International Journal of Surgery Open 16 (2019) 1–6

Contents lists available at ScienceDirect

International Journal of Surgery Open

journal homepage: www.elsevier.com/locate/ijso

Case Series

Outcomes of pelvic resection from malignant pelvic tumors. A case series

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ARTICLE INFO

Article history:
Received 25 May 2018
Received in revised form
7 November 2018
Accepted 12 November 2018
Available online xxx

Keywords:
Pelvic tumor
Pelvic resection
Oncologic outcome
Functional outcome

ABSTRACT

Introduction: Malignant pelvic tumors also tend to present late and therefore pose a major challenge for orthopedic oncologists because of the large tumor size, local extension, and complex anatomy with proximity to major neurovascular structures and intestinal and urinary tracts.
Method: We evaluated the oncologic, functional outcome and complications following internal hemipelvectomy in a consecutive series of patients with malignant muculoskeletal tumors of the pelvis at our hospital between January 2012 and December 2017. The follow-up period was defined as the length of time elapsed from the date of surgery until the death or last date of review following operation. We analyzed survival rate using Kaplan Meier method and it relation with tumor site, tumor size, and type of histology.
Result: Our clinical series of patients were composed of 4 males and 5 females. Of 9 patients, 6 cases were primary malignant bone tumor (3 cases of chondrosarcoma, 2 cases of osteosarcoma, and 1 case of Ewing's sarcoma), 2 cases of metastatic bone disease from thyroid carcinoma, 1 case from breast adenocarcinoma. The Kaplan Meier analysis showed 1 year and 2 years survival rate were 88.9% and 66.7% respectively. The functional outcomes showed the mean MSTS score 16.5 point. The lowest score was 9, which was pelvis resection type I + II on osteosarcoma case. The highest score was 25, which was pelvic resection type II + III and reconstruted with iliofemoral arthrodesis. There was 1 case intra-operative bleeding and 2 cases post operative infections.
Conclusions: The majority of pelvic tumor underwent pelvic resection is chondrosarcoma. Functional outcome MSTS score was still comparable with previous study. Complications of pelvic surgeries were bleeding and infection. In our small case series, pelvic resection with wide margin and reconstruction of bone defect may give good local control and clinical outcome.

1. Introduction

A large number of musculoskeletal tumors can affect the pelvis. Several studies have reported bone sarcomas as the most common lesions, followed by soft tissue sarcomas and metastatic lesions. The most frequently observed pelvic sarcoma is of cartilaginous origin (chondrosarcoma), followed by osteosarcoma [1]. Ten to 15% of all primary bone tumors are located in the pelvic bone of which chondrosarcoma in adults, Ewing's sarcoma in children, and osteosarcoma in adolescents represent the most common histological subtypes [2]. The most common location of pelvic lesions is the type with involvement of only one region, mainly the iliac, followed by the region of the ischial and iliopubic ramus and the acetabulum [1].

These tumors also tend to present late and therefore pose a major challenge for orthopedic oncologists because of the large tumor size, local extension, and complex anatomy with proximity to major neurovascular structures and intestinal and urinary tracts. Subsequently, these are also associated with less favorable outcomes in terms of prognosis and survival compared with other extremity tumors [3]. Even today, treatment of pelvic sarcomas remains one of the most predominant challenges for orthopedic oncologists due to the proximity of visceral organs and neurovascular structures [2].

Previously, the mainstay for treatment of malignant pelvic tumors was external hemipelvectomy (hindquarter amputation), but with advancements in surgical techniques and chemotherapy and radiation therapy, limb salvage procedures (internal hemipelvectomy) have also emerged as viable modalities. This involves

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https://doi.org/10.1016/j.ijso.2018.11.005

resection of the lesion with part or all of the hemipelvis, but preserving the ipsilateral lower extremity. In patients in whom the tumor has not invaded major neurovascular structures, wide resection is possible, and therefore internal hemipelvectomy can be considered without affecting the functionality of the limb. However, in cases of neurovascular invasion and where widemargin excision is difficult, external hemipelvectomy remains the preferred option [4].

