

Reconstruction of Congenital Pseudarthrosis of the Radius Using a Vascularized Tibial Periosteum Transplant: A Case Report

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ABSTRACT

Congenital pseudarthrosis of the radius is a rare condition, commonly associated with neurofibromatosis or fibrous dysplasia. We present the first reported case of using a vascularized periosteal flap for the treatment of congenital pseudarthrosis of the radius. The patient was a 14-month-old boy with neurofibromatosis type 1, presenting with severe shortening of the forearm, radial deviation of the wrist, and limitation of pronation-supination (60°-80°). Radiographs revealed pseudarthrosis in the distal third of the right radius. Surgical treatment involved debridement, bone fixation with a Kirschner wire, and placement of a vascularized periosteal flap harvested from the contralateral tibia to cover the pseudarthrosis site. Radiographic evidence of callus formation was observed two weeks postoperatively, accompanied by full wrist flexion-extension and improved pronation-supination (90°-90°). At five weeks, the patient resumed activities without orthotic protection. The use of vascularized tibial periosteum represents an innovative approach for treating congenital pseudarthrosis of the radius, demonstrating rapid bone healing and early return to activity, with no morbidity at the donor site.

Keywords: Vascularized periosteum; congenital pseudarthrosis; reconstruction; flaps; neurofibromatosis.

Level of Evidence: IV

Reconstrucción de una pseudoartrosis congénita de radio mediante un trasplante de periostio vascularizado de tibia: reporte de un caso

RESUMEN

La pseudoartrosis congénita de radio es un cuadro raro, comúnmente asociado a neurofibromatosis o displasia fibrosa. Presentamos el primer caso de un colgajo de periostio vascularizado utilizado para tratar la pseudoartrosis congénita de radio. Se trata de un varón de 14 meses de edad con neurofibromatosis tipo 1, acortamiento severo del antebrazo y desviación radial de la muñeca. La prono-supinación estaba limitada (60°-80°). En las radiografías, se observó una pseudoartrosis del tercio distal del radio derecho. El tratamiento quirúrgico consistió en desbridamiento, fijación ósea con una aguja de Kirschner y la colocación de un colgajo de periostio vascularizado de tibia de la pierna contralateral cubriendo el sitio pseudoartrosico. A las 2 semanas de la operación, se observó la formación de callo en la radiografía, con flexo-extensión completa de la muñeca y prono-supinación de 90°-90°. A las 5 semanas, reanudó sus actividades sin protección ortésica. El uso de periostio vascularizado de tibia es una opción innovadora para tratar la pseudoartrosis congénita de radio, ya que la consolidación ósea y el retorno a las actividades son rápidos, sin morbilidad asociada en el sitio donante.

Palabras clave: Periostio vascularizado; pseudoartrosis congénita; reconstrucción; colgajos; neurofibromatosis.

Nivel de Evidencia: IV

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INTRODUCTION

Congenital pseudarthrosis of the radius is a very rare condition, commonly associated with neurofibromatosis or fibrous dysplasia.^{1,2} The local bone repair process is defective, similar to what occurs in congenital pseudarthrosis of the tibia. This is likely due to the presence of an abnormal periosteum, which leads to the formation of a fibrous hamartoma at the pseudarthrosis site following a fracture of a previously dysplastic bone.

Several therapeutic options have been described, including conventional bone grafting, vascularized bone grafts, bone shortening, and single-bone forearm procedures.³

According to the literature, vascularized fibula grafting is currently the preferred treatment for this specific disorder.²⁻⁵ The use of vascularized periosteal flaps to treat complex pseudarthroses in children has recently gained popularity^{6,7} and has proven highly effective, even in challenging cases such as congenital pseudarthrosis of the tibia.^{8,9}

We present the first reported case of a vascularized periosteal flap used for the treatment of congenital pseudarthrosis of the radius.

CLINICAL CASE

A 14-month-old male with neurofibromatosis type 1 presented with severe forearm shortening and radial deviation deformity of the right wrist (Figure 1). On physical examination, passive pronation-supination of the forearm was limited to 60-80°, with no flexion-extension deficit. Radiographs revealed an area of atrophic pseudarthrosis with angulation of the distal third of the radius, with an ulnar and volar apex.



Figure 1. Deformity of the right forearm. Dorsal (A) and volar (B) clinical images. Forearm radiographs, anteroposterior (C) and lateral (D).

Surgery was performed through a volar longitudinal approach to the forearm and included debridement of the pseudarthrosis site, extensive resection of the anomalous native radial periosteum, and diaphyseal osteotomy of the middle third of the radius for realignment in both the coronal and sagittal axes. Finally, bone fixation was achieved using a 2-mm retrograde K-wire (Figure 2).

