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## RESEARCH ARTICLE

# Survival and failure modes of the Compress<sup>®</sup> spindle and expandable distal femur endoprosthesis among pediatric patients: A multi-institutional study

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## Abstract

**Background:** Expandable endoprostheses can be used to equalize limb length for pediatric patients requiring reconstruction following large bony oncologic resections. Outcomes of the Compress<sup>®</sup> Compliant Pre-Stress (CPS) spindle paired with an Orthopedic Salvage System expandable distal femur endoprosthesis have not been reported.

**Methods:** We conducted a multi-institutional retrospective study of pediatric patients with distal femoral bone sarcomas reconstructed with the above endoprostheses. Statistical analysis utilized Kaplan–Meier survival technique and competing risk analysis.

**Results:** Thirty-six patients were included from five institutions. Spindle survivorship was 86.3% (95% confidence interval [CI], 67.7–93.5) at 10 years. Two patients had a failure of osseointegration (5.7%), both within 12 months. Twenty-two (59%) patients had 70 lengthening procedures, with mean expansions of 3.2 cm (range: 1–9) over 3.4 surgeries. The expandable mechanism failed in eight patients with a cumulative incidence of 16.1% (95% CI, 5.6–31.5) at 5 years. Twenty-nine patients sustained International Society of Limb Salvage failures requiring 63 unplanned surgeries. Periprosthetic joint infection occurred in six patients (16.7%). Limb preservation rate was 91% at 10 years.

**Conclusions:** There is a high rate of osseointegration of the Compress<sup>®</sup> spindle among pediatric patients when coupled with an expandable implant. However, there is a high rate of expansion mechanism failure and prosthetic joint infections requiring revision surgery.

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**Level of evidence:** Level IV, therapeutic study.

**KEYWORDS**

expandable endoprosthesis, limb-salvage surgery, pediatric sarcoma

## 1 | INTRODUCTION

Managing limb length discrepancy is challenging following lower extremity tumor resection and endoprosthetic reconstruction in the pediatric population. The majority of primary bone sarcomas in the pediatric patient occur in the distal femur, thus affecting the most active growth plate in the lower extremity. Reconstruction with an expandable endoprosthesis enables limb length equalization, achieved by sequential lengthening of the expandable portion.<sup>1-5</sup>

The durability of an endoprosthesis and fixation of the implant to bone is also a challenge in the pediatric population. Due to improved chemotherapy regimens for osteosarcoma and Ewing sarcoma, patients with localized disease in the lower extremity can expect a 75%-85% 5-year and 60%-65% 10-year survival following chemotherapy and wide resection.<sup>6,7</sup> Endoprostheses secured to the bone with compressive osseointegration technology have a favorable long-term aseptic failure profile,<sup>8-11</sup> however, there is limited evidence regarding implant survivorship and functional outcomes when osseointegration is coupled with an expandable component in the pediatric population.<sup>12</sup> Implant survivorship, mean limb lengthening, cause of revisions, evaluation of expansion mechanism failures, and infection rates for the distal femoral Compress<sup>®</sup> Compliant Pre-Stress (CPS) spindle paired with the Orthopedic Salvage System (OSS) Expandable Device (Zimmer-Biomet) at the distal femur have not been previously reported.

The aims of this study were to report the midterm endoprosthetic-related outcomes among pediatric patients with a primary bone sarcoma at the distal femur reconstructed with a Compress<sup>®</sup>/OSS distal femur expandable endoprosthesis and to determine the survivorship of the CPS spindle, modes of failure using the International Society of Limb Salvage (ISOLS) classification and the rate of and risks associated with prosthetic joint infections.

## 2 | METHODS

### 2.1 | Study design and setting

We performed a multi-institutional retrospective cohort study of skeletally immature individuals with primary bone sarcomas located in the distal femur who underwent distal femoral resection and reconstruction with a CPS spindle and expandable OSS distal femur endoprosthesis with a rotating platform, hinged total knee replacement. Institutional Review Board approval was obtained at each individual study center. Data use agreements allowed the transfer of deidentified data, collected using a standardized data form, to one

centralized study center for data compilation and analysis. Ten orthopedic oncologists contributed data from five West Coast tertiary referral centers in the United States.

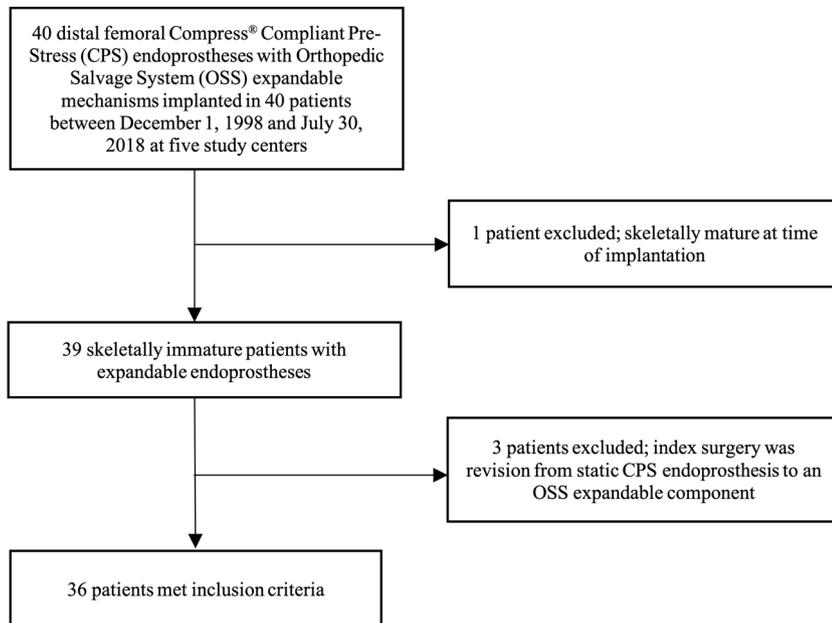
Surgeons at the participating study institutions preferentially used the Zimmer-Biomet expandable OSS implant paired with a CPS spindle to achieve bone fixation via compressive osseointegration for all primary bone sarcoma reconstructions in skeletally immature patients who were not undergoing radiation treatment.

