Results of Single-staged Rotational Osteotomy in a Child With Congenital Proximal Radioulnar Synostosis: Subjective and Objective Evaluation

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Background: For congenital proximal radioulnar synostosis, both conservative and operative treatments have been described. Most of the studies describing surgical interventions are based on subjective evaluation of the forearm function and have used severe degree of forearm pronation as an indication for surgery. This study describes a single-staged rotational osteotomy of the proximal third ulna and distal third radius. The aim of the study was to assess the utility of the described surgical procedure by subjective and objective evaluations of the forearm function.

Methods: Forty-eight children with congenital proximal radioulnar synostosis were evaluated by subjective and objective assessments and were followed up prospectively. Subjective evaluation consisted of a set of 12 questions regarding the basic activities of life. Objective evaluation was made using the Jebsen-Taylor hand-function test and a classification system used by Failla and colleagues for 15 tasks described by Morrey and colleagues. Eleven children were treated conservatively. Thirty-six children underwent a single-staged rotational osteotomy of the proximal third ulna and distal third radius. After surgery, the evaluations were repeated. The mean age at surgery was 8.6 ± 3.7 years, and the mean postoperative follow-up period was 54 ± 13 months.

Results: All operated forearms showed a statistically significant improvement in functioning after surgery as per the subjective and objective evaluations. The mean time taken to carry out all activities before surgery was 47.7 + 10.0 seconds, which significantly reduced to 33.3 + 6.6 seconds after surgery ($P = 0.0001$) as per the results of the Jebsen-Taylor hand-function test. All good (n = 19) and fair (n = 11) results were converted to excellent (n = 30) after surgery as per the modified Failla classification. There were no neurovascular injuries as compared with other published techniques. Only 1 child had delayed union, and 1 had persistent dorsal angulation at the radial osteotomy site.

Conclusions: For patients with congenital radioulnar synostosis and pronation deformity interfering with function and quality of life, the single-staged rotational osteotomy of the radius and ulna is a good alternative procedure.

Level of Evidence: Level IV—therapeutic.

Key Words: congenital, radioulnar synostosis, congenital proximal radioulnar synostosis, rotational osteotomy, osteotomy, Jebsen Taylor, Jebsen-Taylor hand-function test, subjective evaluation, objective evaluation, surgery

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Congenital proximal radioulnar synostosis is a rare malformation of the upper extremity and always results in a fixed position of the forearm ranging from neutral rotation to severe pronation.[1] When the deformity is mild, little disability is evident, as the ipsilateral shoulder and wrist can compensate effectively.[1–7] With severe degree of pronation, activities of daily living can be severely impaired.[2,3,7]

Large numbers of treatment options, including observation,[1,7] rotational osteotomies,[2–4,8–11] mobilization of synostosis with the use of a free vascularized fascio-fat graft,[12–14] the Ilizarov method,[15,16] and an external fixation device,[17] have been described in the literature with different success rates. Most of these studies are based on the subjective evaluation of hand function and have used severe forearm pronation (> 60 degrees[3]) as an indication for surgery.

Our previous experience revealed that even 30 degrees of pronation can impair the forearm function markedly; hence, the surgical decision should be based on the difficulties in forearm functioning affecting the quality of life, rather than just the degree of pronation.

In the present study, a surgical technique is described, which is a single-staged rotational osteotomy of the proximal third ulna and distal third radius. When we consider the rotation of the forearm in a normal child, the supination as well as pronation is 90 degrees, with the elbow kept in flexion (Fig. 1A). Once the child keeps the elbow in extension, the compensatory movements at the shoulder and wrist allow excessive pronation (around 135 degrees) but only 90 degrees of supination (Fig. 1B). This indicates that the shoulder and wrist are less able to