Currently, so much experience has been gained from primary tumor surgery that limb-salvage surgery is also performed for pelvic metastases in patients with favorable cancer types, solitary metastases, and long expected survival rates. The advantages of amputation over resections at the pelvis are a lower incidence of complications, a limited area at risk for recurrence, and a faster recovery time compared with all but the most limited pelvic resections. The disadvantages, especially after periacetabular resections, are the inevitable discrepancy in leg length and impaired hip and gait function [4].

Survival after hemipelvectomy might be related to several different factors, such as tumor histopathology and size, disease stage, patient physical status, and resection type. In patients with soft tissue tumors, the 5-year survival rate might be as low as 10%. Large tumors and bone and vascular involvement might be indicators of poor survival. For bone tumor resection, the 5-year survival rate can be as high as 100%, depending on the resection type. A large previous series reported a survival rate of 50% after hemipelvectomy [5]. Postoperative complications are not uncommon. Large dissections can compromise the viability of the muscle flaps and a large dead space is often present, which leads to the formation of collections, and consequently surgical site infection [1].

In this study, we evaluated the oncologic, functional outcome and complications following internal hemipelvectomy in a consecutive series of patients with primary malignant muculoskeletal tumors and secondary (metastatic) bone diseases of the pelvis. This study has been reported in line with the PROCESS criteria [6].

2. Methods

We reviewed malignant pelvic tumors which were treated with pelvic resection at our hospital between January 2012 and December 2017. The surgeries were conducted by two musculoskeletal oncologic surgeon. Among 10 patients, there was 1 patient that cannot be reviewed due to loss of control.

At the time of diagnosis, all the patient underwent a plain radiography and magnetic resonance imaging (MRI) of the affected site, chest plain radiography or computed tomography scan. On Pelvis MRI we evaluated the size of the tumor and categorized tumor largest diameter size into <10 cm, 10–20 cm, and >20 cm (Fig. 1). After imaging completed, the patient underwent biopsy. Final diagnosis and treatment of every case were established in the clinico-pathological meeting regarding the clinical, radiological, and histopathological findings. The type of pelvic resection was based on Enneking and Durham classification (Fig. 1) [4,7,8].

The follow-up period was defined as the length of time elapsed from the date of surgery until the death or last date of review following operation. We evaluated oncologic and functional outcomes, and complications during and after surgery. Overall survival was taken from the date of surgery to the last date when the patient was documented to be alive or the date of death.

In case of survival, functional outcome and complication were evaluated which performed at a final out patient visit or a telephone interview. Functional outcome was defined based on musculoskeletal tumor society (MSTS) functional classification [9]. The scale is composed of six items such as pain, function, emotional acceptance, external support, walking ability and gait.

3. Results

Our clinical series of patients were composed of 4 males and 5 females, whose mean age was 37.7 (range, 14–62 years). These patients had a mean follow up period of 16.1 months (range, 3–63 months).

Based on histopathology result, 6 cases were primary malignant bone tumor (3 cases of chondrosarcoma, 2 cases of osteosarcoma, and 1 case of Ewing's sarcoma), 2 cases of metastatic bone disease from thyroid carcinoma, another 1 case from breast adenocarcinoma (Table 1). Among 6 cases of malignant primary bone tumor, based on Enneking staging, 5 cases were stage II B, and 1 case was stage III.

The average duration of surgery was 567.8 minutes (range 420–870 minutes). The longest duration of surgery was pelvis resection type I + II and reconstruction using fibular graft and arthrodesis. The fastest surgery in this study was 420 minutes when performing type I pelvic resection. The average blood loss was 3233.3 cc (range: 1000–11000 cc). The most blood loss during surgery was on pelvic resection type I + II for osteosarcoma.

As the method of resection, there were 1 case type I (P1 ilium), 5 cases type I + II (P1P2 ilium and periacetabular), 1 case type III (P3 pubic), 1 case type II + III (P2 P3 periacetabular and pubic), and 1 case type I + II + III (P1P2P3). All cases were resected with wide (clear) surgical margin. Among 5 patients with type I + II resection, 1 case was reconstructed with extracorporeal irradiation autograft and total hip replacement, and 1 case was reconstructed with bone cement spacer + THR, and the others were constructed with arthrodesis. Among patient with arthrodesis, in two cases we used non vascularized fibula graft, plate, and screws to achieve sacrofemoral arthrodesis. Other case was ischiofemoral arthrodesis using reconstruction plates and screws. One patient with type II + III resection was reconstructed with iliofemoral arthrodesis using reconstruction plate and screws. One patient with type I + II + III resection were reconstructed using non vascularized fibular graft to fuse between the remaining of iliac bone and femur. One patient with type III pelvic resection was not reconstructed. One patient with type I resection underwent reconstruction with bone cement spacer to fill the gap between sacrum and acetabulum (Fig. 2).

