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Congenital radio ulnar synostosis, analysis of functional outcome using double rotation osteotomy and osteotomy at synostosis site

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Abstract

Aim: To evaluate the functional outcome using double rotation osteotomy and osteotomy at synostosis site in congenital radio ulnar synostosis

Materials and methods: A prospective study has been done in Institute of child health and hospital for children, Egmore, Chennai for congenital radio ulnar synostosis. There were 11 females and 9 males of which 7 were bilateral CRUS. Cases were classified using cleary omer classification and pre op deformity measured and operated. Mean pre op hyperpronation deformity was 72° (range 55° to 85°)

Results: All the children showed appreciable improvement and their parents were happy about the outcome achieved and improvement in day to day activities. Mean post-operative supination achieved is 20° (range 12° to 35°).

Conclusion: the need for surgical management present in congenital radio ulnar synostosis in specific indications like bilateral hyperpronated deformity with restriction of supination which is not compensated by shoulder. So achieving 20°-25° of supination is our aim. This can be achieved by gradual osteotomies. To start with Initial osteotomy at the synostosis site, If the required supination is not achieved second osteotomy can be done at distal 1/3rd shaft of radius, Double osteotomy at both radius and ulna can be reserved as a choice for older children with bilateral hyperpronation deformity

Keywords: Radio ulnar synostosis, double rotation osteotomy, hyperpronation deformity

Introduction

Congenital proximal radioulnar synostosis is often severely disabling, particularly if it's bilateral or if severe hyperpronation exists [1, 2]. Functionally, patients with severe deformity will find difficulty in doing day to day activities in obtaining a cup to the mouth, difficulty holding or using small objects, inability to dress themselves, difficulty feeding themselves, inability to accept small objects in their hand and difficulty in typing in later part of life.

In an extended elbow supination is not compensated adequately by shoulder like pronation. So in hyperpronated forearm of CRUS needs position correction in supination to achieve some useful hand function. This can be achieved by osteotomy at the synostosis site/osteotomy at the synostosis site and distal shaft of radius/double osteotomy at radius and ulna (if pronation is very severe) [1, 2, 3]

Materials and Methods

Of 34 patients with radioulnar synostosis that we found during the period January 2012 to April 2017 in Intitute of Child Health and Hospital for Children, Egmore, only 20 patients met the criteria for operation. Of which 7 patients had bilateral synostosis. The mean age at presentation was 4 years for operated patients (1-7 years). The most common complaint of patients was limitation of range of movements in elbow and cosmetic. We had 11 female and 9 male patients operated.

All patients were included in the study after getting written informed consent and appropriate ethical committee clearance.

We used Green and Mital methods who suggested that a longitudinal K-wire in ulna and the radius is fixed based upon the need and stability of the osteotomy site either by k wire or external fixator. If radius osteotomy is stable it is left with cast immobilization [18].

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Results

All patients were satisfied with their extremity and their functions improved considerably. The mean age at the time of presentation for surgery is 3.8 years (range 2-9 years).mean postoperative follow up period 2.2 years (range 7 months to 4 years). The mean postoperative position of operated limb was 22° of supination (range 12° to 35°). The results of the operated cases are summarized in table.

We used Green & Mital criteria who advised 20° of

supination post operatively [18]. From that table we could see out of 20 children who underwent surgery, 17 children got good range of supination and were happy about the outcome improvement. In 3 children we could not achieve supination of 20° as per Green & Mital, but we have improved the hyperpronation deformity compared to preoperative deformity. And even those three children could do their daily day to day activities independently as expected of their age.