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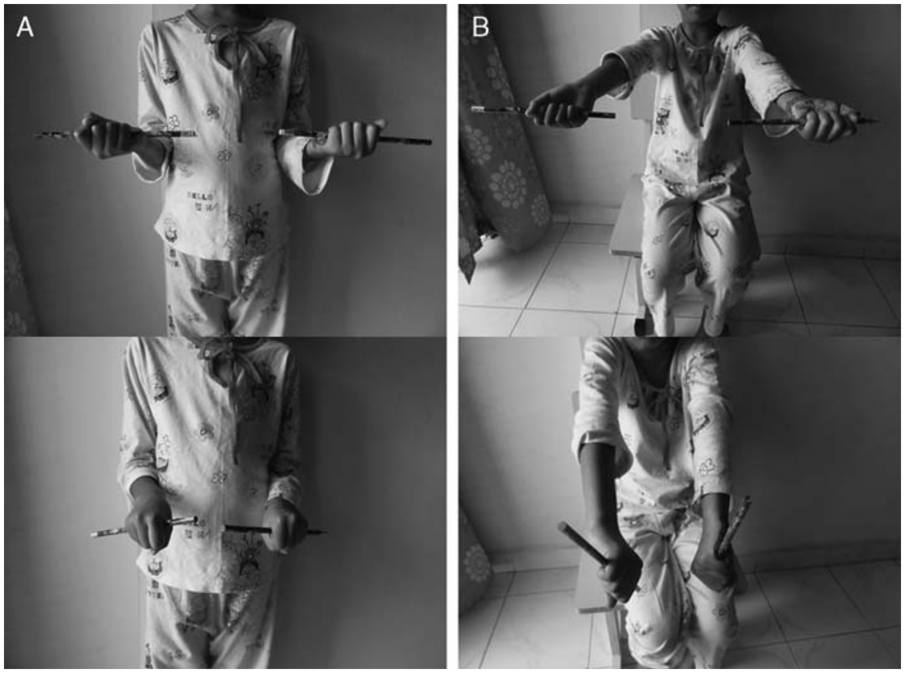

FIGURE 1. A, Rotation of the forearm in a normal child when the elbow is in flexion. The supination as well as pronation is 90 degrees. B, Once the elbow is kept in extension, the compensatory movements at the shoulder and wrist allow excessive pronation (around 135 degrees) but only 90 degrees of supination, indicating that the shoulder and wrist are less able to compensate for the loss of supination than of pronation. Therefore, surgically increasing the supination will improve the function and the child can easily compensate for the loss of pronation.

compensate for the loss of supination than of pronation. Therefore, surgically increasing the supination will improve the function and the child can easily compensate for the loss of pronation. Therefore, the following hypothesis was postulated: "forearm excessive pronation in congenital proximal radioulnar synostosis which interfered with function could be improved by corrective osteotomy of 20-30 degrees of supination." The aim of the study was to assess the effectiveness of the described surgical procedure by subjective and objective evaluation of the forearm function.

METHODS

This prospective study was conducted at a tertiary-level pediatric orthopaedic hospital during the period 2004 to 2012. Forty-eight children having proximal congenital radioulnar synostosis were assessed by performing subjective and objective evaluations.

The study was approved by the Institutional Review Board. Informed consent was obtained from the parents. All evaluations were carried out independently by a registered occupational therapist who was not a part of the study.

Parents and children were asked 12 questions regarding the basic activities of life (Table 1) that the child performed repeatedly throughout the day as far as the function of the upper limb is concerned. Difficulty in a child was assessed by giving a score of 0 or 1 for every activity. The scores were totaled for each child.

Objective evaluation of the forearm function was carried out using the Jebsen-Taylor hand-function test[18,19] and the classification system used by Failla et al[20] for 15 tasks described by Morrey et al.[21] The pronation deformity was measured with a goniometer.[22]

The Jebsen-Taylor hand-function test is a standardized timed test that includes 7 components: writing, turning cards, picking up small objects, simulated feeding, stacking checkers, picking up large heavy objects, and picking up large light objects. The reference range for