Chemotherapy and radiotherapy were not conducted in chondrosarcoma cases. Patient with Ewing's sarcoma received neoadjuvant chemotherapy and radiotherapy before surgery and adjuvant chemotherapy following surgery Patient with osteosarcoma received neoadjuvant chemotherapy and adjuvant chemotherapy following surgery. Patient with breast and thyroid adenocarcinoma received radiotherapy after surgery.

The follow up ranged from 3 until 63 months (mean: 16.1 months). At the time of surgery, there was one case with pulmonary metastasis. Overall, 3 patients died until the last follow up due to the consequence of metastasis to the lung. The mean interval from surgery to death was 16.6 months (range: 12–21 months). Local recurrence was not identified in all patient until last follow up. The Kaplan Meier analysis showed 1 year and 2 years survival rate were 88.9% and 66.7% respectively.

At a final follow up, the functional outcomes were evaluated in 9 patients which showed the mean MSTS score 16.5 point. The lowest score was 9, which was pelvis resection type I + II on osteosarcoma case. The highest score was 25, which was pelvic resection type II + III and reconstruted with iliofemoral arthrodesis (Table 2).

There were three complications from 9 patients in this study. There was 1 case intraoperative complication and 2 cases post operative complication. One case of complication during surgery

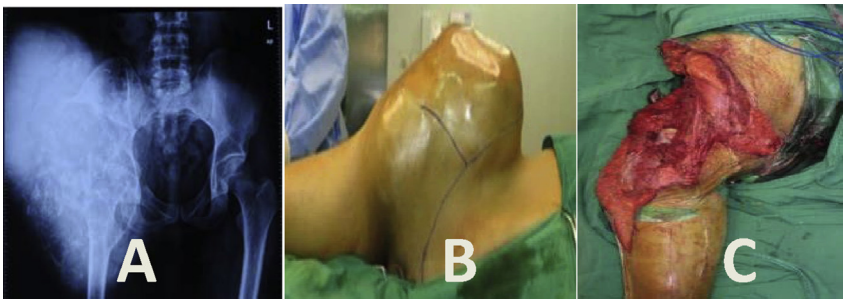

Fig. 1. (A) Anteroposterior plain radiograph of a patient with chondrosarcoma with diameter tumor size > 20 cm. (B) Clinical picture and utilitarian pelvis incision design. (C) Post internal hemipelvectomy.

Table 1
Patient, tumor, and surgery type of 9 patients underwent internal hemipelvectomy.

Case	Age/ Sex	Histology	Tumor site	Resection type	Tumor size[a]	Reconstruction	MSTS score	Complication	Survival[b]	Follow up[b]	Metastasis
1	51/F	Breast Adeno Ca	P3	III	<10	no reconstruction	10	–	17	17	lung
2	23/M	Ewing sarcoma	P1,P2	I + II	10–20	autograft ECI + THR	10	infection, second operation	21	21	lung
3	33/F	Osteo sarcoma	P1,P2	I + II	>20	fibula graft + arthrodesis	9	intraop massive bleeding, second operation	12	12	lung
4	37/M	Chondro sarcoma	P1,P2,P3	I + II + III	>20	fibula graft + arthrodesis	19	–	alive	9	–
5	31/M	Chondro sarcoma	P3	II + III	10–20	arthrodesis	25	–	alive	10	–
6	14/M	Osteo sarcoma	P1,P2	I + II	10–20	fibula graft + arthrodesis	19	–	alive	6	–
7	52/F	Chondro sarcoma	P1,P2	I + II	10–20	arthrodesis	20	–	alive	4	–
8	37/F	Thyroid Adeno Ca	P1,P2	I + II	10–20	bone cement spacer + THR	20	–	alive	3	–
9	62/F	Thyroid Adeno Ca	P1	I	10–20	bone cement spacer	18	infection, second operation	alive	63	lung

[a] in centimeter.
[b] in month.

was massive bleeding. We needed to stop the operation and performed next surgery one week later. All of postoperative complications were an infection that needed the second operation for debridement.

