Derotation osteotomy for congenital radioulnar synostosis

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Background
Congenital radioulnar synostosis is a rare condition. It is the result of failure of segmentation between the radius and ulna. It may be isolated or associated with other abnormalities. It may also be found as a part of well-known syndromes. It is bilateral in 60% of the cases.

Patients and methods
This series included five patients with congenital radioulnar synostosis. There were three female and two male patients. The mean age at surgery was 10 years (range 5–22 years). All patients were unilateral. The right forearm was involved in four patients and the left in one patient. One patient had associated ipsilateral symbrachydactyly. The indication for surgery was limitation in performing the activities of daily life regardless of the degree of pronation deformity. They were treated by transverse rotational osteotomy through the synostosis in four cases. In the fifth case, the osteotomy was performed at the diaphysis of the radius because the synostosis was so proximal and short. The osteotomy was fixed by small set dynamic compression plate (DCP) plate in all cases.

Results
The mean time of union of osteotomy was 7 weeks (range 5–12 weeks). The functional results after surgery were satisfactory in all patients.

Conclusion
Rotational osteotomy is a good option to treat the congenital radioulnar synostosis. The preferred position is the neutral as the compensatory movement of the shoulder and wrist will allow proper positioning of the forearm.

Keywords:
congenital radioulnar synostosis, derotational osteotomy, deformity

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Introduction

Congenital radioulnar synostosis is a rare upper limb malformation and was first described by Sandifort's in 1793. The elbow is first identifiable 5 weeks after conception. At this stage, the cartilaginous anlagen of the humerus, radius, and ulna are continuous. Subsequently, longitudinal segmentation produces separation of the distal radius and ulna. However, temporarily, the proximal ends are united and continue to share a common perichondrium. Abnormal genetic or environmental factors operating at this time could interrupt subsequent proximal radioulnar joint morphogenesis [1,2].

Congenital radioulnar synostosis may be isolated or associated with other abnormalities such as brachydactyly, polydactyly, syndactyly, thumb aplasia, and Madelung's deformity. It may also be found as a part of well-known syndromes (arthrogryposis, Apert's syndrome, Carpenter's syndrome, Williams' syndrome) and chromosomal abnormalities (Klinefelter's syndrome). Both sexes are affected with 3 : 2 male to female ratio. It is bilateral in 60% of the cases [3].

Different classifications of radioulnar synostosis are present. Cleary and Omer [4] proposed four radiographic types:

(1) Type I: fibrous synostosis with a reduced and normal-appearing radial head.
(2) Type II: osseous synostosis with normal radius.
(3) Type III: osseous synostosis with a hypoplastic and posteriorly dislocated head.
(4) Type IV: a short osseous synostosis with an anteriorly dislocated radial.

The patients with congenital radioulnar synostosis have a fixed forearm pronation deformity. In cases with severe deformity, performing activities of daily living is difficult. Generally, there are two surgical options to treat such patients. One is the mobilization operation to separate the radioulnar synostosis to restore forearm rotation. The results of mobilization operation are disappointing with high rate of recurrent fusion [5–7]. Recently, a free vascularized fascial flap placed between the separated forearm bones has been reported to successfully block postoperative recurrence of the synostosis [8,9]. The second surgical option is osteotomy to realign the forearm in a position suitable for performing the activities of daily living. There are many types of rotational osteotomy that can be summarized into osteotomy at the synostosis, osteotomy at two sites in the diaphysis of the radius and the ulna, and osteotomy at one site in the distal diaphysis of the radius. The generally accepted surgical

treatment is derotation osteotomy using Kirschner wires (K-wires), plate, external fixator, or even a cast to fix the osteotomy [3,10–14].