TABLE 1. Evaluation of Each Child for 12 Essential Activities of Daily Living

Sr. No.	Activities of Daily Living	Difficulty Present: Score 1 Difficulty Absent: Score 0
1	Taking food to the mouth	1/0
2	Cleaning the perineal area for hygiene	1/0
3	Holding plates and cups	1/0
4	Accepting coins and small objects in an open palm	1/0
5	Dressing	1/0
6	Writing	1/0
7	Taking the palm to the occiput	1/0
8	Combing hair	1/0
9	Catch a ball	1/0
10	Grasping	1/0
11	Holding	1/0
12	Playing with toys	1/0
	Range of possible scores	0 to 12

comparing the findings in normal children aged 6 years and above is given in the original article of Jebsen and Taylor.[18,19] The time taken by each study subject for each activity was noted. The findings were compared with available reference mean values in normal children standardized for age, sex, and hand dominance.[18,19] A total time of >2 SD around the mean was considered an abnormal result. The children were also assessed objectively by the classification system used by Failla et al[20] for 15 tasks described by Morrey et al.[21] They were asked to perform the following 15 tasks without an electrogoniometer: touch hand to the vertex (head), touch hand to the occiput, touch hand to the neck, touch hand to the chest, touch hand to the waist, touch hand to the sacrum, touch hand to the shoe, pour from a pitcher, put glass to the mouth, cut with a knife, put fork to the mouth, use a telephone, read a newspaper, rise from a chair, and open a door. The forearm function was considered excellent (if the child was able to perform all 15 tasks), good (10 tasks), fair (6 tasks), and poor (3 tasks) on the basis of the child's performance with respect to these 15 activities.

On the basis of the subjective and objective evaluation scores, a guideline for surgical indications was formulated. The indications for surgery included subjective functional limitations (a score of ≥ 4) and objective functional limitations (abnormal test results in at least 2 subtests of Jebsen and colleagues or nonperformance of at least 2 Morrey and colleague's tasks) irrespective of the degrees of pronation of the forearm. Children scoring less than these cutoff scores were treated conservatively.

The mean age of the nonoperated 11 children (7 boys and 4 girls; 13 forearms) was 9.73 ± 3.47 (range 4 to 16) years. The mean position of the forearm was 15.08 ± 3.64 (range neutral to 40) degrees of pronation.

Of the total 48 children, 37 fulfilled the criteria for surgical indications. One child did not opt for surgery. A total of 36 children underwent the single-staged surgical procedure. Postoperatively, all operated forearms were evaluated subjectively and objectively. Only those children who had a minimum postoperative follow-up of 3 years (n = 28) were included in the final evaluation. The presence or absence of pain at the wrist, elbow, shoulder, or any other joint was noted before and after the surgery.

Operative Technique

Under general endotracheal anesthesia, the patient was placed supine and a tourniquet was applied to the arm. A 2-cm posterior incision was made over the ulna, distal to the synostosis level. The periosteum was reflected. With an oscillating saw, under radiographic control, the ulnar osteotomy was carried out approximately a centimeter distal to the synostosis level. Using an oscillating saw, through a 2-cm incision placed dorsally at the distal forearm, the radial osteotomy was carried out at the distal diaphyseal-metaphyseal junction. The forearm was rotated to a position of 20 to 30 degrees of supination. The tourniquet was released and the distal circulation was confirmed. Wounds were closed keeping the fascia open. No implants or wires were used to stabilize the radius or the ulna. A thick cotton padding roll was applied, followed by an above-elbow cast with 90 degrees of flexion at the elbow and 20 to 30 degrees of supination at the forearm.