### 2.2 | Participants

Patients met the inclusion criteria if they had a primary bone sarcoma at the distal femur treated with limb salvage using a CPS spindle with an OSS expandable endoprosthesis (Figure 1). Participating surgeons elected to use a cemented stem in cases where adjuvant radiation was used or planned, and those patients were not included in this study. Additionally, patients must have been skeletally immature at the time of index surgery and had a minimum follow-up of 2 years or reached a primary endpoint (death due to disease or removal of the CPS spindle) before 2 years. Patients were excluded if an expandable CPS endoprosthesis was used for revision of a prior oncologic component or to achieve limb length equality in a skeletally mature individual. Clinical data were obtained from the electronic medical records and included age at index surgery, gender, diagnosis, receipt of adjuvant treatments, resection length, type of expandable mechanism, date of surgery, time to revision, time to follow-up, spindle survival, mode of failure using the ISOLS classification system<sup>13,14</sup> number of operations (making note of whether the operation was planned or unplanned), length of implant expansion, residual limb length discrepancy, limb preservation status, and patient survival. Deidentified patient data was compiled at each participating institution and analyzed by researchers at the centralized study center.

### 2.3 | Description of treatment

During the study period, three different OSS expandable implant mechanisms were used. Before 2004, custom-expandable implants were used utilizing the first-generation C-clamp (CC). Expansion required a full exposure of the implant, removal of a modular clip or collar, and replacement with a longer clip or collars secured in place by cables, with or without cement. In 2004, both the CC device and the second-generation design, which utilized a worm-drive accessible via an intercondylar (IC) screw, became Food and Drug Administration

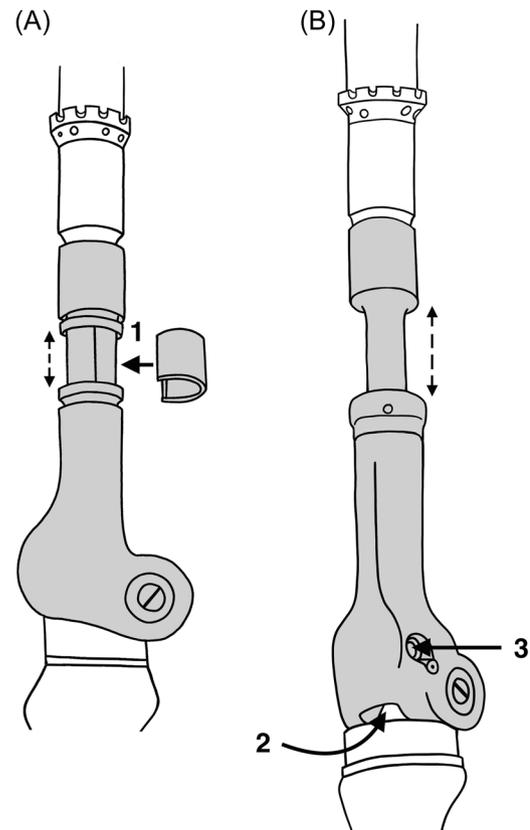


**FIGURE 1** Inclusion and exclusion criteria

approved for use with reduced-sized distal femur components. The second-generation expansion device required an open arthrotomy to access the expansion mechanism. Eight full counterclockwise rotations resulted in 1 cm of expansion. The third-generation implant, which has been available since 2014, has the same internal expansion mechanism as the second-generation design, but the access screw is located on the lateral condyle (LC), resulting in a minimally invasive surgery. One hundred and forty-two full counterclockwise rotations result in 1 cm of expansion (Figure 2A,B). Intraoperative use of C-arm intensifier imaging was utilized during lengthening procedures to confirm correct instrument alignment and final expansion length. To reduce neurovascular compromise, limb-lengthening surgeries did not exceed the recommended 1–2 cm expansion length per procedure,<sup>3,15,16</sup> and serial lengthening procedures were generally performed no more frequently than once every 6 months.

## 2.4 | Description of study population

Thirty-six patients met the inclusion criteria from the five study institutions (Figure 1). There were 24 male patients and 12 female patients with an average age of 10.7 years (range: 5–15) at the time of distal femur resection and reconstruction with an expandable CPS device. All patients were treated for primary oncologic diagnoses of osteosarcoma (34) or Ewing sarcoma (2). Thirty-five patients (97%) received neoadjuvant and/or adjuvant chemotherapy and no patients received radiation. Distal femur resection averaged 19.4 cm (range: 15–30) and a mean body mass index at index surgery was 21.9 (range: 15–35). The mean follow-up was 87.4 (range: 10–246) months (Table 1). One patient (3%) received a first-generation CC implant. Twenty-one patients (58%) received a second-generation implant with the IC access screw, and 14 (39%) received a third-generation implant with the LC access screw. Patient outcomes are outlined below (Table 2).



