

■ ONCOLOGY

Outcomes of type BIIIa rotationplasty in children

M. Pruthi,
R. Agarwal,
P. Nayak,
P. Hegde,
A. Gulia,
A. Puri

From Tata Memorial
Hospital, Mumbai,
India

Aims

Total excision of the femur and its reconstruction pose a substantial challenge in children. We present the long-term results of total femoral excision and type BIIIa rotationplasty in children.

Methods

A total of 18 patients who had undergone a type BIIIa rotationplasty over a period of 20 years were included in the study. Their mean age at the time of surgery was 7.1 years (3.3 to 11). Two patients underwent a hip disarticulation in the perioperative period. Five died in the first two years from surgery. The mean follow-up of the remaining 11 patients was 124 months (24 to 244). We evaluated their long-term radiological, functional, and oncological outcomes and measured any limb length discrepancy at final follow-up.

Results

The hip was stable in eight of 11 patients. Radiologically appreciable remodelling of the proximal tibia was seen in all patients. At final follow-up, four patients had reached skeletal maturity, five were adolescents (aged 12 to 17 years), and two were still pre-adolescent (aged < 12 years). The mean shortening in the skeletally mature patients was 2.25 cm (0 to 4). Limb length was measured in four of the five adolescent patients: the mean shortening was 0 cm (-1 to +1). Other than the two patients who underwent a hip disarticulation in the immediate perioperative period, no patient needed a subsequent surgical procedure. None of the patients had a local recurrence. The five-year probability of overall survival was 70% (95% CI 51 to 96). The mean Musculoskeletal Tumor Society score was 23 (22 to 23). The mean Toronto Extremity Salvage Score calculated in nine patients was 91 (82 to 100).

Conclusion

A type BIIIa rotationplasty is a reliable option for reconstruction after total femoral excision in children. Long-term follow-up shows good functional and oncological outcomes, without the need for additional surgical procedures.

Cite this article: *Bone Joint J* 2026;108-B(1):125–131.

Introduction

The femur is the most common bone affected by sarcoma.¹ Occasionally, the entire femur is involved, or there is a skip metastasis proximal or distal to the primary tumour, necessitating total resection of the femur to clear the tumour. In adults, a total femoral resection is usually reconstructed using a mega-prosthesis, which gives an acceptable functional outcome. However, total femoral resections pose a substantial reconstruction challenge in children. A noninvasive expandable total femoral prosthesis is one reconstructive option in the management of

these defects in children.² However, in addition to being expensive, they are fraught with the need for multiple revision procedures, acetabular dysplasia, and may also limit participation in active sports. Winkelmann³ described the tibia into hip (BIIIa) rotationplasty after total femoral excision in childhood, which exploits the biological plasticity of the proximal tibia to remodel and replicate the femoral head. The literature on long-term outcomes of type BIIIa rotationplasty after total femoral resection is limited: it is not commonly resorted to, even in experienced oncology centres.

Correspondence should be sent to A. Puri; email: docpuri@gmail.com

© 2026 The British Editorial Society of Bone & Joint Surgery
doi:10.1302/0301-620X.108B1.
BJJ-2025-0309.R1 \$2.00

Bone Joint J
2026;108-B(1):125–131.

Table I. Characteristics of 18 patients with follow-up details.

No.	Age at surgery, yrs	Sex	Histology	Pathological fracture	Vessel anastomosis	Length of follow-up, mths	Hip instability	LLD, cm	MSTS score	TESS	Current status/ occupation
1	8.7	M	Osteosarcoma	No	No	244	No	2	22	88	Software engineer
2	10.4	M	Ewing's	No	No	204	No	4	22	99	Stockbroker
3	9.4	M	Ewing's	No	No	203	No	0	23	98	Farmer
4	7.7	F	Ewing's	No	Yes	197	Yes	3	22	82	Competitive shooter (para-athlete)
5	10.8	M	Ewing's (post ECRT)	No	No	12	-	-	-	-	Dead
6	3.4	M	Anaplastic RMS	Yes	No	8	-	-	-	-	Dead
7	5.6	M	Ewing's	No	No	9	-	-	-	-	Dead
8	4.7	M	Ewing's	No	No	119	No	-1	22	86	Schooling
9	4.2	M	Ewing's	No	No	96	NE	NE	23	83	Schooling
10	11.0	M	Osteosarcoma	No	Yes	Disarticulated	-	-	-	-	N/A
11	8.2	M	Ewing's	Yes	No	95	No	0	23	85	Schooling
12	5.6	M	Osteosarcoma	Yes	No	Disarticulated	-	-	-	-	N/A
13	3.8	M	Ewing's	Yes	No	6	-	-	-	-	Dead
14	7.2	F	Ewing's	Yes	No	83	Yes	1	23	95	Competitive swimmer (para-athlete)
15	4.8	F	Ewing's	No	No	57	No	NE	23	NE	Schooling
16	10.7	F	Ewing's	Yes	Yes	50	No	0	23	100	Schooling
17	9.1	M	Ewing's	No	No	25	-	-	-	-	Dead
18	3.3	M	Ewing's	Yes	No	24	NE	NE	23	NE	Schooling