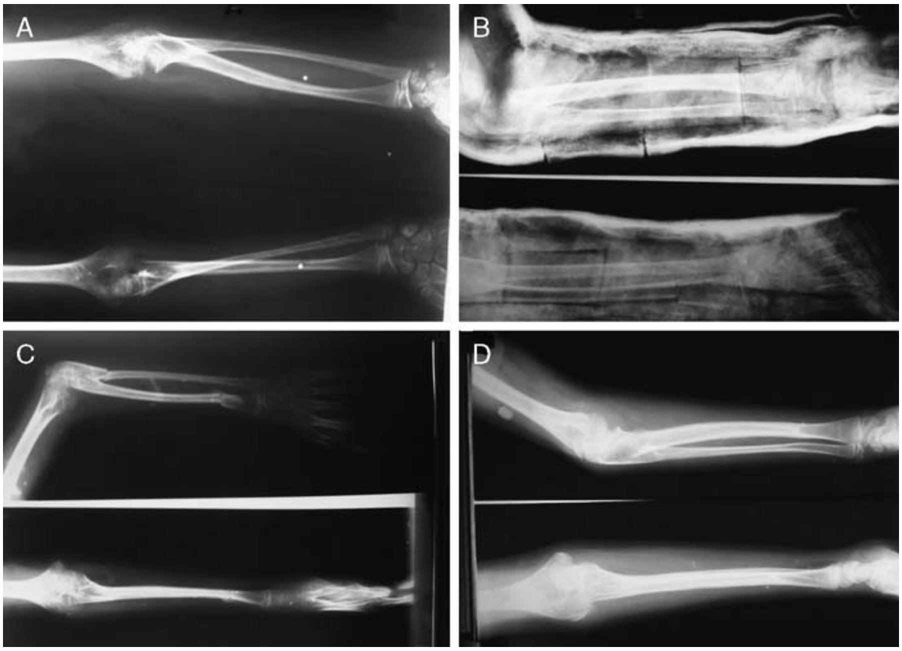

FIGURE 2. A, Presurgery radiograph of the child suggesting proximal radioulnar synostosis. B, Postsurgery radiograph showing osteotomy of the radius and ulna with cast in situ. C, Radiograph after 5 weeks suggesting early union at osteotomy sites. D, Radiograph at subsequent follow-up showing good remolding at osteotomy sites.

Postoperatively, the limb was kept elevated and the patient was observed for signs of neurovascular compromise. The wounds were inspected through the windows in cast and the patient was discharged on the fourth postoperative day.

The cast was removed after 5 weeks. Union at the osteotomy site was confirmed radiographically (Fig. 2). Elbow, wrist, and shoulder mobilization was started. In case of delayed union, the above-elbow cast was reapplied for a further 3 to 4 weeks.

Statistical Methods

Data were analyzed with STATA software (Version 9.0, StataCorp LP, College Station, TX). Before-surgery and after-surgery differences in scores were analyzed with a paired t test and Wilcoxon matched-pair signed-rank test. For categorical variables, differences between proportions were analyzed using the Pearson χ^2 test. For small frequencies in categorical data, the Fisher exact test was applied. A P-value <0.05 was considered statistically significant. Agreement between ≥ 2 criteria for evaluating the degree of impairment in forearm function before and after surgery was assessed with Pearson correlation coefficient.

RESULTS

Data on 30 forearms in 28 operated children who completed >3 years of follow-up were analyzed. Mean age at the time of surgery was 8.6 ± 3.7 years (range 2 to 15 y) in 14 boys and 14 girls. Postoperatively, they were followed up for a mean duration of 54 ± 13 months (range 36 to 84 mo). The mean preoperative pronation

FIGURE 3. A, A 9-year-old girl with bilateral proximal radioulnar synostosis. The presurgery photograph suggests severe restriction of supination for both forearms. She experienced difficulty in taking food to the mouth, holding plates and objects in the open palm, and cleaning the perineal area for hygiene before surgery bilaterally. B, The right forearm was operated upon first and was fixed in 26 degrees of supination. C, After surgery of the right side, with compensatory movements at the shoulder and wrist, almost full supination was possible on the right side. The left side (not operated) shows severe restriction of supination. D, After surgery, with compensatory movements at the shoulder and wrist, almost full pronation was possible on the right side and this is comparable to pronation of the left forearm, which was not operated upon. E, Notice the difficulty in taking food to the mouth on the left side as compared with the right side. F, Notice the difficulty in holding the plate on the left side as compared with the right side. G, After surgery all activities that need pronation of the forearm, including writing, are possible without any difficulty, although the forearm is fixed in 26 degrees of supination.