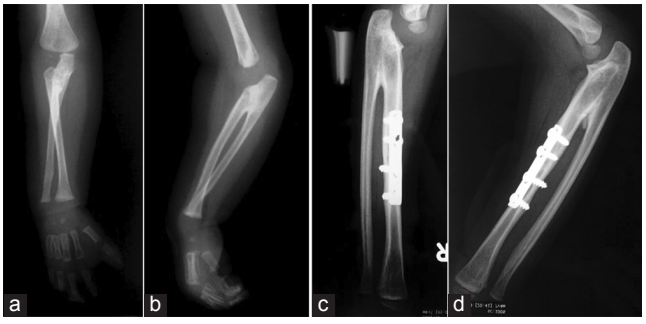
Patients and methods

Between 2004 and 2009, five patients with congenital radioulnar synostosis were managed with derotation osteotomy. There were three female and two male patients. The mean age at surgery was 10 years (range 5–22 years). All patients were unilateral. The right forearm was involved in four patients and the left in one patient. One patient had associated ipsilateral symbrachydactyly. The indication for surgery was limitation in performing the activities of daily life regardless of the degree of pronation deformity (Table 1).

Preoperative assessment was performed for all patients, including both clinical and radiological assessment. The clinical assessment included the ability of performing the activities of daily life, the degree of pronation deformity, and the range of motion of the elbow and wrist. The radiographic assessment was based on the classification of Cleary and Omer [4].

Preoperative assessment of the patients revealed that all of them had limitation of the activities of daily life. All patients were type III according to Cleary and Omer [4], with visible osseous synostosis with a hypoplastic and posteriorly dislocated head.

Figure 1



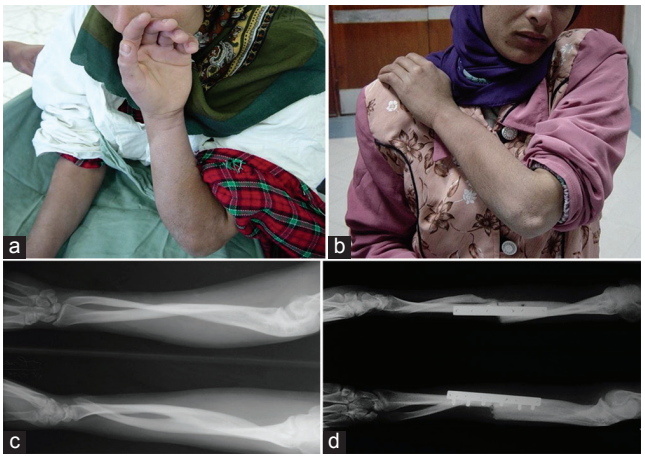
Male patient, 5-year-old with right congenital radioulnar and ipsilateral symbrachydactyly synostosis. (a) Preoperative radiographs. (b) Postoperative radiographs.

Surgery was performed under general anesthesia with the patient in the supine position under tourniquet control. The posterior Thomspon's approach was used to expose the synostosis and the radius. The osteotomy was performed at the synostosis in four cases. In the fifth case, the site of synostosis was so proximal and short; hence, the osteotomy was performed at diaphysis of the radius (Fig. 1). Fixation was performed by small set DCP plate in all cases. The position of derotation osteotomy was neutral in four patients and 25° pronation in one patient. Postoperatively, close observation for neurovascular complications was performed in the first 24 h. In children less than 10 years, an above elbow plaster cast was applied for protection for 4 weeks. The stitches were removed after 2 weeks. The patients were followed up after that every month to check the union.

Results

Bone union after osteotomy was achieved in all patients. The mean time of union was 7 weeks (range 5–12 weeks). The mean postoperative follow-up period was 23 months (range 8–52 months). In patient with severe pronation deformity (180°), acute correction was achieved intraoperatively to reach the final position of 25° pronation (Fig. 2). The mean correction achieved

Figure 2



Female patient, 22-year-old with left congenital radioulnar synostosis. (a) Preoperative photo of the patient. (b) Postoperative photo of the patient. (c) Preoperative radiograph. (d) Postoperative radiographs.

Table 1 Preoperative and postoperative data of the patients

Cases	Sex	Age (years)	Affected side	Associated anomalies	Preoperative pronation deformity (deg.)	Postoperative position	Time of union (weeks)	Follow-up duration (months)
1	Female	22	Left	None	>90 (180)	25° pronation	12	52
2	Male	5	Right	Symbrachyductyly	90	Neutral	5	23
3	Male	8	Right	None	70	Neutral	6	18
4	Female	7	Right	None	60	Neutral	6	14
5	Female	10	Right	None	80	Neutral	6	8

in the other four patients was 75° (range 60–90°). All patients were satisfied and showed improvements in the ability to perform the activity of daily life. There were no neurological or vascular complications even in the patient with severe acute correction of 155°.

