

Derotational osteotomy of the proximal radius and the distal ulna for congenital radioulnar synostosis

Nguyen Ngoc Hung

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Abstract

Objective To evaluate the clinical and functional results of a technical procedure in the surgical treatment of congenital radioulnar synostosis in children.

Materials and methods A prospective study had been undertaken from January 1992 to December 2004. Thirty-four patients with congenital radioulnar synostosis that are fixed in pronation were recruited. Congenital radioulnar synostosis was classified for two types according to Tachdjian's criteria. All patients were treated by resection of the proximal radius and the distal ulna to remove a segmental bone of both parts of the forearm. After K-wires are inserted intramedullarily into both bones, the forearm is derotated manually, followed by cast immobilization.

Results There were 34 patients (52 forearms) with congenital radioulnar synostosis, whom the average age at surgery was 6 years and 3 months. There were two types of congenital radioulnar synostosis: Type 1 in six forearms (11.6%) and Type 2 in 46 forearms (88.4%). The preoperative forearm rotation ranged from 65° to 85° pronation. The postoperative forearm rotation angle was corrected from 0° to 30°; the best end position appears to be 70–100% of pronation. Of the patients, 78.8% had good or excellent results. All patients were operated on without complications; five patients had loss of correction during cast immobilization. Overall, the patient's ability to perform daily activities showed a marked improvement after surgery.

Conclusion This method is a simple and safe technique to derotate the forearms of patients with congenital radioulnar synostosis that are fixed in pronation.

Keywords Congenital radioulnar synostosis · Head radius dislocation · Rotational osteotomy · Derotational osteotomy

Introduction

Congenital radioulnar synostosis is an uncommon deformity of the upper limb in which proximal portions of the radius and ulna fuse with each other and restrict the rotation of the forearm. In 1932, Fahlstrom reviewed all of the reported cases in the world literature and found only 185 since Sandifort's original description in 1793 [1]. In cases in which the forearm is fixed in pronation over 60° and the patient complains of disability in daily life, derotational osteotomy is recommended to achieve a more functional position. Disappointing results of earlier attempts to resect the synostosis and restore forearm rotation are reported [2, 3]. Osteotomy is usually performed through the site of fusion, but extensive release of the soft tissue around the osteotomy is mandatory for adequate correction [1, 4, 5]. Otherwise, soft-tissues tightness may lead to loss of correction or vascular complication because considerable rotation takes place in a very limited area [6, 7, 8]. Most efforts to separate the synostosis and obtain rotation of the forearm have ended in failure.

Since 1992, we have developed a method of osteotomy for congenital radioulnar synostosis in which osteotomy is performed at the shafts of the radius and ulna. It is combined to enable the removal segmental bone of both parts

N. N. Hung (✉)
Vietnam National Hospital of Pediatrics,
18/879 La Thanh Road, Dong Da District, Hanoi, Vietnam
e-mail: ngocyenhung@gmail.com

of the forearm. The purpose of this study is to evaluate the long-term results of this technique.

Materials and methods

From January 1992 to December 2004, we performed in 39 patients (60 forearms), a derotational osteotomy of the proximal radius and distal ulna using the method mentioned above. Five patients (eight forearms) were lost to follow-up. The remaining 34 patients (52 forearms as 18 patients had a bilateral synostosis) formed the basis of this study. The patients was operated by only one surgeon (author). There were 22 males (64.7%) and 12 females (35.3%). All pertinent clinical and operative records were prospectively reviewed.

Clinical and roentgenographic check-up

All patients completed a questionnaire and were interviewed by a member of staff at the study hospital. A complete clinical examination of the upper limb was performed, which included measurements of the range of movement, position of the forearm, carrying angle, and length of the forearm.

The range of pronosupination of the forearm was measured by the angle between the longitudinal axis of the humerus and the line of the radial and ulnar styloid processes (forearm rotation). In patients with congenital radioulnar synostosis, a pronosupination degree of the palm is possible by the means of compensatory rotation motion in the wrist and the carpometacarpal joints. Therefore, the range of pronosupination of the palm was defined by the angle between the longitudinal axis of the humerus and the axis of metacarpophalangeal joints from the index to the little finger in the palm (apparent rotation) [5].