**FIGURE 2** (A, B) Illustration of the expansion mechanisms of the distal femoral OSS expandable device paired with the Compress® CPS spindle. The area of expansion is shown between dashed double-headed arrows. (A) Lateral view of first-generation C-clamp OSS utilizing clips of incrementally larger size for expansion (arrow 1). (B) Frontolateral view of second- and third-generation OSS with an internal expansion mechanism with either an intercondylar access screw (arrow 2) or lateral condylar access screw (arrow 3), respectively. CPS, Compliant Pre-Stress; OSS, Orthopedic Salvage System.

**TABLE 1** Patient demographics (n = 36)

Characteristic	Number
Age at index surgery (years)	10.7 (range: 5–15; SD: 2.2)
Female	10.4 (range: 8–13; SD: 1.4)
Male	10.8 (range: 5–15; SD: 2.5)
Sex	
Female	12
Male	24
BMI (kg/m <sup>2</sup> )	21.9 (range: 14.5–34.5; SD: 6.3)
Resection length (cm)	19.4 (range: 14.5–30; SD: 3.9)
Surgical indication	
Primary oncologic reconstruction	36
Tumor diagnosis	
Osteosarcoma	34
Ewing sarcoma	2
Chemotherapy	
Neoadjuvant and/or adjuvant (both)	35 (33)
Type of expansion mechanism used	
Clip (%)	1 (3%)
Intercondylar screw (%)	21 (58%)
Lateral condylar screw (%)	14 (39%)
Follow-up (months)	87.4 (range: 10–246; SD: 71.3)

Abbreviation: BMI, body mass index.

## 2.5 | Statistical analysis

Statistical analysis of spindle survival, expansion mechanism survival, and failure-free survival was determined using the Kaplan–Meier technique<sup>17</sup> in Stata<sup>®</sup> (STATA Corp LP). Competing risk analysis and cumulative incidence estimation<sup>18</sup> for spindle failure, expansion mechanism failure, and first ISOLS failure was performed in R (version 4.0.0; R Core Team) using the “cmprsk” package.<sup>9,19,20</sup> This study was designed and reported using the STROBE Criteria.<sup>21</sup>

## 3 | RESULTS

### 3.1 | Survivorship of endoprosthesis spindle

Spindle survivorship was 91.3% (95% CI, 75.4–97.1) at 2 years and 86.3% (95% CI, 67.7–93.5) at 5 and 10 years (Figure 3A); four patients underwent removal of the spindle for either aseptic loosening (1) or at the time of an above knee amputation (3). Two patients (5.7%; 95% CI, 1.46–21.1) experienced spindle failure due to incomplete osseointegration within 12 months of index surgery. One spindle rotated after early weight-bearing within 3 weeks of

implantation; however, by the time of surgical revision, osseointegration at the bone–implant interface was achieved, the spindle was retained, and the rotational deformity was corrected by rotating the modular implant components. The second patient received an undersized centering sleeve that caused angulation of the endoprosthesis resulting in a femoral fracture that was corrected by removal of the original spindle and revision to a new spindle apparatus proximal to the fracture.

### 3.2 | Number of expansions and total length gained

Twenty-two patients (61%) collectively underwent 70 lengthening procedures, resulting in a mean overall expansion of 3.2 cm (range: 1–9 cm) over an average of 3.4 surgeries (range: 1–9). Mean lengthening per procedure was 1.1 cm (range: 0.5–2.25). Fourteen patients (39%) did not undergo expansion of their OSS device including nine who died due to progressive oncologic disease, two who had early above knee amputations, and three who had not yet developed limb-length inequality requiring an expansion procedure.

### 3.3 | Rate of expansion device failure

The expandable component failed in eight patients (22%) at a mean time of 47.3 months (range: 22–91). Expansion mechanism survivorship was 85.1% (95% CI, 67.7–93.5) at 2 years, 66.2% (95% CI, 43.3–81.6) at 5 years, and 43.3% (95% CI, 22.9–66.9) at 10 years (Figure 3B). Cumulative incidence estimation of failure of the expansion mechanism using death or removal of the expandable component (above knee amputation or revision to non-expandable component) as competing events showed an increasing failure rate over time: 2.8% (95% CI, 0.2–12.6) at 2 years, 16.1% (95% CI, 5.6–31.5) at 5 years, and 29.8% (95% CI, 13.3–48.4) at 10 years (Table 3 and Figure 4B). Failures included rotation of the expandable component (3), fragmentation and dislodging of mechanical components (2), and failure resulting in loss of expansion (3). These failures occurred in all three expansion mechanism types (1 CC, 4 IC, and 3 LC). Failure of the expansion mechanism did not affect the integrity of osseointegration of the spindle, and revision did not require removal of the osseointegrated spindle. Five patients were revised with a new expandable OSS endoprosthesis; two patients were revised to a static endoprosthesis. One patient experienced two expansion failures and was ultimately revised to a static endoprosthesis with no additional failures or unplanned reoperations. Figure 5A–C shows the radiographs of a patient who experienced failure, resulting in the loss of expansion. Figure 5A shows expansion after three lengthening procedures. However, shortly thereafter the patient noted a worsening leg length discrepancy and radiographs demonstrated loss of expansion (Figure 5B). The patient was revised to a new, longer expandable component (Figure 5C).

