

## Original Articles

# Tumour Prosthetic Replacement in Paediatric Bone Cancer

KC WONG, SM KUMTA, KW CHEUNG, KH CHIU, LF TSE, MK SHING, CK LI

### Abstract

Surgical management of paediatric bone sarcoma is challenging. Effective chemotherapy and advances in surgical techniques make limb sparing surgery possible and offers adequate disease control comparable to the results obtained by amputations. With advances in design, material technology and surgical techniques, modern implanted tumor prostheses are more durable, less short term complications and can achieve good limbs function. This study is to review the experience of using prostheses in limb sparing surgery for 35 patients with paediatric bone sarcoma managed at one of quaternary referral centers in Hong Kong.

### Key words

Limb sparing surgery; Paediatric bone cancer; Tumour prostheses

### Introduction

Osteosarcoma is the most common primary malignant bone tumours in children. It occurs most often in the bones of lower extremities and in humerus. The development of effective chemotherapy has increased dramatically the survival of patients with osteosarcoma.<sup>1</sup> In addition, the advances in imaging and surgical techniques enables

surgeons a more accurate preoperative planning and limb sparing surgery is feasible. Results of multiple studies available in the literature support the use of limb sparing techniques if an adequate margin of resection can be achieved. Limb sparing surgeries offer adequate disease control comparable to the results obtained by radical amputations.<sup>2,3</sup> Reconstruction in limb sparing surgery can be in the form of arthrodesis or arthroplasty. Arthroplasty replaces a natural joint with an artificial joint. It can be accomplished by either cadaveric allograft or tumour metallic prosthesis. As there are high rate of complications in allograft reconstruction<sup>4-7</sup> and limited bone donors in Hong Kong, we have shifted totally from using allograft to tumour prostheses in limb sparing reconstruction since 2003. The study is to investigate our experience of using tumour prostheses in paediatric bone sarcoma and the advances in this field will be discussed.

Department of Orthopaedics and Traumatology, The Chinese University of Hong Kong, Prince of Wales Hospital, 30-32 Ngan Shing Street, Shatin, N.T., Hong Kong, China

KC WONG (黃國全) *FRCSEd(Ortho)*  
SM KUMTA (古明達) *MS(Ortho), PhD*  
KW CHEUNG (張堅穎) *FRCSEd(Ortho)*  
KH CHIU (趙國慶) *FRCSEd*  
LF TSE (謝龍峰) *FRCSEd(Ortho)*

Lady Pao Children's Cancer Centre, Department of Paediatrics, Prince of Wales Hospital, The Chinese University of Hong Kong, 30-32 Ngan Shing Street, Shatin, N.T., Hong Kong, China

MK SHING (成明光) *MRCP, FHKAM(Paed)*  
CK LI (李志光) *MD, FHKAM(Paed)*

Correspondence to: Dr KC WONG

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### Methods

From July 2003 to July 2008, we studied 35 patients (17 males, 18 females), between 6 and 22 years (mean, 15 years) of age, with primary malignant bone sarcoma treated by limb sparing surgery with reconstruction of prostheses at one of the quaternary referral centers of paediatric cancers in Hong Kong (Table 1). Tumour types