S.no	Age/ sex	Duration of follow up	Pre op pronation deformity	Post op elbow rom	Final position of forearm	Cosmetic improvement	Functional improvement
1.	3/F	3 years	60°	Good	Supination	Yes	Yes
2.	4/F	1 year	85°	Good	Supination	Yes	Yes
3.	2/M	8 months	80°	Acceptable	Mid Prone	No	Yes
4.	3/M	1 year	70°	Acceptable	Mid Prone	No	Yes
5.	2/F	2 years	85°	Good	Supination	Yes	Yes
6.	3/M	18 months	60°	Good	Supination	Yes	Yes
7.	3/F	10 months	60°	Good	Supination	No	Yes
8.	2/F	7 months	80°	Acceptable	Mid Prone	No	Yes
9.	3/F	2 years	65°	Good	Supination	Yes	Yes
10.	3/F	15 months	70°	Good	Supination	Yes	Yes
11.	4/M	30 months	80°	Good	Supination	Yes	Yes
12.	2/F	2 years	85°	Good	Supination	Yes	Yes
13.	3/F	2 years	60°	Good	Supination	Yes	Yes
14.	9/M	5 years	70°	Good	Supination	Yes	Yes
15.	9/F	3 years	75°	Good	Supination	Yes	Yes
16.	8/F	4 years	80°	Good	Supination	Yes	Yes
17.	3/M	3 years	80°	Good	Supination	Yes	Yes
18.	3/F	2 years	70°	Good	Supination	Yes	Yes
19.	4/M	3 years	75°	Good	Supination	Yes	Yes
20.	3/M	42 months	55°	Good	Supination	Yes	Yes

Discussion

Congenital radioulnar synostosis, a deformity characterized by a fixed position of the forearm ranging from neutral rotation to fixed pronation deformity, is a very rare anomaly [1, 2].

Embryologically it is due to defect in the longitudinal segmentation of upper limb bud. Upper limb bud which forms the upper limb bones and muscles usually arises at 26 days of intra uterine life and growth and differentiation continue until 46 days of age. Longitudinal segmentation of upper limb bud produces separation of the radius and ulna. For some time the proximal part of radius and ulna are united and both share a common perichondrium. Abnormal genetic factors or teratogenic factors influencing the upper limb bud at this time would interfere with proximal radioulnar joint morphogenesis [6, 7].

Only 350 cases have been reported in literature heretofore. The condition will be disabling particularly once it happens bilaterally or if there's severe hyperpronation where supination is needed to do normal day to day activities.

Congenital radioulnar synostosis is also presented as associated with congenital syndromes or as an isolated anomaly

Wilkie classified the synostosis roentgenographically into two kinds:

Type 1: Proximal or true radioulnar synostosis (the radius and ulna are uniformly fused at their proximal borders for a variable distance.)

Type 2: Radioulnar synostosis associated with congenital dislocation of the head of the radius and the fusion is just distal to the proximal radial epiphysis [9].

Cleary and Omer explained four types

Type I: synostosis does not involve bone, associated with reduced radial head.

Type II: visible bony synostosis, associated with normal reduced radial head.

Type III: visible bony synostosis with radial head is posteriorly dislocated and hypo plastic.

Type IV: short bony synostosis with radial head is anteriorly dislocated and mushroom shaped [10].



Fig 1: Bilateral radioulnar synostosis



Fig 2: Child comfortable in doing day to day activities with pronation deformity of 50°

Surgery indications are hyperpronation and synostosis in both limbs, in both of them disability must be severe. The seriousness of hyperpronation that requires surgery is dubious however as we would like to think it must be decided on individual case to case basis and the significant concern is the function of the limb.

As authors would see it, patients with inborn radioulnar synostosis who doesn't have extreme disability and functional impairment require no surgical treatment. As of late, a few surgeons introduce new techniques for manipulation and mobilization of the forearm, yet long term outcome have not been studied clearly.

Surgeries that have been proposed for congenital radioulnar synostosis that can be arranged in 2 noteworthy categories: 1) operations that were intended to reestablish the rotational movement of forearm (i.e. supination and pronation)

associated with correction of synostosis, 2) operations that enhance the settled position of forearm in a more useful position [1, 2, 3].

Some surgeons have revealed osteotomy of the synostosis and interposition of fat or muscle or silicone, yet their outcomes have not been successful what's more, repeat of the ankylosis has been the major drawback. Hansen and Andersen played out a partial resection of the synostosis in a sixteen-year-old young lady. Eighteen month postoperatively, rigid union was noted radiologically at surgical site [2, 3, 4, 5, 6].