Eight patients were revised to static implants after an average of 71.5 months (range: 19–190); three due to failure of the expansion

**TABLE 2** Patient outcomes (*n* = 36)

Characteristic	Number
Spindle failure	2 (6%)
Mean time to spindle failure	8.1 months (4–12)
Reason for failure	Incomplete osseointegration
Expandable mechanism failure	6 (18%)
Mean time to expandable mechanism failure	47.2 months (22–91)
Revision to nonexpandable components	2
ISOLS unplanned surgical reoperation	63 in 29 patients (81%)
Time to first revision	16.9 months (1–61)
ISOLS classifications of all revisions	Type 1A (20), Type 1B (9), Type 2A (2), Type 3A (17) Type 3B (5), Type 4A (2), Type 4B (4), Type 5B (4)
Infection	6 (18%)
Mean time to infection	44.6 months (2–127)
Type of expansion mechanism	6 intercondylar screw access point
Above knee amputation	3 (9%)
Mean time to amputation	28.2 months (12–59)
Indication for amputation	1 infection, 1 severe arthrofibrosis, 1 local recurrence
Periprosthetic fracture	6 (18%)
Mean time to fracture	68.2 months (9–178)
Death due to progressive disease	10 (28%)
Mean time to death	28.2 months (10–61)

Abbreviation: ISOLS, International Society of Limb Salvage.

mechanism (listed above) and five for elective revisions to a static component at skeletal maturity. The elective revisions generally incorporated one final centimeter of lengthening with the exchange.

### 3.4 | Prevalence of ISOLS failure modes

Twenty-eight patients (78%) sustained one or more ISOLS failures requiring 63 surgical reoperations with a mean time to first failure of 16.9 months (range: 0.7–61). The 63 ISOLS failures included 29 soft-tissue failures (46%, Type 1), two aseptic loosening events (3%, Type 2), 22 structural failures (35%, Type 3), six prosthetic joint infections (10%, Type 4), and four local tumor progressions (6%, Type 5) (Figure 6). Patients most frequently encountered soft-tissue functional failures, Type 1A (*n* = 20, 56%), which consisted exclusively of arthrofibrosis and were treated with manipulation under anesthesia, lysis of adhesions, chemodenervation, and/or revision

of the implant. Patients who experienced a failure required on average 2.29 unplanned procedures.

### 3.5 | Prosthetic joint infection rate and risk factors

Prosthetic joint infection affected six patients (16.7%) with a mean time to infection of 44.6 months (range: 2–127). Five patients underwent secondary surgical procedures before development of infection, including three with multiple expansions of their OSS device (2, 4, and 5 surgeries) and two with revisions for arthrofibrosis. The sixth patient had central venous line sepsis in the week before index surgery and developed complications of PJI within 2 months; this was the only patient who experienced PJI during adjuvant chemotherapy. Patients with PJI underwent a two-stage exchange with retention of the osseointegrated spindle and removal of the modular components, placement of an antibiotic-impregnated cement spacer, and revision to a new endoprosthesis after resolution of the infection. PJI occurred exclusively in patients with an IC expansion mechanism (*p* = 0.048); additional risk factors identified include multiple surgeries for expansion or arthrofibrosis. One PJI progressed and the patient elected to have an above knee amputation, one patient died due to progressive oncologic disease after the first stage of implant exchange, and four patients had successful infection clearance.