4. Discussion

Pelvic resections are challenging and complex. They are technically difficult due to sometimes the size of the tumor is very large, and anatomically it closes to pelvic viscera and neurovascular structure. The patient usually needs a large amount of blood transfusion and postoperative intensive care. As shown in our series, mean time of surgery was 567.7 minutes. A Study from Umer et al. showed mean time of surgery was 356 minutes [3]. The mean intraoperative blood loss in our study was 3233.3 cc, and compare to Umer et al. study that showed 1900 cc blood loss [3]. Our study showed longer time of surgery and more blood loss during surgery, it was probably due to most of the tumors in our study (72%) were 10–20 cm in size, compare to Umer et al. which showed 62,5% of the tumor had a diameter less than 10cm. Although study from Delloye et al. showed a quite similar result with average blood loss 4359 cc (range 1000–11.300 cc) [10].

Complications after pelvic tumor resection are very common. Complications that had been reported were infection, hematoma, wound healing problem, nerve injury, ureter injury, bladder injury, bowel injury, extensive hemorrhage, thrombosis, and implant failure [11–13]. The complication rate in our study was 33.3%, it was lower compared to other studies that showed 50–60% [13–16]. The most common complication was the infection. As shown in our study, 2 from 9 surgeries (22.2%) had postoperative infection that needed debridement and antibiotic treatment. As shown from other studies, infection rate was reported 21–29.2% [2,3,17].

There are many factors that increase infection. During pelvic resection, many adjacent muscles and soft tissues should be dissected to obtain a safe margin that sometimes leading to devascularization and create dead space. Prolong surgical time during pelvis resection increases the risk of infection. Reconstruction technique also involved in increasing infection after surgery. The allograft and reimplantation of resected bone after autoclaving or irradiating play a role like a foreign body that fills the empty space, this greatly increases the risk of infection [12,18]. One of our infection cases was reconstructed by reimplantation of resected bone after extracorporeal irradiation (ECI), according to other studies rate of infection of this procedure was 16–23% [19–21].

One of our complication in our study was extensive hemorrhage during pelvic resection type I + II for osteosarcoma, with the diameter of tumor >20 cm. A wide variety of risk factors influence perioperative blood volume loss. Resection of massive pelvic tumor always requires a longer period of time and high-grade malignancy always has an abundant blood vessel supply. There are studies that suggest performing embolization before pelvic resection surgery. However the indication for embolization in these cases still uncertain. According to Tang et al. limb salvage surgery for pelvis tumors is mainly influenced by the location of the tumor, volume, and surgery time. Pelvis tumor that involves the acetabulum and sacrum, have a volume greater than 400 cm^3, and anticipated

