pelvic tumors) to better predict the clinical outcome of these complex diseases [63]. For pelvic bone sarcomas, the T criterion is described in the eighth edition according to a different principle than for limb bone tumors. However, unlike the same sarcomas of the bones of the limbs, the stage of sarcomas of the bones of the pelvis remained undetermined [3].

Conclusions

Despite significant advances since the first attempt at hemipelvectomy in 1891, a number of challenging issues in pelvic tumor surgery remain unsolved. The need for reconstruction of pelvic bone defects after massive resections is debated. It probably provides a better functional result at the cost of higher complications, but the choice of reconstruction method and biased treatment in a specific medical institution clearly influence these results. Long-term follow-up of patients are rarely conducted and have shown the expected decline in functional outcomes in individuals over time [64].

Uncertainty about the role and method of reconstruction is increased in pediatric patients, for whom there is little published information to guide surgeons [65]. In resection of the acetabulum, reconstruction is considered in patients starting from adolescence, e. g. 14 years and older. Younger patients are mostly treated with resection arthroplasty.

The search for a more adequate, least traumatic and functionally beneficial method of reconstruction of post-resection defects of pelvic bones, both using biological materials and various types of individual/serial endoprostheses, is ongoing, especially in cases concerning the acetabulum.