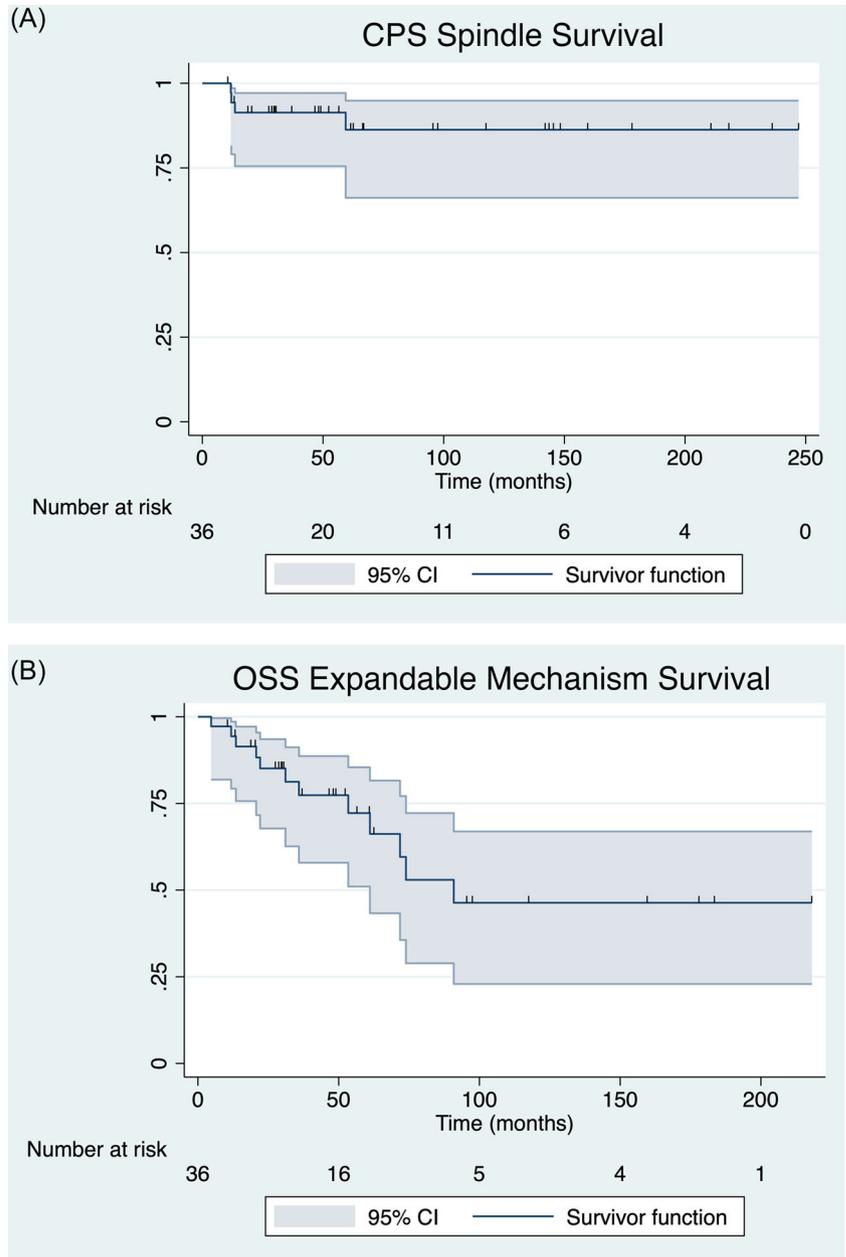
The overall limb-salvage rate in this cohort was 91% at 5 and 10 years. Three patients underwent an above-the-knee amputation (8%), one each for PJI, arthrofibrosis, or local recurrence, at a mean time of 28.2 months (range: 12–59); two occurred within 24 months of index surgery. Ten patients with intact expandable CPS devices died due to progressive oncologic disease at a mean time to death of 35.7 months (range: 8–97). The overall patient survival rate was 72% at 5 years.

## 4 | DISCUSSION

Despite the relatively small size of this cohort, to our knowledge, this is the largest cohort to examine outcomes of a single manufacturer's expandable device used in a single anatomic location (distal femur) among pediatric patients with primary bone sarcomas. Additionally, it is the only study to specifically report on the implant survivorship when an osseointegrated spindle is coupled with a distal femur expandable endoprosthesis. This multicenter study followed 36 skeletally immature patients for a minimum of 2 years, or until death or removal of the Compress<sup>®</sup> spindle, with an average of 87.4 months (range: 10–246). The use of this implant achieved an overall 91% limb-salvage rate at 10 years. However, as has been reported in other series,<sup>4,20,22</sup> there was a high complication rate requiring multiple revision procedures; 78% of our cohort experienced a failure requiring an average of 2.29 additional surgeries.

Several manufacturers offer expandable implants with minimally invasive to noninvasive lengthening mechanisms,<sup>1,3,7,15,23</sup> and there

**FIGURE 3** (A, B) Kaplan–Meier survival curves showing (A) spindle survivorship of 91.3% (95% CI, 75.4–97.1) at 2 years and 86.3% (95% CI, 66.1–94.9%) at 5 years, and (B) expandable mechanism survivorship of 85.1% (95% CI, 67.7–93.5) at 2 years, 66.2% (95% CI, 43.3–81.6) at 5 years and 43.3% (95% CI, 22.9–66.9) at 10 years. CI, confidence interval; CPS, Compliant Pre-Stress; OSS, Orthopedic Salvage System.