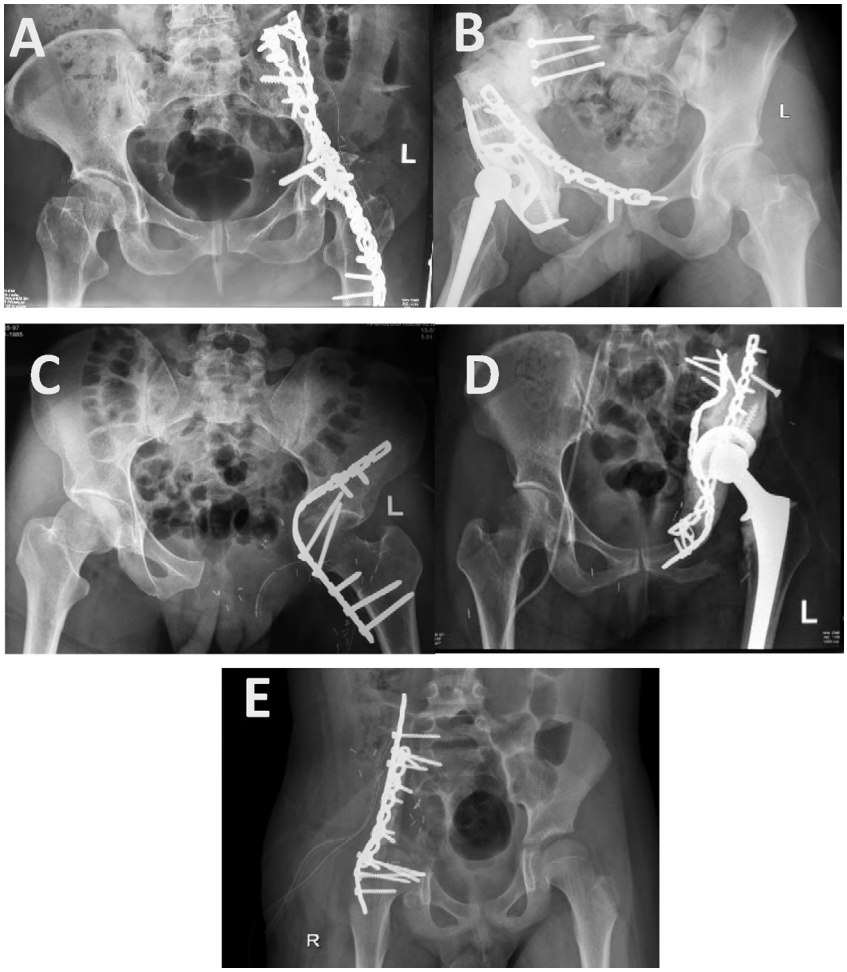

Fig. 2. Anteroposterior plain radiographs of the pelvis. (A) Hip arthrodesis following resection type I + II. (B) ECI and THR following resection type I + II. (C) Hip arthrodesis following resection type II + III. (D) Bone cement spacer + THR following resection type I + II. (E) Nonvascularized fibula graft + arthrodesis following resection type I + II.

Table 2
Functional score according to MSTS score.

Case	Pain	Function	Emotional	Support	Walking	Gait	Total
1	3	1	3	0	1	2	10
2	3	3	2	0	1	1	10
3	2	1	3	1	1	1	9
4	4	3	4	2	3	3	19
5	4	4	5	5	4	3	25
6	5	4	5	1	2	2	19
7	5	4	5	0	3	3	20
8	5	4	5	0	3	3	20
9	5	4	5	1	1	2	18
Average	4	3,11	4,11	1,11	2,11	2,22	16,5

operation time more than 200 minutes are likely to have a large amount of blood loss [22]. Therefore, more practiced surgical skill and simple reconstruction method which would result in less operation time were important. A Large amount of transfused blood and platelets should be prepared and blood vessel control should be considered before and during surgery.

Our study showed no local recurrence until last follow up. It was probably due to we can achieve wide margin during surgery. Han et al. showed that surgical margin was the factor most closely related to the local recurrence [13]. Several studies showed local recurrence rate 12, 5–19% [3,17,23]. Puchner et al. estimated probability of experiencing local recurrence according to competing risk (CR) analysis was 5%, 14%, and 14% at 1, 5, and 10 years respectively [2]. Length of follow up in our study was still limited compared to those studies, more than half of our cases were followed less than 1 year. This can affect lower local recurrence rate than other studies. Meanwhile, the overall survival rate in this study using Kaplan Meier analysis for 1 year and 2 years were 88.9% and 66.7%. These had the same result with study from Umer et al. [3].

A total 3 patients (33.3%) developed metastatic disease to the lung after surgery. The overall distant metastasis rate after pelvic resection had been shown to range from 26 to 29.2% [3,16]. These results were almost similar to our study. In our study, metastasis to the lung occurred in one osteosarcoma patient, and two in metastatic disease from carcinoma. In our study, the patient with osteosarcoma who developed metastasis to the lung died after 12 months of surgery. Fuchs et al. reported the outcome of pelvic osteosarcoma, the cumulative incidence of metastasis with death as a competing risk factor was 48% at 5 years, and with overall survival 5 years was only 38%. The overall survival rate of a patient with osteosarcoma of the pelvis was lower compared to extremity osteosarcoma [24]. As expected, this osteosarcoma patient in our study had tumor diameter >20 cm, large volume tumor resistance to chemotherapy is associated with higher risk of pulmonary metastases and carry a very poor prognosis in pelvis osteosarcoma [25].