ECRT, extracorporeal radiotherapy; LLD, limb length discrepancy; MSTS, Musculoskeletal Tumor Society; N/A, not applicable; NE, not evaluated; NED, no evidence of disease; RMS, rhabdomyosarcoma; TESS, Toronto Extremity Salvage Score.

We aimed to evaluate the long-term radiological, functional, and oncological outcomes after total femoral resection treated with type BIIa rotationplasty in children aged younger than 12 years.

Methods

Over a period of 18 years (January 2004 to March 2022), 18 patients with sarcoma of the femur aged younger than 12 years were treated surgically with a Winkelman type BIIa rotationplasty at our hospital (Table I). These patients were identified by a retrospective review of a prospectively maintained database. Written informed consent was obtained from the patients for their anonymized information to be published, and this being a retrospective audit, ethical approval was not required.

The medical records, radiographs, and oncological and functional outcomes at follow-up of these cases were reviewed. A total of 14 patients had a Ewing's sarcoma, three had an osteosarcoma, and one a rhabdomyosarcoma. All patients received chemotherapy as per existing hospital protocols.

A detailed review of the surgical technique is published elsewhere.³ A brief summary of the surgical procedure is presented here.

Circumferential oblong incisions along the hip joint and knee joint are connected by a vertical incision along the thigh. The proximal dissection is similar to a hip disarticulation, while preserving as much capsule and muscle around the hip as is oncologically safe. Iliopsoas, sartorius, rectus femoris, and the abductors are tagged before cutting. The sciatic nerve and femoral neurovascular bundle are then dissected along their

length in the thigh (if there is a need to resect the vascular bundle for oncological margin, it is cut at the level of the hip and knee and re-anastomosed). The distal part of the dissection is similar to that for a knee disarticulation and preserves as much of the knee joint capsule and cruciates on the tibia as possible. The muscular heads of gastrocnemius are preserved and released at their tendinous origin. The specimen is then excised, leaving the tibia attached to the proximal body only by the sciatic nerve and femoral vascular bundle (Figure 1). The leg is rotated 180° externally and the limb is shortened, to position the lateral tibial condyle in the acetabulum. The neurovascular structures are coiled and placed anteriorly. The capsule of the hip joint is sutured to the capsule of the knee joint using interrupted nonabsorbable braided sutures. The tendon of the biceps femoris is used to augment the anterior capsule and the cruciate ligaments are sutured to the ligamentum teres. The gastrocnemius muscle ends are sutured to the iliopsoas and rectus femoris muscles. The hamstrings are sutured to the anterior tibial muscles and the abductors to the insertion of the pes anserinus. The patient is given a pelvic harness to help support the weight of the limb during standing for the first three months (Figure 2). Subsequently, final prosthesis fitting and gait training is undertaken.

As per our institutional protocol, patients were followed up every six months for the first five years and annually thereafter, with radiographs of the chest and of the rotationplasty limb including the hip.