TABLE 2. Comparison of Results of "Jebsen-Taylor Hand-Function Test" Subtests Before and After Surgery in the Operated Forearms (n = 16) in Boys

Subtest	Dominance	Mean (± SD) Time (s) Before Surgery	Mean (± SD) Time (s) After Surgery	*P*
Writing*	Dominant hand (n = 3)	36.3 ± 29.2	28.3 ± 20.5	0.2529
	Nondominant hand (n = 7)	56.7 ± 11.8	47.7 ± 10.3	0.001
Turning cards	Dominant hand (n = 4)	11.2 ± 3.2	7.2 ± 2.9	0.0109
	Nondominant hand (n = 12)	12.1 ± 3.5	8.2 ± 3.0	0.0001
Picking up small objects	Dominant hand (n = 4)	6.0 ± 1.4	4.7 ± 1.7	0.0154
	Nondominant hand (n = 12)	7.9 ± 1.1	6.1 ± 1.0	0.0001
Simulated feeding	Dominant hand (n = 4)	16.7 ± 3.8	9.2 ± 3.2	0.0154
	Nondominant hand (n = 12)	21.9 ± 9.6	13.4 ± 4.1	0.0006
Stacking checkers	Dominant hand (n = 4)	5.5 ± 1.3	4.0 ± 0.8	0.0138
	Nondominant hand (n = 12)	7.4 ± 4.7	5.2 ± 2.7	0.0038
Large light objects	Dominant hand (n = 4)	5.2 ± 2.2	4.0 ± 1.4	0.2394
	Nondominant hand (n = 12)	6.1 ± 1.7	4.7 ± 1.5	0.0586
Large heavy objects	Dominant hand (n = 4)	5.2 ± 2.2	4.2 ± 1.5	0.0917
	Nondominant hand (n = 12)	6.3 ± 2.1	4.6 ± 1.6	0.0001
Total	Dominant hand (n = 4)	50 ± 13.6	33.5 ± 11.1	0.0079
	Nondominant hand (n = 12)	61.8 ± 20.8	42.3 ± 12.9	0.0001

*Writing was not possible for 6 forearms and hence they were excluded.

deformity was 56.3 ± 13.7 degrees (range 30 to 86 degrees). The mean position of the forearm after surgery was 27.2 ± 4.1 degrees (range 20 to 30 degrees) of supination. There was significant ($P < 0.0001$) association between the degree of pronation (> 30 degrees) and surgical treatment. In the operated group, 93.3% of forearms had pronation > 30 degrees as compared with 23.1% in the nonoperated group. Thus, forearms with > 30 degrees of pronation were positively affected by correction to 20 to 30 degrees of supination.

Preoperative subjective evaluation revealed difficulty in all 30 forearms for taking food to the mouth, holding plates and cups, accepting coins and small objects in an open palm, combing hair, and catching a ball. All 17 nondominant affected forearms had difficulty in cleaning the perineal area for hygiene. Difficulty in dressing, taking the palm to the occiput, writing as well as playing with toys, grasping, and holding was present in 18 (60%), 13 (43.3%), 8 (26.7%), 5 (16.7%), and 3 (10%) forearms, respectively. After surgery, all 30 forearms could perform all the 12 activities (Table 1) without difficulty, as reported by the parents (Fig. 3).

In the objective evaluation using the Jebsen-Taylor hand-function test, no reference range[19] was available for 8 forearms (children less than 6 y) to evaluate the subtest results as normal or abnormal. The mean time for performing all activities in these 8 forearms before surgery was 79.5 ± 19.1 seconds, which significantly reduced to 55.4 ± 14.6 seconds after surgery ($P = 0.0001$). In the remaining 22 forearms (in children aged more than 6 y), the mean time taken to perform all activities before surgery was 47.7 ± 10.0 seconds, which significantly reduced

TABLE 3. Comparison of the Results of "Jebsen-Taylor Hand-Function Test" Subtests Before and After Surgery in Operated Forearms (n = 14) in Girls