Patient with Ewing's sarcoma in our study presented with lung metastasis at the time of surgery, and died 19 months after surgery. This patient had delayed performing surgery because choosing

non-surgery treatment (chemotherapy + radiotherapy) at first. Despite advance in adjuvant therapy, Ewing's sarcoma of the pelvis remains an anatomic site with a poor prognosis, 5 years survival rate approximately 50%. The use of neoadjuvant chemotherapy, followed by secondary local control for the primary site with surgical resection and radiation is an accepted regimen. Retrospective studies of Ewing's sarcoma treatment suggest that survival rates improve and local recurrences decrease significantly when surgery is performed. The patient underwent surgery with or without adjuvant chemotherapy had higher survival rates than those treated with radiotherapy or chemotherapy alone [26–28].

The pelvis has functions to transmit the weight of the upper body to the lower extremity and contains the hip joint. If there is bone loss due to a tumor and wide excision, then the surgeon needs to restore femorosacral continuity for weight-bearing [13]. In this study, we always did reconstruction to maintain continuity between sacrum and femur. We did not reconstruct tumor in P3 location as it did not involve in weight-bearing.

The reconstructive option today include using autograft, allograft, patient sterilized resected bone, custom-made prosthesis, saddle prosthesis, stemmed prosthesis, resection arthrodesis, and local improvised reconstruction using plate screw with bone cement. Each reconstruction modality has its own risks and benefits [4,29]. In our study patients that involved pelvis resection type II were reconstructed with either arthrodesis or total hip arthroplasty to achieve mobile hip. Beside that, we always tried to preserve continuity between sacrum and femur bone as in pelvis resection type I, we reconstructed with either bone cement spacer or autograft. We did not use a saddle or stemmed prostheses for reconstruction because those were unavailable in our country and very expensive.

The principle of arthrodesis after pelvis resection is to achieve solid union between proximal femur and the remaining pelvis (iliofemoral, ischiofemoral, or sacrofemoral) using plate, screw, or wire. The disadvantage of arthrodesis includes loss of the hip joint function. Fusion rates of femoropelvic arthrodesis are <50%, in most patient, a stable and painless pseudoarthrosis develops, but with a comparable functional result to that of the alternative reconstruction options [4]. Arthroplasty reconstructions are recommended when adequate ilium and pubis remain for fixation of pelvis allograft or autograft composite with a unipolar or total hip prosthesis. We used some technique according to Kolender et al. to replace mega prosthesis after pelvis resection [4,29,30].

In our study mean MSTS score was 16.5 point or 55%. Systemic review by Shao et al. showed MSTS score ranged widely from 16% to 100%, in the studies of the last 10 years, the mean MSTS scores were more congruent, ranging from approximately 50%–70%. Regarding the specific type of reconstructive technique used, the protheses group demonstrated a mean MSTS score that ranged from 33.4% to 72.0%. The allograft reconstruction group showed a mean MSTS score ranging from 54.8% to 75.4%. The arthrodesis group reported a mean MSTS score ranging from 56.2% to 69.2% [29]. In our study mean MSTS score arthrodesis groups were 18.4 point or 61.3%, and non arthrodesis group were 14.5 point or 48.3%. It was still comparable with Shao et al. study.

5. Conclusion

The majority of pelvic tumor underwent pelvic resection is chondrosarcoma. Functional outcome MSTS score was still comparable with previous study. Complications of pelvic surgeries were bleeding and infection. In our small case series, pelvic resection with wide margin and reconstruction of bone defect may give good local control and clinical outcome. Since complication in surgery of pelvis tumor is quite high, approach in the management of pelvis tumor needs multidiciplinary team.

Ethical approval

This is a case report, patient' written consent has been obtained and will we available on request.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Author contribution

MW contributed to performed the operation, data collection, analysis and interpretation, manuscript drafting, revising, and approval for publishing; AFK contributed to performed the operation, data collection, analysis and interpretation, manuscript drafting, revising, and approval for publishing; YP contributed to assist the operation, data collection, analysis and interpretation, manuscript drafting, revising, and approval for publishing.

Conflict of interest statement

The authors declare that there is no conflict of interests regarding the publication of this paper.

Guarantor

Guarantor in this study is AFK.

Research Registration Number

researchregistry3660.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.ijso.2018.11.005.

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