At follow-up, patients were examined clinically to assess the stability of their new 'neo' hip and ranges of flexion and abduction. Limb length discrepancy was determined by comparing the level of the rotationplasty limb heel pad with the medial

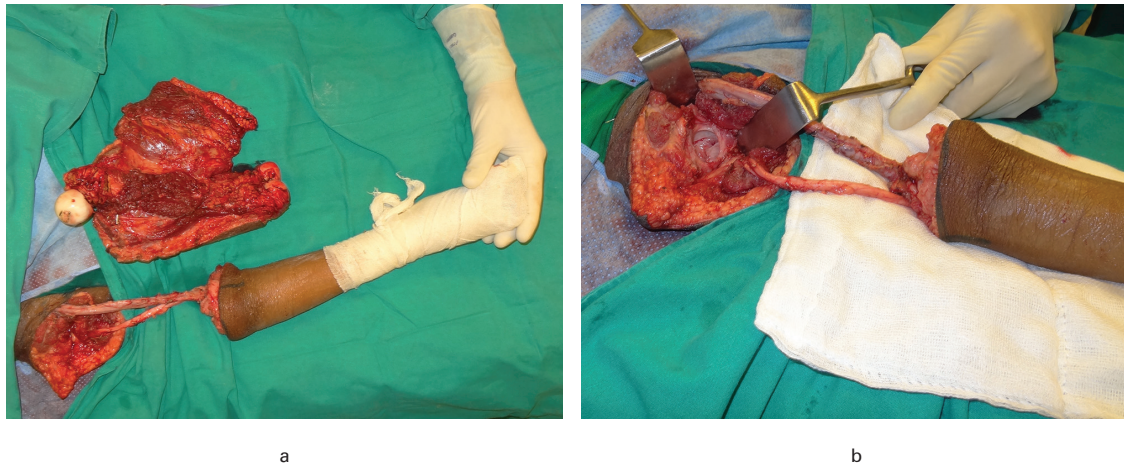


Fig. 1

a) Intraoperative photograph with excised specimen and distal limb continuity maintained with only neurovascular bundle. b) Acetabulum with intact labrum prior to positioning the proximal tibia.

knee joint line of the contralateral femur on a squared pelvis. Radiological assessment was done with radiographs in two planes to assess the remodelling of proximal tibia.

Survival curves were calculated according to the Kaplan-Meier method using SPSS v. 26.0 (IBM, USA).

Functional outcomes were evaluated using the Musculoskeletal Tumor Society (MSTS)⁴ score and the Toronto Extremity Salvage Score (TESS).⁵ The MSTS, being a physician-evaluated score, was completed by the medical team. Translated and validated questionnaires for TESS in the regional language were completed by the patients.

The MSTS scoring system evaluates the functional outcomes of patients treated for musculoskeletal tumours, focusing on six aspects for the lower limb: pain, function, emotional acceptance, use of walking aids, walking ability, and gait. Each aspect is scored from 0 to 5, with 5 indicating the best outcome.⁴ The total score, with a maximum of 30, is calculated by summing the scores of all six components. TESS is a self-reported assessment tool used to measure physical disability in individuals aged 12 to 85 years who have undergone limb salvage surgery for a sarcoma of the lower limb. It includes 30 questions that evaluate challenges in various daily activities such as dressing, grooming, mobility, work, sports, and leisure. Each question is scored on a scale of 0 to 5, where 5 represents 'no difficulty' and 0 indicates that the activity is 'impossible'.⁵ The total raw score is converted to a scale of 0 to 100, with higher scores reflecting better physical function.

Results

There were 14 male and four female patients. The mean age of patients at the time of surgery was 7.1 years (3.3 to 11). A pathological fracture was present in seven of the 18 patients. All but one of the patients were metastasis-free at presentation. All except one patient underwent rotationplasty as a primary procedure (one patient who had failed reimplantation of sterilized tumour bone with persistent infection underwent rotationplasty at eight months after their index procedure). Vessel anastomosis



Fig. 2

a) Front view of type BIIIA rotationplasty patient using pelvic harness. b) Side-on view of type BIIIA rotationplasty patient using pelvic harness.

was carried out in three of the 18 cases (16.7%) while vascular continuity was maintained in the rest. Of the 18 patients, two (11.1%) underwent a hip disarticulation in the immediate perioperative period due to vascular insufficiency (one of these had undergone a vascular anastomosis at index surgery). Five patients (27.8%) died early (three of metastatic disease, one



Fig. 3

a) Pre-chemotherapy anteroposterior radiograph of a seven-year-old girl with Ewing's sarcoma of femur with pathological fracture in proximal diaphysis. b) Post-chemotherapy anteroposterior radiograph showing fracture healing. c) Seven years' follow-up anteroposterior radiograph with remodelling of proximal tibia.

due to complications of chemotherapy, and one from an unrelated cause) after a mean duration of 11.8 months (6 to 24). The remaining 11 patients were available for follow-up: mean follow-up was 124 months (24 to 244). At final follow-up, four patients had reached skeletal maturity, five were adolescents (aged 12 to 17 years), and two were still pre-adolescent children (aged < 12 years).