Subtest	Dominance	Mean (± SD) Time (s) Before Surgery	Mean (± SD) Time (s) After Surgery	*P*
Writing*	Dominant hand (n = 8)	28.4 ± 15.3	22.1 ± 12.7	0.0016
	Nondominant hand (n = 3)	54.7 ± 5.0	44.7 ± 12.1	0.1383
Turning cards	Dominant hand (n = 9)	11.7 ± 8.0	6.9 ± 4.7	0.005
	Nondominant hand (n = 5)	12.0 ± 3.7	8.2 ± 2.5	0.027
Picking up small objects	Dominant hand (n = 9)	6.7 ± 1.1	5.0 ± 1.6	0.0004
	Nondominant hand (n = 5)	7.4 ± 1.7	5.6 ± 1.3	0.0086
Simulated feeding	Dominant hand (n = 9)	15.2 ± 5.2	10.4 ± 3.8	0.0001
	Nondominant hand (n = 5)	20.8 ± 3.4	14.2 ± 2.6	0.01
Stacking checkers	Dominant hand (n = 9)	5.4 ± 2.4	4.3 ± 1.9	0.0027
	Nondominant hand (n = 5)	6.0 ± 1.0	4.6 ± 0.5	0.0046
Large light objects	Dominant hand (n = 9)	5.8 ± 3.5	4.9 ± 3.6	0.0436
	Nondominant hand (n = 5)	5.6 ± 1.3	4.2 ± 0.8	0.089
Large heavy objects	Dominant hand (n = 9)	5.4 ± 2.8	4.6 ± 3.0	0.0022
	Nondominant hand (n = 5)	6.6 ± 1.3	5.0 ± 1.0	0.0028
Total	Dominant hand (n = 9)	50.2 ± 22.0	36.1 ± 17.7	0.0001
	Nondominant hand (n = 5)	58.4 ± 11.8	41.8 ± 6.8	0.0083

*Writing was not possible for 3 forearms and hence they were excluded.

TABLE 4. Test-Retest Reliability in 30 Operated Forearms with Radioulnar Synostosis as per the Jebsen-Taylor Hand-Function Test

Subtest	Correlation Coefficient (*r*) for Dominant Hand (*P*)	Correlation Coefficient (*r*) for Nondominant Hand (*P*)
Writing	0.9999 (0.0001)	1.0 (0.0001)
Turning cards	0.9445 (0.0001)	0.8646 (0.0001)
Picking up small objects	0.8718 (0.0001)	0.7236 (0.001)
Simulated feeding	0.8526 (0.0002)	0.8435 (0.001)
Stacking checkers	0.9479 (0.0001)	0.9884 (0.0001)
Picking up large light objects	0.9643 (0.0001)	0.9516 (0.0001)
Picking up large heavy objects	0.9686 (0.0001)	0.9286 (0.0001)
Total score	0.9679 (0.0001)	0.9369 (0.0001)

to 33.3 ± 6.6 seconds after surgery ($P = 0.0001$). On preoperative evaluation, picking up small objects was the only subtest that gave a normal score in all forearms. However, the turning cards and simulated feeding subtests gave abnormal results in 20 (90.9%) forearms, stacking checkers in 7 (31.8%) forearms, and picking up large light objects and large heavy objects in 13 (59.1%) forearms. After surgery, all these abnormal tests were converted to normal, except simulated feeding in 2 forearms. Preoperative and postoperative findings differed significantly in male and female subjects (Tables 2 and 3).

The mean number of possible tasks (as described by Morrey and colleagues[20,21]) increased significantly ($P = 0.0001$) from 10 ± 1.3 preoperatively to 15 ± 0 postoperatively. All good (n = 19) and fair (n = 11) results were converted to excellent (n = 30) after surgery, as per the evaluation system described by Failla et al.[20,21]

Test-retest reliability in 30 operated forearms as per the Jebsen-Taylor hand-function test assessed before and after surgery was found to be excellent for both dominant hands (range 0.87 to 0.99) and nondominant hands (range 0.72 to 1.0) (Table 4). A fair correlation ($r = 0.40$, $P = 0.0292$) was observed when the position of the forearm before and after surgery was correlated with the results.

Subjective evaluation of 12 activities revealed that before-surgery and after-surgery reliability was excellent ($r = 1.0$, $P = 0.0001$) and comparable to the results of both the Jebsen-Taylor hand-function test ($r = 0.95$, $P = 0.0001$) and the evaluation system of Failla and colleagues ($r = 1.0$, $P = 0.0001$).