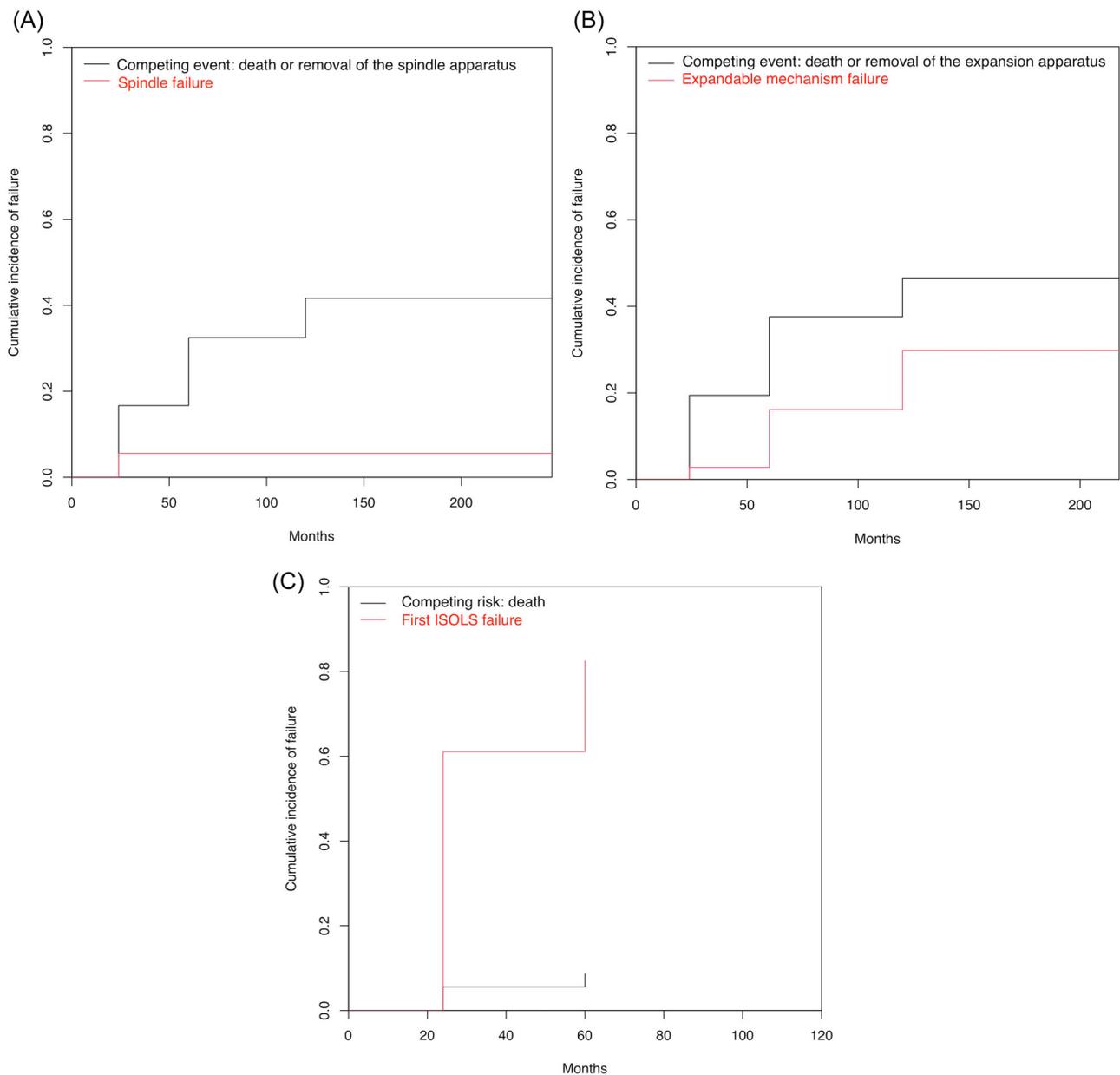


**TABLE 3** Competing risk analysis: The cumulative incidence of events shown at 2, 5, and 10 years of follow-up ( $n = 36$ )

Event type	Patients ( $n = 36$ )					
	Cumulative number of events			Cumulative incidence of events (95% CI)		
	2 years	5 years	10 years	2-year follow-up	5-year follow-up	10-year follow-up
Spindle failure	2	2	2	5.6% (1.0–16.5)	5.6% (1.0–16.5)	5.6% (1.0–16.5)
Expandable failure	2	5	8	2.8% (0.2–12.6)	16.1% (5.6–31.5)	29.8% (13.3–48.4)
First ISOLS failure	22	28	29	61.1% (42.8–75.1)	82.5% (60.7–92.9)	
Death	4	8	10	5.6% (1.0–16.5)	16.9% (5.8–33.0)	16.9% (5.8–33.0)

Note: Competing events for spindle failure included death or removal of the spindle apparatus. Competing events for expandable component failure included death, removal of the spindle, or conversion of the expansion mechanism to a nonexpandable component. Competing events for ISOLS failure included death or removal of the spindle.

Abbreviations: CI, confidence interval; ISOLS, International Society of Limb Salvage.