Nine patients were available for clinical examination at final follow-up. The hip was examined clinically to assess range of motion and stability. Mean active flexion was 85° (30° to 120°) and mean active abduction 60° (50° to 70°). We defined clinical hip stability of the incongruous 'neo' hip as the ability of the patient to control and hold the hip in active flexion and abduction. The hip was stable in seven (77.8%) and unstable in two (22.2%) of the nine patients. Nine of 11 patients (81.8%) were participating in recreational sports. Hip stability did not correlate with participation in sports or activity levels.

Although no objective criteria could be established for radiological remodelling, on the anteroposterior radiographs a beaking of the lateral tibial condyle was seen, and on the lateral radiographs a rounding of the proximal tibial articular surface was seen in all cases (Figure 3).

Other than the two patients who underwent a hip disarticulation in the immediate perioperative period, no patient needed a subsequent surgical procedure.

The mean shortening in the skeletally mature patients was 2.25 cm (0 to 4). The limb length was measured in four of the five adolescent patients: the mean shortening was 0 cm (-1 to +1).

None of the patients had a local recurrence. The five-year probability of overall survival was 70% (95% CI 51 to 96) (Figure 4).

MSTS was calculated for all 11 of the patients with a mean score of 23 (22 to 23). TESS was calculated in nine of the 11 patients (two patients aged below 12 years were excluded). Their mean score was 91 (82 to 100).

Discussion

Malignant bone tumours have to be resected with adequate margins which occasionally requires excision of the entire bone. In children, if the tumour involves the femur, this can be achieved with a hip disarticulation, resection, and reconstruction with an expandable endoprosthesis or with a type BIIIa rotationplasty. There is limited literature on the type BIIIa rotationplasty, with no published data on its long-term outcome.

Ablative surgery in the form of a hip disarticulation has a high morbidity and results in poor function.⁶ A total femoral endoprosthesis gives reasonable function after a total resection of the femur in adults, but reconstruction in children is challenging. An expandable endoprosthetic total femoral arthroplasty, besides being expensive, has a high incidence of complications and invariably needs revision. In Medellin et al's² series of expandable total femoral prostheses, the mean overall implant survival times for minimally invasive and noninvasive prostheses were 87 months (4 to 312) and 55 months (4 to 133), respectively. The rates of revision-free implant survival were 69% and 61% at five years, and 52% and 61% at ten years, respectively. They observed early failures due to tumour progression or infection,

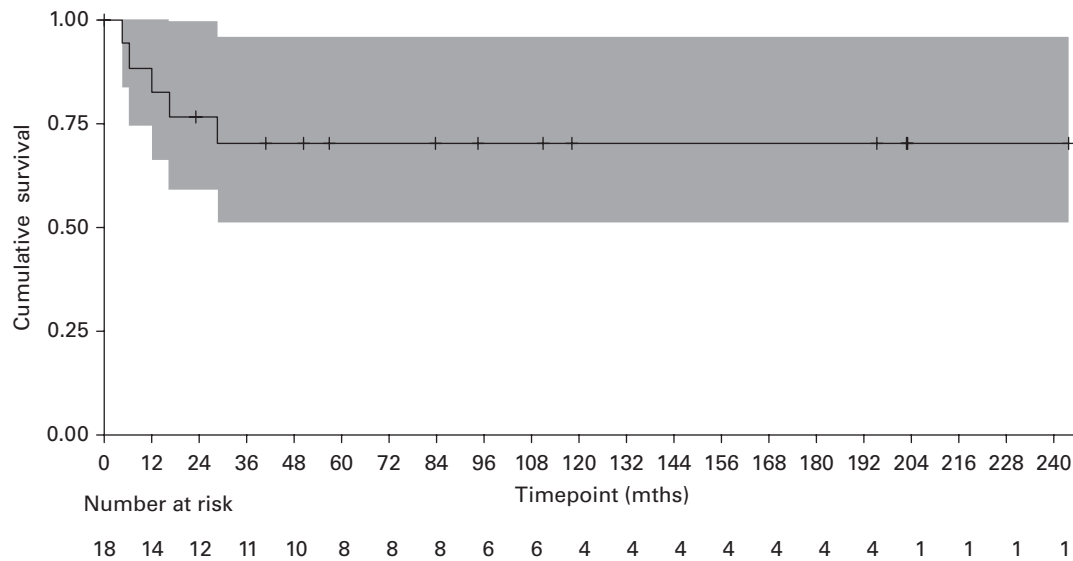


Fig. 4

Kaplan-Meier curve demonstrating overall survival for patients. The overall two-year survival was 79% (95% CI 59 to 100).