**Table 1** Demographic data for patients treated with metallic prostheses

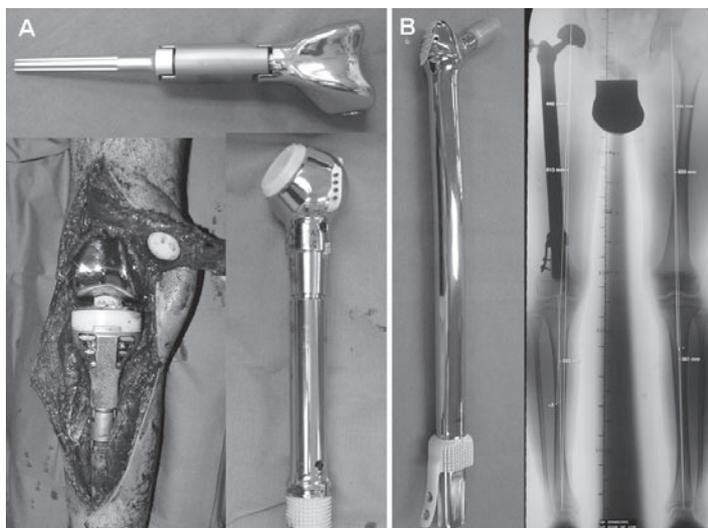
	Patients with primary tumours	Patients with revision surgery
Number	21	14
Mean age	14 (6 to 20)	16.6 (6 to 22)
Male : Female	11:10	6:8
Type of prostheses		
Modular	9	4
Custom-made (expandable)	12 (6)	10 (8)
Site		
Femur	14	8
Tibia	4	4
Humerus	2	2
Pelvis	1	0
Operative time (hours)	4.2 (2.3 to 6.6)	4.8 (2.5 to 7)
Follow-up (months)	40.4 (14 to 71)	46.5 (13 to 67)
Complications	<ul style="list-style-type: none"> <li>• Total 5 patients (23.8%)</li> <li>• 3 early postoperative wound edge necrosis</li> <li>• 1 delayed acute infection and one chronic infection</li> <li>• 1 unexplained knee pain</li> </ul>	<ul style="list-style-type: none"> <li>• Total 1 patient (7%)</li> <li>• 1 delayed acute infection</li> </ul>
Mortality	7 (33.3%)	1 (7.1%)
Limbs function by MSTs score*	28 (24 to 30) (93% of the normal function)	27.2 (24 to 29) (90.6% of the normal function)

\*MSTs, musculoskeletal tumor society score for assessing function after limb-sparing surgery; total score and normal function is 30 points.

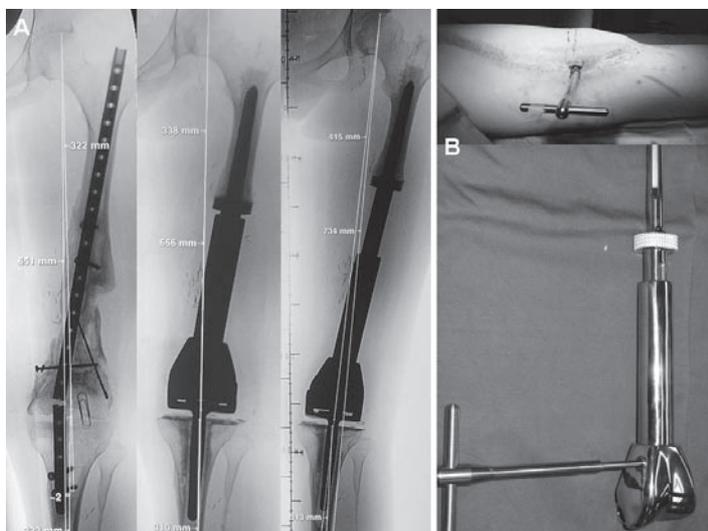
included osteosarcoma (34) and Ewing's sarcoma (1). Locations of tumour were femur (22), tibia (8), humerus (4) and pelvis (1). Twenty one patients had primary tumour resection and reconstruction following neoadjuvant chemotherapy (according to the standard protocol from Hong Kong Paediatric Haematology & Oncology Study Group). Fourteen patients who were in disease remission of their previously treated primary bone sarcoma underwent revision surgery as they experienced functional disability as a result of complications from limb sparing surgery with allograft reconstruction. Custom-made tumour prostheses were used if the size of modular system did not match with the skeletal defect created after tumour resection or removal of previous allograft (Figure 1). Custom-made expandable tumour prostheses (minimally invasive or noninvasive type) were used if leg shortening was greater than two centimeters or anticipated at the end of skeletal maturity (Figures 2 & 3). For proximal tibia tumours, reconstruction of extensor

mechanism was performed for better knee function and local muscle flaps were used for adequate soft tissue coverage of prostheses in order to minimise chance of infection. Leg lengthening procedures were performed under general anesthesia after the first index surgery when patient developed leg shortening. Immediate weight-bearing walking with protective brace was allowed soon after wound was satisfactory at the first week of operation. Patients attended intensive physiotherapy to restore joint motion and muscle control during period of postoperative chemotherapy. Plain radiographs were taken regularly to assess any bone ingrowth at bone-implant junction and any evidence of implants loosening.