The roentgenographic examination included both elbows and wrists in two planes and were compared with previous radiographs. The roentgenograms taken in the anteroposterior and lateral positions show a radioulnar synostosis, involving the upper one-and-a-half inches of the bones. As a result of this synostosis, the distal end of the ulna seen is displaced posteriorly. The bases of the metacarpal bones appear somewhat atrophic, possibly the result of disuse.

Classification of congenital radioulnar synostosis

Our patients were classified according to Tachdjian's criteria [9]:

Type 1 Figure 1: the radial head may be fused to the ulna or it may be completely absent (known as the "headless type")



Fig. 1 Radioulnar synostosis with hypoplastic ulna and without dislocation of the radial head

Type 2 Figure 2: the radial head is malformed and possibly dislocated

Operative procedures and postoperative management

Indication

The average preoperative degree of fixed rotation of the operated forearms is over 60°. In such a case, a child has



Fig. 2 Radioulnar synostosis with dislocation of the radial head

significant functional limitation when they try to hold a rice bowl, drink water from a glass, or receive a coin in the open palm and they have an inability to use spoons or chopsticks and to wash their face. The patient with obvious osseous synostosis and with or without dislocated radial heads appear on the radiographs.

Operative procedure

In the first skin incision according to Thompson's posteromedial approach for the ulna, the ulna was approached through a longitudinal incision along the subcutaneous border starting 5 cm proximal to the styloid process and continuing in a proximal direction for 4 cm. In the second skin incision according to Henry's anterolateral approach [12], the radius was approached through a longitudinal incision along the subcutaneous border starting from fusion site and continuing in a distal direction for 4 cm. The superficial radial nerve, the ulnar nerve, and the radial and ulnar artery were identified and gently retracted so that they are protected. A shortening bone was performed and the bone was resected at the diaphysis, that is, at the distal one third of the ulna and the proximal one third of the radius, where approaches to the bones are straightforward and better bone healing is anticipated. This method is different from osteotomy at the fusion mass. The proximal radius was exposed and the distal margin of the synostosis was identified with the use of injection needles, and the insertion of the pronator teres muscles followed. The radius was resected approximately 2 cm distal to these landmarks. The ulna was resected approximately 5 cm proximal to the ulnar styloid. After both bones were exposed subperiosteally, several holes were made according to two transverse lines per bone at the diaphysis by using a Kirschner wire to ensure that the hand saw will not slip on the bone, guided by these holes (Figs. 3a, 4). The transverse osteotomy was performed using a hand saw. After the osteotomy, a segmental bone of approximately 1.5 cm of both bones of the forearm is removed (Fig. 5), and K-wires—1.5 mm in diameter—were inserted into the radial styloid and the ulnar styloid, which passed through to the resecting bone sites (Fig. 3b). The two bones were reduced and the K-wires were advanced across the resecting bone sites (Fig. 3c). Subsequently, the K-wires were controlled to exit at that point without damage to the growth plate or joints, confirmed by radiography. After the intramedullary K-wires were inserted into both bones, the forearm was derotated manually to the position that is neutral to 30° pronation for the dominant hand and neutral for the nondominant hand (Figs. 6, 7). Adequate rotation could be achieved without difficulty at both the distal radioulnar joint and the osteotomy sites. This procedure was followed by

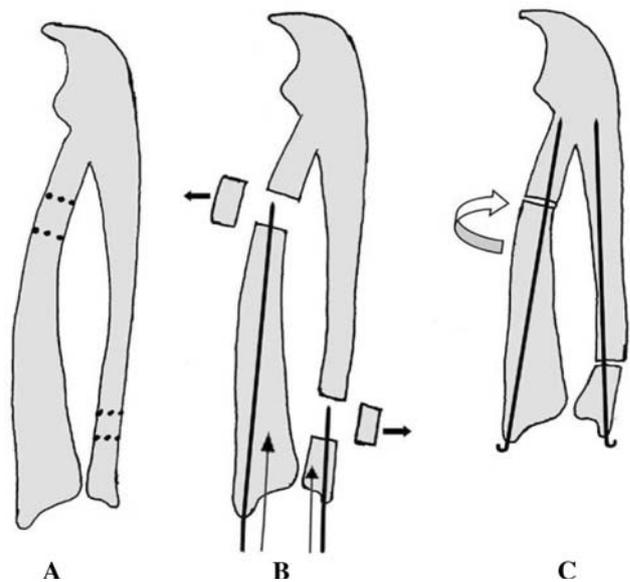


Fig. 3 Surgical method. **a** The osteotomies/resections are drawn on both parts of the forearm. **b** Both bone resections are performed, a segmental bone of approximately 1.5 cm of both bones of the forearm is removed, and K-wires are inserted in the radial styloid and the ulnar styloid. **c** The two bones are reduced and the K-wires are advanced across the resecting bone sites; the forearm was derotated manually to the position neutral to 30° pronation for the dominant hand and neutral for the nondominant hand