There was no incidence of major complications or neurovascular injury during the intraoperative and postoperative period. There was no loss of correction during cast immobilization. A 15-year-old child had delayed union (9 wk) at the distal radial osteotomy site. The remaining osteotomies united in 5 weeks' time. Another child of the same age had persistent residual dorsal angulation at the radial osteotomy site and had cosmetic concern. No child was seen to be suffering from pain during the follow-up period.

DISCUSSION

The Jebsen-Taylor hand-function test[18,19] is a validated method for evaluating the hand function objectively; however, its reference values are available only for children aged 6 years and above. Morrey and colleague's 15 tasks could be easily performed by a child aged 2 years and above. The classification system of Failla et al[20] not only evaluates the results of posttraumatic radioulnar synostosis but also assesses the function of the forearm and hence was specifically used for the objective evaluation of the forearm function in this study.

High incidence of neurovascular injury was reported by Simmons et al,[3] Hankin et al,[11] and Cleary et al[1] when osteotomies were performed through radioulnar synostosis. However, no neurovascular injuries were found in the present study, as osteotomies were performed at different sites at the radius and ulna, excluding the synostosis site. These findings are comparable to the results of other 2-staged techniques such as drill-assisted osteotomy by Lin et al[10] and double-level rotational osteotomy by El-Adl.[9] The single-staged procedure in the present study produced results similar to 2-staged procedures.

Murase et al[23] and other authors[2,8] have used pins for stabilizing the osteotomy and found loss of correction during cast immobilization. The subjects in the present study did not have any loss of correction during cast immobilization, thereby indicating that immobilization with implants may not be necessary.

Mobilization procedures require the experience of a microvascular surgeon and the application of extensive surgical dissections.[12–14] The Ilizarov method[15,16] and external fixation devices[17] need expensive implants and are associated with a high risk for pin tract infections. These limitations can be easily avoided by the described surgical procedure. Simmons et al[3] advised surgery in the forearm with > 60 degrees of fixed pronation. Evaluations of the present study confirmed that with > 30 degrees of pronation, the forearm function does get affected; however, it is more dependent on the individual's ability of compensatory movements at the shoulder, elbow, and wrist. Hence, surgical indications should be based on functional impairment in addition to a fixed degree of pronation.

The optimum position of the forearm after osteotomy remains controversial.[2] The ideal position depends on the side involved, dominance, and sociocultural environment of the child.[4] The findings of the present study suggested that, for supination activities, a slight supinated position is beneficial; this is consistent with the findings of Ramchandran et al.[2] As our study population has the custom of eating rice with their hand (the dominant hand requiring almost full supination) and cleaning the perineal area with water in the palm (the nondominant hand requiring almost full supination), postoperatively forearms were kept in 20 to 30 degrees of fixed supination rather than in neutral rotation. In all operated forearms, the arc of motion occurred in more functional hand positions (with compensatory movements at the shoulder, elbow, and wrist), with no difficulty in supination as well as pronation postoperatively (Fig. 3).

On the basis of these observations, our current treatment algorithm advocates that forearms with > 30 degrees of pronation and having impaired functions be kept in 20 to 30 degrees of supination after surgery. This may not be universally applicable to all other cultures, as in some countries the neutral or slight pronated position may be more effective[3–5] for using technological devices such as computers and for performing other tabletop activities. The described surgical procedure is single staged, easy, safe, and reproducible. No implants are required.

Both children in the present study who had delayed union and dorsal angulation were 15 years old at surgery. These findings are consistent with those of Dalton et al[24] and Hung[25] and state that higher the age at surgery, more is the chance of having complications. The limitation of the study is the small sample size. The ideal design for formulating the surgical indications would be validation of cutoff scores by a randomized controlled trial with combinations of different subjective and objective scores, and a large sample size, which is not possible because of the rare nature of the disease.

From the study findings, it can be concluded that, for children with congenital radioulnar synostosis and pronation deformity interfering with function and quality of life, the single-staged rotational osteotomy of the radius and ulna is a good alternative procedure.

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