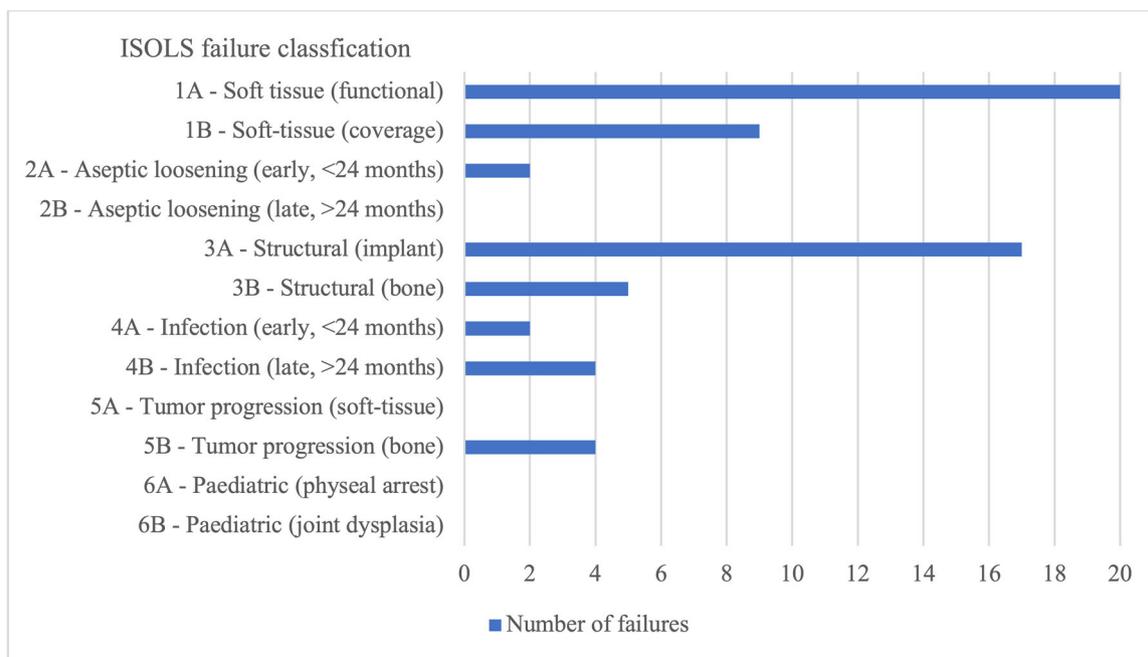
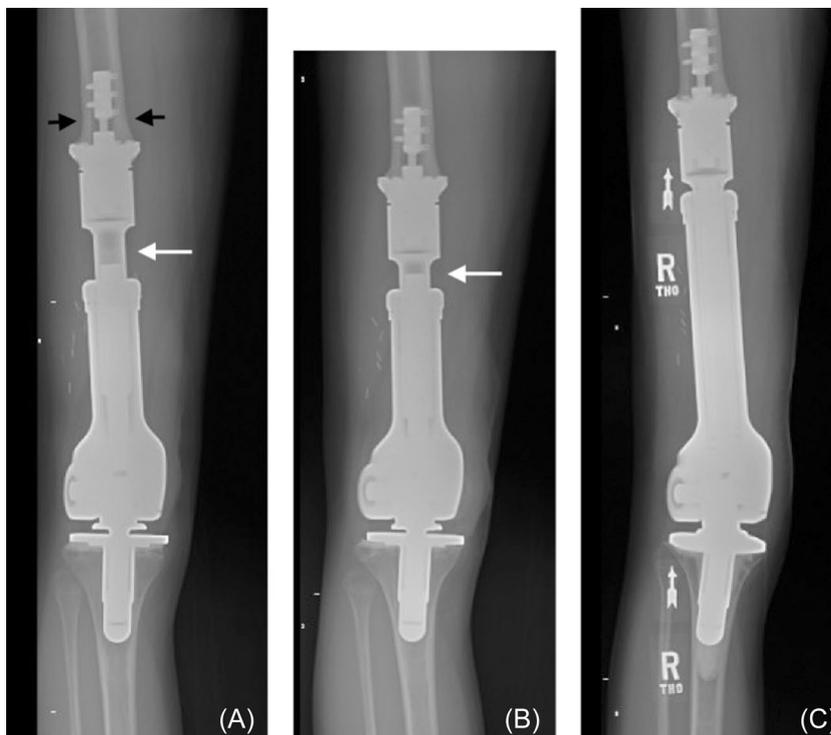
**FIGURE 4** (A–C) Competing risk analysis with the cumulative incidence of events shown at 2, 5, and 10 years follow-up ( $n = 36$ ) for (A) spindle failure versus death or removal of the spindle, (B) expandable component failure versus death or removal of the expansion mechanism, and (C) and first ISOLS failure versus death. ISOLS, International Society of Limb Salvage.

currently is no consensus or guidance on choosing an expandable endoprosthesis.<sup>3</sup> Proponents of the compressive osseointegration for bone anchorage of large endoprosthesis cite a low aseptic failure rate and a low long-term failure of the osseointegrated spindle.<sup>10,24,25</sup> Similar to previously published studies evaluating the Compress spindle in adult patients, we found a similar survival of the spindle when coupled to an expandable OSS endoprosthesis<sup>8,11,24–27</sup> with only two spindle failures (5.5%) in this cohort. It should be noted that both were failures of osseointegration within 12 months of surgery and we found no late failures at the bone–implant interface. This pattern of stability once osseointegration has been achieved has been previously reported.<sup>8,25</sup> The rate of aseptic loosening at 10-year

follow-up in our series (5.5%) was lower than traditional cemented and uncemented stems (13.2%–52%) when paired with expandable components.<sup>4,22,28</sup> A benefit of durable osseointegration is the ability to retain an osseointegration spindle when revising a failed expandable mechanism or during two-stage exchange for the treatment of PJI. Despite spindle retention during two-stage implant exchange for infection, we had a 67% rate of infection eradication, which is comparable with other series.<sup>29</sup> Retention of the spindle at the time of revision preserves bone stock and allows these patients to be immediately weight-bearing postrevision.

Serial lengthening provides a meaningful approximation of limb equality for skeletally immature patients and preserves gait and

**FIGURE 5** (A–C) Postoperative radiographs of a pediatric patient with an expandable CPS endoprosthesis (lateral condyle screw expansion mechanism). (A) Expandable 18 cm CPS endoprosthesis lengthened to 21 cm after three procedures. There is evidence of osseointegration at the bone-spindle interface and development of an “elephant’s foot” bone hypertrophy from the compressive forces (black arrows). (A, B) Radiographic evidence of limb length loss due to a failed expansion mechanism (white arrows). (C) The patient was revised to a new 23 cm expandable component and returned to functional status. CPS, Compliant Pre-Stress.



**FIGURE 6** Classification of the 63 ISOLS failures affecting 29 of 36 (81%) pediatric patients after limb preservation surgery with an expandable Compress<sup>®</sup> endoprosthesis. Nineteen patients sustained multiple ISOLS failures, including seven patients with two failures, eight with three failures, and four with four failures. Failures are grouped by the ISOLS classification system. ISOLS, International Society of Limb Salvage.

function as it is adjusted to match the continued growth of the contralateral unaffected lower extremity. The percentage of our patients who underwent an expansion surgery (61.1%) is comparable to previously published rates (51.3%–96%),<sup>4,20,28</sup> with a similar number of expansions (3.2 vs. 4.0–4.2) and expansion lengths (3.4 vs.