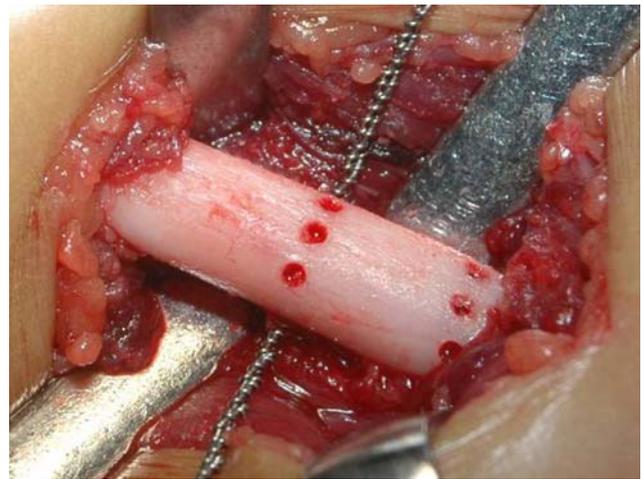


Fig. 4 Several holes were made and osteotomized by a hand saw

forearm immobilization by an above-the-elbow plaster cast applied on the operating table (Fig. 8).

After operation

At 6 to 8 weeks after surgery when the formation of sufficient callus was evident radiographically, the cast was removed and the forearm was protected with a removable splint. The K-wires were left through the skin and removed

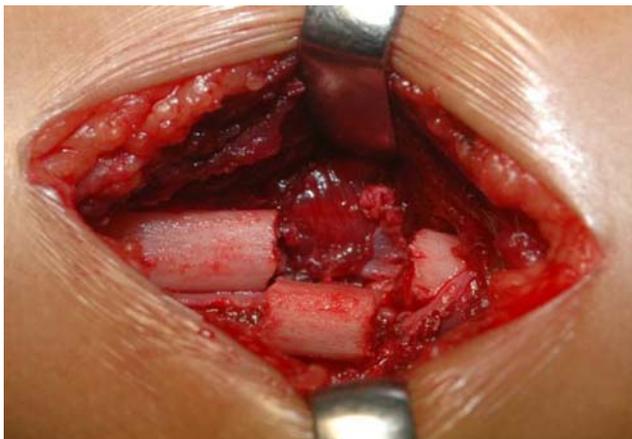


Fig. 5 Defect of the bone and removing a bone segment



Fig. 6 K-wires are inserted intramedullarily into both bones of the forearm

in the operating room under local anesthesia after the radiographic confirmation of bone union.

Follow-up

Patients were re-examined at 3 weeks, 6 weeks, 3 months, 6 months, 1 year, and afterwards every year. All patients were followed up to clinical or radiographic healing of the forearm bones.

Results

From January 1992 to December 2004, 34 patients (52 forearms) were operated. The mean age of the patients at



Fig. 7 Postoperative union of bone after 8 weeks



Fig. 8 The upper extremity was immobilized in a long arm cast to hold the elbow in 90° of flexion and retain the forearm position according to the planned preoperation

surgery was 6 years and 3 months (range 3 years and 9 months to 9 years and 11 months). The distribution was nine right forearms and seven left forearms; 18 patients (52.9%) had bilateral involvement.

None patients had other congenital deformities, and there were no family history of congenital radioulnar synostosis.