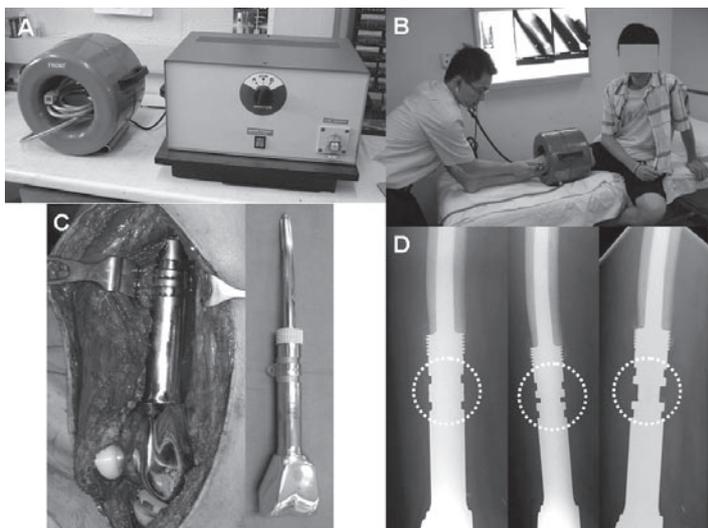
The data recorded included the number of custom-made or modular tumour prostheses used; operative time; complications; disease status; limb function as assessed by using Musculoskeletal Tumor Society (MSTs) score.<sup>8</sup>



**Figure 1** (A) Modular tumour prostheses for different regions (distal femur, proximal tibia and proximal humerus). (B) Custom-made femur tumour prosthesis for subtotal femur tumour resection.



**Figure 2** (A) A patient had failed limb sparing surgery with previous allograft reconstruction. (B) After revision surgery with prosthetic reconstruction, the leg was gradually lengthened by inserting and turning with a metal key via a one centimetre skin incision. (C) Expandable custom-made tumour prosthesis.



**Figure 3** (A) The electromagnetic external drive machine. (B) The limb with implanted prosthesis is placed at the centre of a rotating electromagnetic field created by the external drive machine. The powerful magnet mounted inside the prosthesis is captured to rotate in synchrony and this allows expansion of prosthesis at a rate of 1 mm in 4 minutes. (C) By putting a stethoscope over patient's bone, surgeons can check the proper functioning of the non-invasive expandable prosthesis by listening to the noise from the rotating magnet inside the prosthesis. (D) X-ray showed the expansion of the prosthesis at the telescopic junction.

## Results

The mean follow-up was for 42.8 months (13-71 months). At this stage, 28 patients (77.1%) remained alive. Seven patients died of disseminated disease while one patient died of systemic sepsis at early postoperative period. Twenty two custom-made tumour prostheses (62.9%) and thirteen modular prostheses (37.1%) were used. The average operative time was 4.4 hours (2.3-7). Fourteen expandable custom-made prostheses (13 minimally invasive; one noninvasive) were required for reconstruction and subsequent leg lengthening procedures. Five out of 14 patients with expandable prostheses had completed the whole lengthening process, with a mean total lengthening of 4.6 cm achieved, requiring on average, 2.4 operations. The patient with noninvasive expandable prosthesis had undergone three noninvasive lengthening at outpatient clinic (12 mm expansion of prosthesis with 4 mm per session). Three out of 21 patients (14.3%) in the group with primary tumours developed early postoperative wound edge skin necrosis that required operative debridement. One patient therefore had his postoperative chemotherapy regime postponed for one week. There were no other patients who were withheld or postponed of postoperative chemotherapy because of operation related complications. One patient developed solitary bone metastasis in the same operated leg 5 years after the first index surgery. She underwent further chemotherapy and limb sparing surgery with prosthetic reconstruction. She finally received hip disarticulation after another soft tissue local recurrence 8 months later. She remained disease free 2 years after the last operation.

Two patients (5.7%) developed acute prosthetic infection secondary to fever and sore throat, one year and four years after the first index operation respectively. Both recovered after immediate operative drainage, debridement and 6 weeks of antibiotic. One patient (2.9%) had severe prosthetic infection at postoperative four years. He only seek medical advice after he experienced leg pain for 3 months following fever and upper respiratory symptoms. The uncontrollable infection finally ended with an amputation. One patient (2.9%) has several operative debridement because of suspected metal allergy.

For 25 patients who survived and retained their limbs at latest follow-up, the mean functional MSTS score was 28 out of 30 (93% of the normal function) in the group with primary tumours while 27.2 out of 30 (90.6% of the normal function) in the revision group. For patients with tumour prostheses of lower extremities, all could walk unaided

except one patient who had hip disarticulation following local recurrence five years after the first limb sparing surgery. Follow-up plain radiographs showed satisfactory bone ingrowth at bone-implant junctions in all except three patients who were at the early study period. There was no evidence of loosening or mechanical failure of implants that required further revision surgery.