Miura *et al.* worked in eight forearms points in seven patients. They put anconeus between the isolated radius and ulna, yet bony union repeated in the entire patient. [15] Kelikian and Doumanian announced great outcomes with utilization of a swivel prosthesis in post-traumatic proximal radioulnar synostosis patients; [16] but, Tachdjian noted bad outcomes with the swivel prosthesis in patients who had congenital radio ulnar synostosis, with repeat of the ankylosis at eighteen months [17].

Kanaya announced that separation of congenital proximal radioulnar synostosis along with interposition of vascularised fascio-fat graft and osteotomy of the radius could accomplish pronation and supination of the forearm [19].

Right now, osteotomy to accomplish more useful position is acknowledged for treating the patients who have severe pronation deformity. The perfect position relies upon the type of involvement in the patient (both forearms/one-sided, dominant and nondominant hand), the socio-cultural environment of the patient, and anticipated future activities of the patient. Since it is impractical to anticipate patient's future activities, it is difficult to decide the ideal position of the forearm.

Various suggestions for ideal position are given in table

Authors	Recommendation	Comments
Green & Mital [18]	20 degrees of supination	20-35 degrees for the dominant hand in bilateral cases
Simmons and Waters [12]	20 degrees of pronation	Neutral for the nondominant hand in bilateral cases
Ogino and Hikino [14]	Neutral to 20 degrees of supination for unilateral or nondominant hand in bilateral cases	Allows the use of chopsticks
Hung [8]	70-100% pronation	
Tsujino [11]	Neutral to 30 degrees of pronation for the dominant hand, neutral for the nondominant hand	
Ramakrishnan <i>et al.</i> [13]	10 degrees of supination for all	Compensatory movements of the shoulder and wrist allow most activities of daily living

We worked on both unilateral and bilateral congenital radio ulnar synostosis. We wanted to convey their forearm to 15-25 degree supination after osteotomy. We did osteotomy at the radio ulnar fusion mass for 10 patients and osteotomy distal to the fusion mass (double rotation osteotomy) for 4 patients and for patients we did osteotomy at both radius and ulna [7, 8, 14, 17, 18].

Osteotomies distal to fusion mass have a tendency to have more soft tissue restrictions. Continuous redress utilizing an Ilizarov frame diminishes the danger of neurovascular injuries and enables the patient to choose the most useful position. Utilizing a little external fixator permits exact rotational alignment manages satisfactory adjustment yet stays away from cast immobilization



Fig 3: Pre op



Fig 4: Immediate post op



Fig 5: 10 wks post op Double rotation osteotomy



Fig 6: Pre op



Fig 7: Post op-osteotomy at fusion site



Fig 8, 9, 10: Post operatively pronation deformity corrected using osteotomy at fusion site

A few surgeons suggest that osteotomy is best to do between the ages of 4 and 10 years, yet we prescribe the osteotomy should be as early as possible to mitigate the neuro vascular injuries and soft tissue complications [20, 21].

Conclusion

Complications of both type of osteotomy are

1. Neurovascular injury
2. Compartment syndrome
3. Surgical site infection
4. Bony union at osteotomy site and loss of derotation
5. Reduced range of movements at elbow joint

All these all rare complications. We encountered surgical site infection in one case and it was treated accordingly. Both type of osteotomies yield comparative results.

To conclude the need for surgical management present in congenital radio ulnar synostosis in specific indications like bilateral hyperpronated deformity with restriction of supination this is not compensated by shoulder. So achieving 20°-25° of supination is our aim. This can be achieved by gradual osteotomies. To start with

1. Initial osteotomy at the synostosis site
2. If required supination is not achieved second osteotomy can be done at distal 1/3rd shaft of radius,
3. Double osteotomy at both radius and ulna can be reserved as a choice for older children with bilateral hyperpronation deformity

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