Criteria used for assessing the results of the operation

The criteria used for assessing the results of the operation are shown in Table 1. The complications included: bone

Table 1 Criteria used for assessing the results

	Forearm position (° pronation)	Rate of results at latest follow-up (%)	Activities of daily living (holding a rice bowl, using chopsticks, washing face, dressing and buttoning clothes, etc.)
Excellent	>0° to ≤15°	100 to ≤85	No difficulty
Good	>15° to ≤30°	≥70 to <85	Mild difficulty
Fair	>30° to ≤45°	≥50 to <70	Moderate difficulty
Poor	>45°	<50	Severe difficulty or inability or having complications



Fig. 9 Postoperative 5 years and 6 months; forearm pronation



Fig. 10 Postoperative 5 years and 6 months; forearm supination with compensatory movement around the wrist and elbow

union, disturbance of the forearm bone, angulation of the forearm, compartment syndrome, or other complications (vascular compress, Volkmann’s ischemia, nerve palsy, etc.).

Classification of congenital radioulnar synostosis

Type 1 “headless type”: 6 forearms (11.6%) and Type 2: 46 forearms (88.4%) (anteriorly dislocated radial head in 18 forearms and posteriorly dislocated radial head in 28 forearm).

The average length of follow-up was 5 years and 4 months (ranging from 2 years and 6 months to 10 years and 9 months) (Figs. 9, 10).

Long-term follow-up: excellent results were seen in 10 forearms (19.2%), good results in 31 forearms (59.6%), and fair results in 11 forearms (21.2%). There were no poor results and no complications in any of the patients (Table 2).

The preoperative forearm position ranged from 65° to 85° pronation (mean 82° pronation in dominant and 74° in

nondominant hands). The postoperative forearm position angle was corrected from 0° to 30° pronation (mean 6° pronation in dominant and 10° pronation in nondominant hands).

There were 11 forearms with fair results. The postoperative forearm position had a mean of 36° pronation. Those patients still had some difficulty in holding a rice bowl, using chopsticks, and suffers some disability in daily activities, and these patients were also satisfied with the results of the surgery.

There was a slight loss (15°–20°) of correction during cast immobilization in five of the forearms (three forearms in the dominant hand—postoperative forearm position was 20° pronation; when the cast was removed, the forearm position was 40° pronation; two forearms in the nondominant hand—postoperative forearm position was neutral; when the cast was removed, the forearm position was 35° pronation).

Twenty-two patients (13 patients bilateral) were younger than 6 years old were operated. The postoperative follow-up showed a mean 6° pronation in 35 forearms and

Table 2 Compare preoperative range of movement (ROM) and postoperative ROM

	Preoperative ROM		Postoperative ROM	
	Forearm rotation, ° (average)	Apparent rotation pronation/supination, ° (average)	Forearm rotation, ° (average)	Apparent rotation pronation/supination, ° (average)
Dominant (<i>n</i> = 24)	82	110/0	6	31/40
Nondominant (<i>n</i> = 28)	74	115/10	10	34/48

the remaining 12 patients (five patients bilateral) were older and showed a postoperative follow-up with mean 25° pronation in 17 forearms

In the bilateral case, the operated hand was the dominant hand and, thus, the patient's ability to use chopsticks, wash his face, and button his clothes markedly improved after surgery. The unilateral cases also showed great improvement in performing daily activities, such as holding a rice bowl, dressing, and washing, and these patients also were satisfied with the results of the surgery.

Bone union was achieved in all patients at 7.2 weeks (range, 6.6–7.4 weeks), and the time for complete removal of the cast ranged from 6 to 9 weeks (mean 8 weeks).

No complications were observed for the surgical technique by the reviewer.

The growth of the radius and ulna had not been disturbed; great functional improvements in performing activities of daily living, such as operating a keyboard, were achieved in all patients.

Discussion

Congenital radioulnar synostosis is an uncommon deformity of the upper limb. Blaine [13], quoting Mouchet and Leleu [14] stated that radioulnar synostosis was apparently first discovered by Lenoir at an autopsy in 1817. However, the earliest record is that of Sandifort, who, in 1793, reported three cases [1]. Smith, Verneuil and Dubois reported a single case in 1852. In 1856, Malgaigne wrote about one case and was followed in 1863 by Voigt. Then, no case was heard until 1880, as Allen reported one more case. It was followed by Pye-Smith in 1883 with another [15]. In 1892, Morrison reported one case and Abbott contributed to the largest series, comprising seven cases. The latter series occurred in one family across four generations. This was the first authentic evidence of a familial tendency [15–17]. In 1932, Fahlstrom reviewed all of the reported cases in the world literature and found only 185 since Sandifort's original description in 1793 [1].