3.95–4.65 cm).<sup>22,28,30,31</sup> Given that nine patients (25%) did not undergo expansion due to progressive metastatic disease, one consideration would be initially implanting a nongrowing prosthesis. The expandable portion of the prosthesis could be placed at the time of first lengthening, which would not require revision of the CPS

spindle. Residual limb length discrepancy was not reportable here due to incomplete records, lack of standardization in data collection (e.g., unilateral radiographic plain films, bilateral scanograms, clinical assessment), and the fact that several patients in the cohort had not yet reached skeletal maturity.

The integrity of the OSS expandable mechanism, when paired with an osseointegrated CPS spindle, has not yet been reported. In the present series, eight patients (22%) experienced failure of the expansion apparatus at a mean time to failure of 47.3 months (range: 22–91), and our cohort continued to accrue these failures over time. Seven of the eight patients required revision surgery. Five patients underwent elective revision to a nonexpandable implant after reaching skeletal maturity. The expandable component may therefore be seen as a temporary bridge to a static adult endoprosthesis, with exchange occurring after failure or in an elective fashion to avoid a late failure of the expansion mechanism after the desired limb length is achieved.

Patients should be counseled on the likelihood that they will require one or more unplanned surgical reoperations (Table 3) in addition to the expected surgical procedures for serial limb lengthening. Similar to the findings of a recent multi-institutional study in Europe of nonosseointegrated distal femoral expandable endoprostheses from multiple manufacturers<sup>22</sup> and a single-institution study of 124 patients with expandable endoprostheses at 10-year follow-up,<sup>4</sup> most patients in our cohort (80.6% vs. 63%–85%) experienced ISOLS failures requiring multiple surgical revisions (2.2 vs. 2.2–2.7) in addition to the lengthening procedures. Patients should be advised of the expansion mechanism failure rate of 6.7% (95% CI, 1.7–24.2) at 2 years, 20.6% (95% CI, 8.9–43.4) at 5 years, and 41.7% (95% CI, 22.3–68.5%) at 10 years, and that the risk for failure increases over time. Notably, all three types of expansion mechanisms failed in our series. Five patients in our series opted for an elective revision to a static nonexpandable adult component at skeletal maturity after sufficient limb-length equality was approximated. The rate of infection is high in our series (16.6%) and comparable to other studies using distal femoral expansion mechanisms with invasive lengthening procedures (8.6%–22.9%)<sup>22,28</sup> and the Prophylactic Antibiotics Regimens in Tumor Surgery (PARITY) study (15.0% and 16.7%).<sup>32</sup> Four of the six patients with PJI were successfully treated with two-stage exchange revision with an antibiotic-impregnated spacer and spindle retention, which is a useful therapeutic approach in this population, as it avoids a prolonged period of nonweight bearing and bone loss while maintaining the volume of the soft tissue envelope for the placement of the revision implant. Risk factors for PJI in this cohort included repeated surgical revision and the use of the second-generation design with the IC access screw. There were no infections in the patients with the LC access screw, which has a minimally invasive approach for limb-lengthening surgeries, supporting previous reports showing reduced infection rates when moving from invasive to minimally invasive or noninvasive expansion mechanisms.<sup>4,22</sup> There was a high rate of soft tissue failure in this cohort, 56%, all due to arthrofibrosis. Arthrofibrosis requiring surgical interventions can

increase the risk of infection in this population. Causes of arthrofibrosis should be investigated and patients should be started on aggressive physical therapy following initial reconstruction and after lengthening procedures.

Our study was limited by the retrospective nature and a relatively small sample size due to the rarity of sarcomas in the pediatric population. However, by including patients from five medical institutions, we were able to create a homogenous study population of skeletally immature patients with bone sarcomas of the distal femur who had primary limb-salvage reconstruction using the Zimmer-Biomet CPS spindle and an OSS expandable endoprosthesis. By increasing the number of surgeons contributing cases over a period of 20 years, we reduced bias associated with idiosyncratic surgical techniques or experience, making the data more generalizable. With the establishment of our multi-institutional collaboration, future studies can include prospective data and longer follow-up to determine limb-length discrepancy at skeletal maturity, functional outcomes, and patient-reported outcomes of the CPS spindle when paired with an OSS expandable component.

## 5 | CONCLUSIONS

Expandable endoprostheses provide skeletally immature patients undergoing limb-salvage surgery the opportunity for limb length equalization. However, these patients will require multiple revision surgeries over time. Our study adds new information to the literature highlighting the risk of failure of the expansion mechanism, which increases with time. However, using the Compress spindle leads to a high rate of osseointegration and stability at the bone-implant interface. The modular design of this implant allows for spindle retention, thus sparing bone at the time of revision surgery. The Compress spindle coupled with the Zimmer-Biomet expandable OSS distal femur endoprosthesis results in an overall high limb-salvage rate despite a high infection and revision rate among pediatric patients.

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## CONFLICT OF INTEREST

One of the authors (James B. Hayden), or a member of his immediate family, has or may receive payments or benefits, an amount of USD 10 000–100 000, during the study period from Biomet®. The remaining authors declare no conflict of interest.

## DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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