which decreased the overall implant survival rates. The overall rate of complications for all prostheses was 46%. Sevelde et al⁷ documented that the five-year revision-free survival of expandable total femoral prostheses was 30%. Additionally, 80% of their patients developed hip instability, 60% developed contractures due to massive scarring, and 40% had a failure of the expansion mechanism.

A type BIIIA rotationplasty, in which the rotated upper tibia articulates with the acetabulum, is an alternative reconstruction option in children. In our series, the hip was stable in seven of the nine patients (77.8%) examined clinically at final follow-up. Remodelling of the proximal tibia because of its inherent osseous plasticity in children was seen in all patients. On the anteroposterior radiographs, the proximal tibia showed a beaking of the lateral condyle placed in the acetabulum. The lateral view showed a rounding of the proximal tibia in its attempt to be congruous with the hip.

Rotationplasty is not without its share of complications, but most occur early in the perioperative period. While two patients underwent a hip disarticulation in the immediate perioperative period due to vascular insufficiency, no additional surgical procedures were necessary in the remaining patients at a median follow-up of ten years, the longest follow-up being 20 years. This is an advantage of rotationplasty over mega-prosthetic arthroplasty in young children where most of the patients need revision surgery. In the study by Sevelde et al,⁷ the average number of surgical procedures for patients who had an expandable total femur was 7.2 (2 to 20). Hobusch et al,⁸ in their study of rotationplasty patients with a minimum follow-up of 20 years, mentioned that failures after endoprosthetic reconstructions occur frequently in the long term, whereas they rarely occur after rotationplasty. Hence the use of rotationplasty might be a benefit not only to individual patients, but also to stakeholders in healthcare systems.

Unlike other forms of rotationplasty, in a type BIIIA rotationplasty, because the total femur is excised and replaced by the entire tibia, the limb length achieved at skeletal maturity cannot be modified at the time of index surgery based on estimates of remaining growth. While there are no data in the literature on limb length at maturity after a type BIIIA rotationplasty, in our study there was no clinically significant limb length inequality when comparing the length of the rotationplasty limb to that of the contralateral femur. Mean shortening in skeletally mature patients was 2.25 cm (0 to 4), and the mean shortening in adolescents was 0 cm (-1 to +1).

The extensive resection of bone and associated soft-tissue adds to oncological safety as none of the patients had a local recurrence, despite having high-risk features such as large volume disease and a pathological fracture (in seven of the 18 patients). In Medellin et al's² series of expandable total femoral prostheses, three of 20 patients had a local recurrence.

Our mean MSTS score of 23 (22 to 23) was lower than the mean score of 26 reported by Medellin et al² in all their expandable femoral prostheses at final follow-up or before revision. Functional outcome data directly comparing BIIIA rotationplasty with hip disarticulation or total femoral arthroplasty are lacking. The functional scores of patients in our series were comparable to those after conventional rotationplasty. When comparing patients with a minimum 20 years of follow-up with an amputation above the knee or at a lower level with those who had an A1 rotationplasty, patients who underwent rotationplasty scored higher in the EuroQol index with fewer problems in normal activities.⁸ Hillmann et al⁹ evaluated 61 patients after a rotationplasty and found that 85% of patients participated in high-level sports. The most common sport played was swimming followed by cycling, football, basketball, and volleyball. Sporting activities involving prolonged running such as soccer or jogging were done by 25% of patients.⁹ None of the patients