## Discussion

Tumour resection with adequate surgical margin and a functional residual limb with increased patient survival are the goals of modern orthopedic oncology. The goal of limb sparing surgery is to preserve a useful functioning limb without increasing risk to patients. The surgery should enable patients have durable, painless, mobile and stable functional limbs. The use of structural cadaveric allograft for reconstruction of large bone defects after tumour resection was a common treatment option in the past. However, reconstruction with avascular allografts had resulted in significant morbidity. Complications of allograft reconstruction included a high rate of infection, nonunion and fracture that led to failure in approximately 50% of cancer cases.<sup>4-7</sup> In view of reported and our own experience about the complications of allograft reconstruction, our unit had utilised tumour metallic prosthesis as the main reconstructive method in limb sparing surgery of primary paediatric bone sarcoma since 2003. Our mid-term results showed that patients could attain excellent limbs function. Short term complications were minimal and the rapid functional recovery could facilitate the postoperative chemotherapy regime. Protective brace was needed only for initial postoperative period about four to eight weeks. All patients could walk unaided. It was in contrast with allograft reconstruction that patients might require long term brace protection and protective weight-bearing walking.

We are now managing more long-term disease survivors with primary bone sarcoma due to advances in chemotherapy and improvement in surgical techniques. These patients sometimes mainly suffer from the complications of previous limb sparing surgery but not the disease itself. Our results showed that this group of patients could achieve excellent limbs function after revision surgery with tumour prosthetic reconstruction. In the past, there were no other good surgical options. The patients could either accept the functional disability or have amputation if the disability is severe.

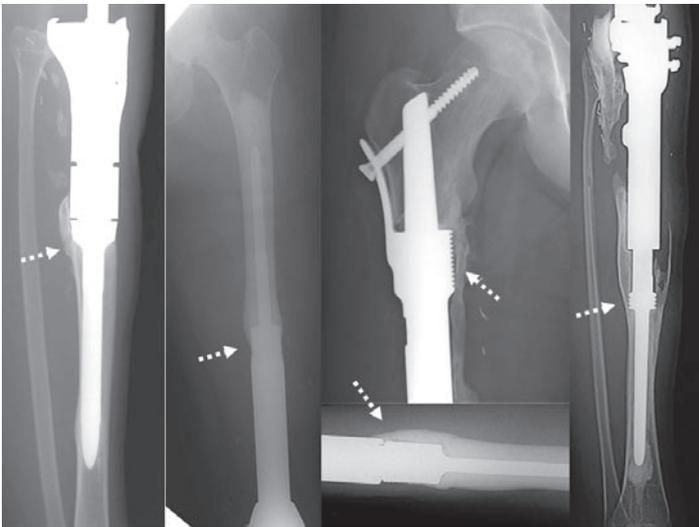
Weight-bearing part of knee joint in modern tumour

metallic prostheses simulates near normal joint function that allows good flexion-extension and some freedom of rotational movement. Hydroxyapatite coating on metal surface and additional bone graft at the bone-implant junction enhance the bone ingrowth to the metal prostheses (Figure 4). The modern knee design and the new concept of bone ingrowth at bone-implant junction actually improve survival of prostheses significantly.<sup>9,10</sup> Being an artificial joint, wear of articulating surface and mechanical loosening are anticipated in the long run. Modern design tumour prostheses are however more durable and long lasting, the risk of revision for mechanical loosening for distal femur and proximal tibia tumour prostheses were reported to be only 0% and 25% at 10 years respectively.<sup>9,10</sup>

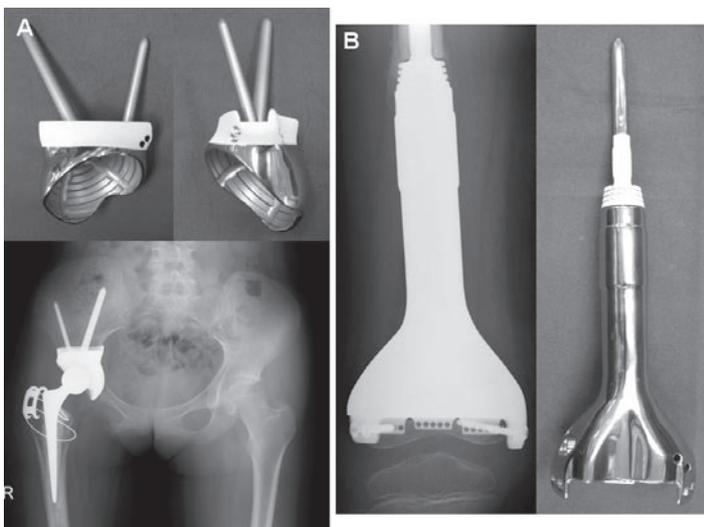
Modern tumour prostheses include modular and custom-