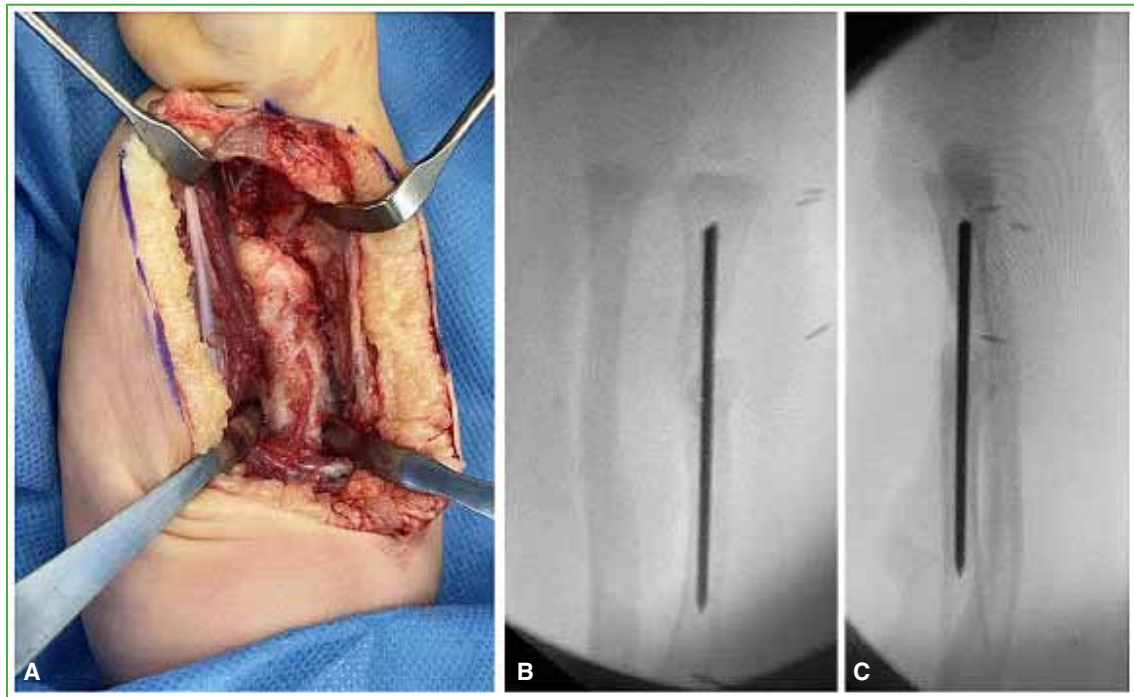


Figure 2. A. Abnormal radial periosteum (fibrous hamartoma). B and C. Anteroposterior and lateral radiographic images, respectively, of K-wire placement.

Subsequently, a vascularized periosteal flap was harvested from the contralateral tibia following the technique described by Soldado et al.⁸ (Figure 3). The flap was positioned to cover the radial periosteum, the pseudarthrosis site, and the osteotomy site, leaving a skin island for postoperative flap monitoring. Anastomosis of the tibial vessels to the radial vessels was performed. There were no immediate or late postoperative complications.