in the study group were members of special sports clubs for handicapped people but of regular sports groups.^{7,9} In a meta-analysis that showed the effectiveness of rotationplasty for lower limb tumours in terms of gait parameters measured by gait analysis, Filis et al¹⁰ mentioned that while stride length was reduced, gait velocity appeared comparable to normal controls. Hobusch et al⁸ emphasized the benefit of rotationplasty as a durable surgical method that enabled patients for high physical performance. Bernthal et al¹¹ reported that patients with a rotationplasty returned to high-impact sports like skiing, running, and wrestling without the concerns or inhibition reported by patients who underwent endoprosthetic arthroplasty. Literature on participation in recreational sports for type BIIa rotationplasty is lacking. In our series, 82% of patients participated in recreational sports, the most common being cricket, badminton, swimming, and cycling. In a study addressing patient and parent perceptions of body image, social acceptance, physical function, and satisfaction with treatment after limb salvage with an expandable prosthesis, Henderson et al¹² reported that satisfaction with body image and appearance was good in most patients. Patients were more likely to be happy with their appearances than with their bodies, indicating a level of frustration with functional ability as these patients were cautioned to refrain from sports involving contact or substantial impact to the lower limbs due to risk to the endoprosthesis.

Although we have not evaluated the emotional acceptance and long-term psychological effects of rotationplasty in this study, they have been extensively assessed in previous studies. Rödl et al¹³ used quality of life questionnaires in 22 patients at a mean ten years' follow-up and found no reduction in psychological adaptation and similar life contentment compared to healthy individuals. In a quality of life study in survivors with a rotationplasty, Veenstra et al¹⁴ reported that one-third to half of the patients reported negative effects of the surgery on initiating social and/or intimate contacts, body image, and sexuality. However, levels of psychosocial functioning, general quality of life, and social support were highly comparable with those of healthy peers and two-thirds of patients engaged actively in sports. Patients reported wearing the prosthesis continuously and were, in general, satisfied with its fit. In Gradl et al's¹⁵ series, the scores for vitality, social functioning, and mental health were significantly higher at long-term follow-up of patients with a rotationplasty when compared with a representative healthy German sample cohort (attributed to post-traumatic growth, benefit finding, and positive reappraisal coping). Most of their patients were well adjusted in terms of social integration and showed good-to-excellent results in terms of physical and mental health.

In conclusion, at a median follow-up of over ten years, BIIa rotationplasty is an oncologically sound procedure and is a reliable, enduring, revision-free reconstruction option after total femoral excision in children, enabling them to participate in all day-to-day activities and sports.



Take home message

- Total femoral resections for sarcomas pose a substantial reconstruction challenge in children.
- Type BIIa rotationplasty (tibia into hip) is an oncologically sound procedure after total femur excisions in children. It offers a reliable, enduring, revision-free reconstruction option, enabling them to actively participate in all day-to-day activities and sports.

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Author information:

M. Pruthi, MS (Orth), Professor
 R. Agarwal, MS (Orth), Fellow
 P. Nayak, MS (Orth), Professor
 P. Hegde, MS (Orth), Assistant Professor
 A. Puri, MS (Orth), Professor & Head
 Orthopaedic Oncology, Department of Surgical Oncology, Homi Bhabha National Institute, Tata Memorial Hospital, Mumbai, India.

A. Gulia, MS (Orth), Professor, Orthopaedic Oncology, Department of Surgical Oncology, Homi Bhabha Cancer Hospital & Research Centre, Tata Memorial Centre, New Chandigarh, India.

Author contributions:

M. Pruthi: Data curation, Formal analysis, Supervision, Writing – original draft, Writing – review & editing.

R. Agarwal: Data curation, Formal analysis, Writing – original draft, Writing – review & editing.
P. Nayak: Data curation, Formal analysis, Supervision, Writing – original draft, Writing – review & editing.
P. Hegde: Data curation, Formal analysis, Writing – original draft, Writing – review & editing.
A. Gulia: Formal analysis, Writing – original draft, Writing – review & editing.
A. Puri: Conceptualization, Formal analysis, Project administration, Validation, Writing – original draft, Writing – review & editing.

Funding statement:

The authors received no financial or material support for the research, authorship, and/or publication of this article.

ICMJE COI statement:

The authors have no disclosures to report.

Data sharing:

The datasets generated and analyzed in the current study are not publicly available due to data protection regulations. Access to data is limited to the researchers who have obtained permission for data processing. Further inquiries can be made to the corresponding author.

Acknowledgements:

The authors would like to acknowledge Mr Roberto Scanferla for his contribution in data acquisition. Written informed consent was obtained from the patients for their anonymized information to be published in this article.

This article was primary edited by A. C. Ross.