Discussion

Congenital radioulnar synostosis is a rare anomaly of the upper limb. It may cause significant disability when the deformity is severe or bilateral. In this study, surgery was performed at the age of first presentation and the mean age was 10 years (range 5–22 years). With respect to the proper age for surgery, most authors recommended that the operation is best carried out between the age of 3 and 8 years [3,10–12]. At these ages, the osteotomy will be easy with sufficient remodeling of the radius and ulna. In addition, the fixation of osteotomy could be accomplished by K-wire and cast or even a cast alone. In older patients, the risk for neurovascular complications may increase.

In these cases, the indication for surgery was limitation in performing the activities of daily life regardless of the degree of pronation deformity. Simmones *et al*. [2] considered that pronation deformity of 60° or more was a definite indication for derotation osteotomy. Yammine *et al.* [15] proposed two main indications for surgery, which were hyperpronation more than 90° and bilateral synostosis. Ramachandran *et al.* [12] reported that a pronation deformity of 60° or more was the indication for osteotomy in unilateral cases and less than that in bilateral. Hung [11] showed that the indication for osteotomy was pronation deformity more than 65°.

Generally, there are two surgical options to treat congenital radioulnar synostosis. The first option is to separate the radioulnar synostosis to restore forearm rotation, but the reports of other authors are disappointing with high rate of recurrent synostosis [5–7]. The second surgical option is derotation osteotomy to realign the forearm in a suitable position.

In this study, the osteotomy was performed through the synostosis in four cases. The procedure was accomplished through a single incision. In the fifth case, the synostosis was so proximal and short; hence, the osteotomy was performed through the diaphysis of the radius. All osteotomies were fixed by plate and screws to ensure adequate degree of intraoperative correction and to avoid postoperative loss of this correction. None of our patients showed loss of correction during postoperative follow-up.

Yammine *et al.* [15] performed reed radial osteotomy and transverse ulnar osteotomy in six patients. Fixation was performed by a plate in three cases, external fixator in two, and by mini-orthofix in one patient. None of their patients showed loss of correction postoperatively [15]. Murase *et al.* [13] described osteotomies in the distal third of the radius and the proximal third of the ulna through two separate incisions in four patients. The osteotomies were fixed by K-wire. They reported a 20° loss of correction during immobilization in a cast in one patient [13]. Hung [11] described osteotomies in the distal third of the ulna and the proximal third of the radius through two separate incisions in 39 patients. The osteotomies were fixed by K-wire. There was loss of correction during cast immobilization in five patients [11]. Hence, proper fixation of the osteotomy is essential to maintain the correction postoperatively.

In this study, all patients were unilateral. The position of derotation osteotomy was neutral in four patients and 25° pronation in one patient. The best position of the forearm after derotation osteotomy is controversial.

Murase *et al.* [13] and Hung [11] preferred to correct the forearm position in the dominant hand between 0 and 30° of pronation and neutral position was preferred for nondominant hand in a unilateral case, as well as bilateral case. This is in agreement with our opinion in this series.

Green and Mital [16] recommended that, for bilateral cases, the dominant hand should be placed 20–35° of supination and the nondominant hand in 30–45° of pronation. In unilateral cases, the ideal position was 10–20° of supination [16]. However, Ogino and Hikino [17] recommended that, for bilateral cases, the dominant hand should be placed 0–20° of pronation and the nondominant hand in 0–20° of supination. In unilateral cases, the forearm position was 0–20° of supination [17]. Ramachandran and colleagues preferred a position of 10° supination in all cases as the compensatory movement of the shoulder and wrist will allow proper positioning of the forearm.

In this study, there were no complications. The reported complications in other series included loss of correction, vascular complications such as Volkmanns' ischemia, neural complications such as posterior interosseous palsy, and delayed union [2,11,13,15–17].

Conclusion

Congenital radioulnar synostosis is a rare anomaly of the upper limb. Derotation osteotomy is a good option to manage the deformity. The indication for surgery was limitation in performing the activities of daily life

regardless of the degree of pronation deformity. There is no consensus about the optimal angle of forearm rotation. The preferred position is the neutral as the compensatory movement of the shoulder and wrist will allow proper positioning of the forearm.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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