In 1924, Davenport [18] reported the largest series of 15 cases and covered this condition most thoroughly. Davenport, as the major essayist of this group, made a complete

ontogenetic and phylogenetic study, and displayed all of the known hypotheses of abnormal origin. He made the statement that, in 55% of the parents out of 15 cases, one of the parents was synostotic. He came to the conclusion that the radioulnar synostosis was partially sex-limited and that its occurrence depended upon several variable factors. He believed that consanguineous marriage was a possible factor and that the condition occurred twice as often in males as in females. Our patients group was composed of 64.7% males and 35.3% females.

Regarding the suitable age for surgery, Griffet et al. [19] recommended that operation is best carried out between the ages of 4 and 10 years. Poueyron et al. [20] proposed that surgical treatment in congenital radioulnar synostosis should consider many problems: indication, technique, and ideal age (due to the more common vascular or nervous postoperative complications if older). We consider that the optimal age ranges from 3 to 6 years. At these ages, the osteotomy is easy, and it is likely to ensure sufficient remodeling of the radius and ulna. Regarding the postoperative results, the patients were operated younger than 6 years old with mean of 6° pronation and patients older than this age had a mean of 25°.

From our evaluation, we concluded that all patients can benefit from this rotational osteotomy. Ideally, it should be used for only a patient who complains of specific limitation or who is employed in or entering an occupation in which a change in the position of the forearm would result in functional improvement. Nevertheless, delaying the performance of this procedure until adolescence or adulthood might increase the risk of operative complications due to long-standing osseous and soft-tissue deformity. However, this study demonstrated that most procedures performed in childhood are probably of functional benefit. There were no patients in this study who demonstrated pathological findings in the wrist or shoulder; therefore, the forearm can be actively compensated by the motion of an adjacent joint.

At present, two major surgical procedures are available for this congenital deformity. One is the operative mobilization to separate the radioulnar synostosis and restore forearm rotation [21–23]. The operative mobilization is thought to be the best method theoretically; however, it requires a vascularized fat graft after release of the

synostosis and it is often difficult to realign the radial head in the proper position in which the radial head shows posterior or anterior dislocation [8]. Another surgical procedure is osteotomy to correct the forearm position that it is suitable for the patient's activities of daily living. Three types of osteotomy procedures have been used to correct forearm rotation: osteotomy at the synostosis involves operative complexity, the critical rotation takes place over a much more narrow space and the excessive soft-tissue tightness may result in the loss of correction, a circulatory compromise, or a neural entrapment and some postoperative complications have been reported, including vascular compromise, such as Volkmann's ischemia, shortening and angulation of the forearm, and posterior interosseous nerve palsy [4–6, 8, 14, 15, 19, 24–26]; osteotomy at one site in the distal diaphysis of the radius [21]; and osteotomy at two sites in the diaphysis of the radius and the ulna [7, 23, 27]. In the osteotomy, at two sites without leveling in the diaphysis of the radius and the ulna, the procedure is easier and there are fewer complications, although internal fixation is necessary, requiring a second surgical operation to remove the hardware.

The length of the synostosis ranged from 3–5 cm [28, 29], and was usually 2–6 cm [18, 24]. We measured the length of fusion mass at an average of 18 mm (range, 13–25 mm) in Type 1 and 15 mm (range, 12–22 mm) in Type 2. Some authors performed shortening of the forearm by resection of the bone from the fusion mass or radius and ulna to decrease the tension of the soft tissues from 3 to 22 mm in length [28, 30–33]; we approved Yammine's opinion of shortening the forearm by resection of the bone by ≤ 2 cm [33] and we performed resection of 1.5 cm of bone. In our technique, the rotation takes place between the two osteotomy sites and the removal of a segment of bone decreases excessive soft-tissue tightness. In addition, because the ulna is osteotomized distal to the insertion of the pronator quadratus muscle, the derotation maneuver does not cause compression of the median nerve and brachial artery at the proximal part of the muscle. The other great advantage of our method is the ease of the surgical technique. The approach to the osteotomy sites of the radius and ulna is quite simple and extensive release of the soft tissue is not required. After the intramedullary K-wires are inserted, the forearm can be derotated manually into the planned position.