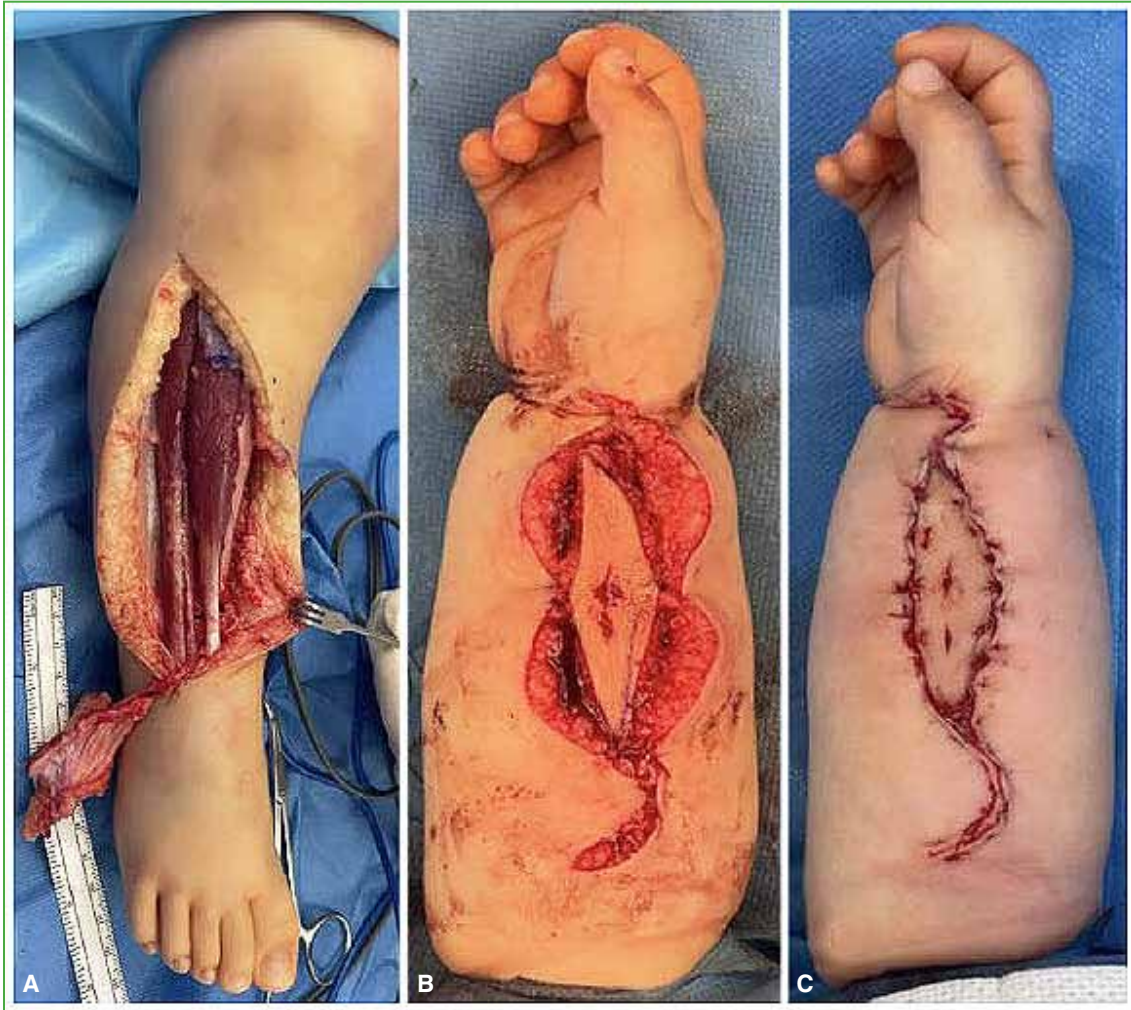


Figure 3. A. Donor area. Vascularized periosteal flap of contralateral tibia. B and C. Recipient site on the right forearm with skin island for monitoring.

Control radiographs taken three weeks after surgery showed solid callus formation, defined as the presence of bone bridges in at least two projections (Figure 4). To assess healing, we used the Radiographic Union Score for Radius, a simple and standardized method described for distal radius fractures.¹⁰ A radial shortening of 5 mm was observed. Passive forearm exercises were prescribed. Due to the COVID-19 outbreak, K-wire removal was delayed until six months postoperatively.

At the last follow-up, the patient had 90° of forearm supination, 90° of pronation, and full wrist flexion-extension. The child resumed all daily and school activities without the need for orthotic protection.



Figure 4. Postoperative radiographic controls. **A.** At 3 weeks. **B.** At 8 weeks.

DISCUSSION

Several surgical techniques have been described for the treatment of congenital radial pseudarthrosis. Standard bone grafting is generally not recommended due to its high failure rate.¹¹ The single-bone forearm procedure is rarely indicated for this condition. While it is a simpler procedure compared to microsurgical options, it sacrifices forearm rotation and may still be associated with pseudarthrosis, as abnormal bone remains at the union site.¹²

Biological procedures have been proposed to improve consolidation rates. Bone resection and free vascularized fibula grafting have shown excellent outcomes in congenital forearm pseudarthrosis.^{13,14}

Allieu was the first to use the vascularized fibula technique in 1981.⁴ He recommended early treatment to maximize the growth potential and remodeling capacity in pediatric patients.¹³ The youngest reported patient to undergo this procedure was one year old.¹⁴

According to Barrera-Ochoa et al., the use of vascularized periosteal flaps for treating complex osseous pseudarthrosis has gained popularity in recent years across a variety of clinical scenarios.^{6,15-18}

Vascularized free periosteal flaps, harvested from the fibula or tibia, have been used for the treatment and prevention of osseous pseudarthrosis in children, with excellent outcomes. Studies suggest that the tibial vascularized periosteal flap, based on the anterior tibial vessels, is easier to harvest and has greater osteogenic capacity compared to the fibular vascularized periosteal flap.^{8,9,19} Using this technique, we performed the first vascularized periosteal flap procedure for congenital forearm pseudarthrosis and achieved rapid consolidation. The advantage of using a tibial vascularized periosteal flap over a fibular one is the avoidance of potential donor site complications associated with fibular removal, which are not negligible.²⁰ To date, no local morbidity has been reported after tibial vascularized periosteal flap harvesting. Another advantage is the early return of the child to daily activities, as this technique leads to rapid consolidation. Indeed, our patient was allowed mobility three weeks after surgery.

A major limitation of our study is that it is a single-case report with short-term follow-up. A larger study including additional cases treated with this technique, along with a comparison to the vascularized fibular periosteal flap and long-term follow-up, would be valuable.

CONCLUSIONS

The tibial vascularized periosteal flap proved to be highly effective and rapid in achieving consolidation in a patient with congenital pseudarthrosis of the radius. Successful reconstruction was achieved using this novel technique.

Conflict of interest: The authors declare no conflicts of interest.

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