made types. The modular type consists of components with different standard sizes. They are off-the-shelf items and can be assembled to match the length of bone resected at the time of surgery. Surgeons therefore have more intraoperative flexibility for bony reconstruction. However, the commercially available modular prostheses are designed and based on the bone geometry of Caucasian and our experience suggested that they might not fit well to the bones of local Chinese with small body built.<sup>11</sup> Therefore, we have increasing demand for custom-made prostheses that are more suitable for local individual requirement.

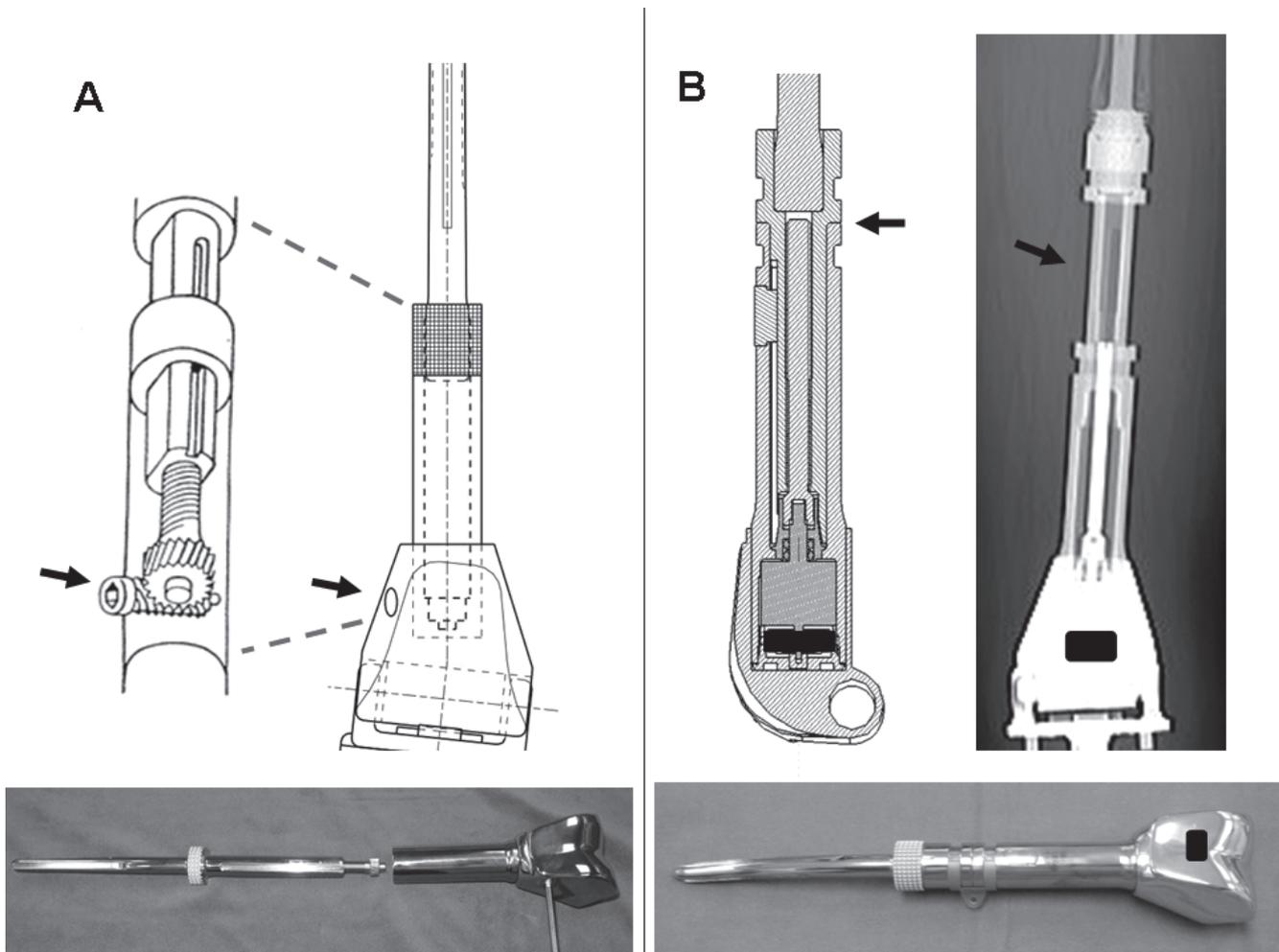
Custom-made prostheses are designed and manufactured precisely according to the exact bone dimension from patients' own computer tomography datasets (Figure 5). Computer navigation technology was recently introduced