Adequate blood supply is another critical factor for bone healing. In the adult ulna, the proximal diaphysis receives its blood supply from a major nutrient artery, which enters the anterior surface of the bone 7.5 cm distal to the tip of the olecranon. The artery courses proximally, providing the primary blood supply to the proximal portion of the ulna. Distal to the entry point of this artery, several small perforating vessels from the anterior interosseous artery

provide the primary blood supply [34]. Therefore, there is a relative watershed zone at the junction of the proximal and middle thirds of the ulna just distal to the nutrient artery [32, 35]. Fractures in this watershed zone of the ulna have been associated with poor healing. The relationship between the healing of one bone in the forearm to the other suggests that factors affecting healing of the ulna also affect the radius in the same extremity [36]. Szabo and Skinner [26] reported nonunions in 7 of 28 closed isolated ulna fractures. They determined that fractures in the proximal third of the ulna were prognostic of increased nonunion. Dalton [37] performed forearm osteoclasia with osteoclasia site in the proximal ulna and stated that 21 ulnas of 69 forearms had either delayed or nonunion. The ulnar union rates decreased significantly when the osteoclasia was performed in the proximal third of the ulna, so we did not carry out osteotomy at this site.

Murase [7] reported a 20° loss of correction in one of four cases, Tsuyoshi [23] reported a 20° loss of correction in one of four cases, and in our 5 of 54 forearms, the loss of correction occurred during the cast immobilization. The surgeon should, therefore, be cautious in regard to the position of the forearm in the cast until bone fusion is achieved. Once solid bone fusion is achieved, loss of correction with skeletal growth is unlikely because no tension is put on the flexors, extensors, or interosseous membrane by this method.

Green and Mital [6] stated that, in bilateral cases, the dominant hand should be placed 20° to 35° of supination and the other hand should be left in considerable pronation, whereas in unilateral cases, a supination of 10° to 20° is ideal. Ogino and Hikino [5] recommended that, for unilateral cases or the nondominant hand of bilateral cases, the forearm should be corrected to between neutral and 20° of supination (Fig. 10), and for the dominant hand of bilateral cases, it should be corrected to between neutral and 20° of pronation. We approved Tsuyoshi's opinion that the forearm should be derotated manually to the position of neutral to 30° pronation for the dominant hand and neutral for the nondominant hand [23]. With the increase in computer use over the past decade, however, people have begun to use keyboards much more frequently. Fixed supination of the forearm requires shoulder abduction and internal rotation to bring the forearm into pronation, and prolonged maintenance of this position during keyboard use is extremely fatiguing. Fixed pronation of the nondominant hand also is undesirable because Asian people hold a rice bowl with the nondominant hand in a slightly supinated position while eating. For these reasons, we prefer to correct the forearm position to between neutral and 30° of pronation for the dominant hand and to neutral for the nondominant hand in a unilateral case, as well as in a bilateral case. Some rotation would be possible through the hypermobile wrist

Table 3 Comparing results of postoperative follow-up with derotational osteotomy

Authors	Postoperative follow-up of forearm position	Complications
Simmons et al. [8] (<i>n</i> = 41 forearms)	10–15% pronation; 82% of patients had good or excellent results	Eight complications, four involving neurovascular compromise
Castelló et al. [10] (<i>n</i> = 4 forearms)	15°–0° pronation	No
Murase et al. [7] (<i>n</i> = 4 forearms)	25° pronation	No
Ramachandran et al. [11] (<i>n</i> = 6 forearms)	10° supination	One forearm with compartment syndrome
Hung (2008) (<i>n</i> = 52 forearms)	10–30% pronation; 78.8% patients had good or excellent results	No

joint. The patients with congenital radioulnar synostosis have considerable compensatory movement around the wrist [11], so performing derotational osteotomy to direct the forearm into a more comfortable position is a reasonable alternative in cases of fixed pronation radioulnar synostosis (Table 3).

In our follow-up results, the preoperative forearm position ranged from 65° to 85° pronation (mean 82° pronation in dominant and 74° in nondominant hands). The postoperative forearm position angle was corrected from 0° to 30° (mean 6° pronation in dominant and 10° pronation in nondominant hands). There were five forearms with 15°–20° loss of correction during cast immobilization. No complications were observed for the surgical technique by the reviewer.

Conclusions

1. Congenital proximal radioulnar synostosis is a rare deformity, frequently bilateral (52.9%), and more commonly seen in male patients (64.7%).
2. This method is a simple and safe technique to derotate the forearms of the patients with congenital radioulnar synostosis that are fixed in pronation.

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