**Figure 4** Solid bone ingrowth at bone-implant junction (white dotted arrows) increases the longevity of modern tumour prostheses.



**Figure 5** (A) Custom-made pelvic tumour prosthesis. (B) Custom-made joint saving intercalated tumour prosthesis.



**Figure 6** (A) Illustrates the internal structures of minimally invasive expandable prosthesis. Black arrow shows the key port that is connected to the gearbox mechanism of telescopic prosthesis. (B) Illustrates the internal structure of noninvasive expandable prostheses. Black rectangle represents the site of magnet inside the implanted prosthesis. Black arrow points to the site where the prosthesis expands. The maximum extension that an expandable prosthesis can allow is already pre-determined by the length of implanted prosthesis. (Pictures are reproduced with permission from Stanmore Implant Worldwide Limited, United Kingdom.)

to help surgeons reproduce surgical planning and insert the custom prostheses more accurately.<sup>12,13</sup> As it normally takes the overseas implant company three-four weeks from manufacturing to actual delivery of prostheses, MRI scan should be repeated at week 5 of neoadjuvant chemotherapy to ensure that patient responds to chemotherapy and there is no tumour progression prior to final purchase of custom-made prostheses.

A relatively novel technique of limb sparing surgery is the use of a custom-made expandable prosthesis for skeletally immature patients with primary bone sarcoma.<sup>14,15</sup> The location of these tumours in the growing areas of bone commonly mandates removal of the

affected growth plate. Subsequent continued growth in the contralateral extremity results in limb length inequality. The expandable prostheses can be gradually extended to correct limb shortening. For minimally invasive expandable prostheses (Figure 6A), the lengthening procedure is performed under general anaesthesia. A key is inserted into a key port of the implanted prosthesis via a skin incision of one centimeter. Turning the key rotates the gearbox mechanism inside and allows longitudinal expansion of telescopic prosthesis. The most advanced expandable prostheses allow noninvasive lengthening without surgical operation (Figure 6B). When the limb with the implanted prosthesis is placed at the center of a rotating

electromagnetic field, a powerful magnet mounted inside the implant is captured to rotate and cause the implant to expand. During extending procedures, the patients report no discomfort, no abnormal sensation and no noise. Patients are able to move and walk immediately after these outpatient procedures.

In general, patients with implanted prostheses have no restriction of their daily activities. However, vigorous exercise and contact sports are generally not recommended as these activities theoretically increase stress to the prostheses, wear rate of articulating surface and may give rise to early mechanical failure of implants. Similar to conventional joint arthroplasty for osteoarthritis, bacteria inoculation to the location of artificial joints can happen during episodes of bacteremia secondary to febrile illness, dental extraction or invasive procedures. Antibiotic prophylaxis is advisable to prevent serious complications of infected prostheses. The earlier patients receive aggressive surgical debridement and antibiotic treatment for prosthetic infection of acute onset, the higher the chance the infection can be eradicated. Delay in treatment may result in osteomyelitis that unavoidably requires removal of the prosthesis, multiple operations or even amputation.<sup>16,17</sup>

The cost of prostheses ranges from HK\$65,000 to HK\$180,000, depending on the complexity of the prostheses. In Hong Kong, patients need to purchase the prostheses by their own and the cost is not funded by the Government. With the help from medical social workers, patients with financial difficulties can apply sponsorship from charity organisations.

In conclusion, surgical management of paediatric bone sarcoma is challenging. Effective chemotherapy and advances in surgical techniques make limb sparing surgery possible and offers adequate disease control comparable to the results obtained by amputations. With advances in design, material technology and surgical techniques, modern implanted tumour prostheses are more durable, less short term complications and can achieve good limbs function.

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