

## Congenital Radioulnar Synostosis Treated Using a Microvascular Free Fascio-fat Flap

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Congenital radioulnar synostosis, which is caused by fusion between the proximal end of the radius and ulna, is an uncommon deformity of the upper extremity. It is characterized by the forearm being fixed in some degree of pronation and patients present with a variety of functional limitations of the involved limb. This condition may lead to difficulties in daily activities, such as writing, eating, and accepting objects in an open palm. Many operative procedures have been developed to mobilize the fixed forearm but the long-term results are disappointing. Herein, we reported a 12-year-old female patient with congenital radioulnar synostosis of the bilateral forearm who was treated with separation of the bony bridge between the left radius and ulna combined with a free groin flap transfer into the separated space. The functional results are good and no recurrent ankylosis occurred during a period of 28 months postoperatively. The flap size and volume of the subcutaneous fat are the key factors for satisfactory results. (*Chang Gung Med J* 2005;28:117-22)

**Key words:** congenital radioulnar synostosis, free fascio-fat flap.

Congenital radioulnar synostosis is a rare malformation that affects the elbow and forearm. It is caused by the fusion of the proximal radius and the ulna with the presentation of forearm fixed in some degrees of pronation. Loss of rotation of the forearm substantially decreases the functions of the upper limb and can have profound restriction on patient's daily activities such as eating, writing, washing, and dressing. The first anatomical description was given by Sandifort in 1793,<sup>(1)</sup> and Morrison also reported one case 100 years later, in which the classic physical findings were described.<sup>(2)</sup> Many operative methods have been reported including simple excision of the bony fusion,<sup>(1)</sup> taking down the synostosis with or without interposition of soft tissue,<sup>(1,3,4)</sup> the Kelikian/Doumanian swivel method, derotational osteotomy distal to synostosis site,<sup>(5,7)</sup> derotational osteotomy through the synostosis mass,<sup>(8)</sup> and derotational

osteotomy with the Ilizarov method.<sup>(9)</sup> The ideal treatment for radioulnar synostosis is to restore the forearm rotation and to prevent recurrence of the bony bridge after correction. In a review of reports of operative procedures and their outcomes, the long-term follow-up results and associated complications had been disappointing,<sup>(1,5,7,10-12)</sup> until a new mobilization technique using free vascularized fascio-fat graft interposition combined with radial osteotomy was developed by Kanaya in 1998. This method provided good results with an average supination 26 degrees (range, 10 to 45 degrees) and pronation 45 degrees (range, 10 to 80 degrees). In addition, none of the patients had recurrent ankylosis during the 4 years of follow up.<sup>(10)</sup>

We present a case of bilateral congenital radioulnar synostosis who was treated with separation of the synostosis combined with a free groin flap

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transfer to fill the space created and discuss the outcome.

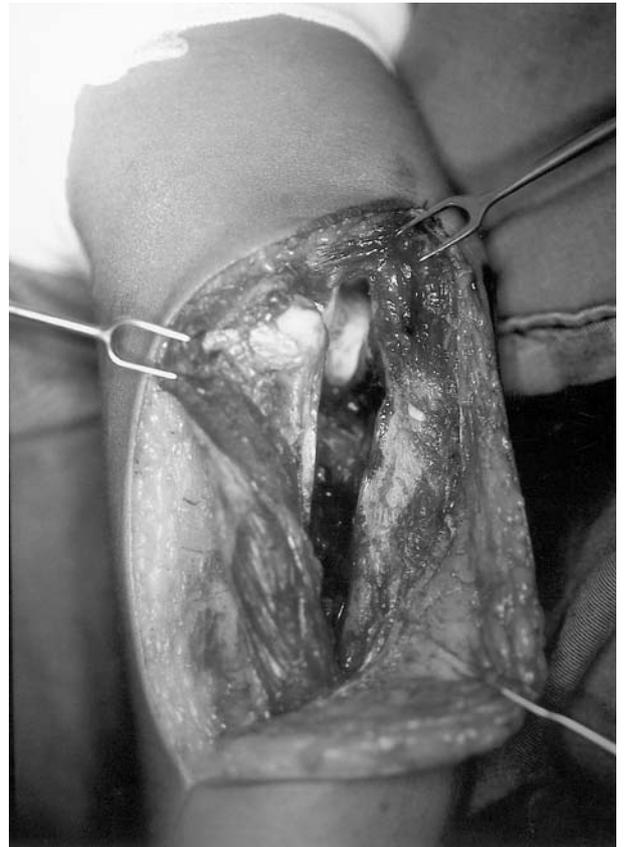
### CASE REPORT

A 12-year-old right-handed girl presented with a fixed pronation deformity of bilateral forearms caused by congenital proximal radioulnar synostosis. The patient complained of disability in her daily life such as washing, dressing, eating with chopsticks and holding a bowl into the palm. The left forearm was also 4 cm shorter than the right forearm. This case was classified as type II according to Cleary and Omer, with a visible osseous synostosis with a normal radius appearing on X-ray film (Fig. 1). Preoperatively, the left forearm was fixed in 90 degrees of pronation and the right forearm was 70 degrees of pronation. The synostosis of the left side was exposed anteriorly. After both bones were exposed subperiosteally, periarticular osteotomy,



**Fig. 1** Preoperative radiographs show a visible osseous synostosis with a normal radius.

trimming of the radial head and ulnar to be flat, and extensive release of interosseus membrane were performed (Fig. 2). After separation of the synostosis, adequate rotation was achieved without difficulty at both the distal radioulnar joint and the osteotomy site. A right groin flap was elevated. Deepithelization was performed and only a small area of skin ( $1.5 \times 3$  cm) was preserved as the monitor. The deepithelized portion was inserted into the separated space between the radius and ulna. The recipient artery was the right radial artery, and the cephalic vein was chosen as the recipient vein. All of the procedures were followed by immobilization using a long arm splint. Active supination and pronation achieved 70 and 50 degrees respectively in the operated forearm but the non-operated side still showed a fixed 70 degrees of pronation after a follow-up of 28 months (Figs. 3,4). No recurrent anky-



**Fig. 2** After periarticular osteotomy, trimming of the radial head and ulna to be flat, and extensive release of interosseus membrane, a space was created.

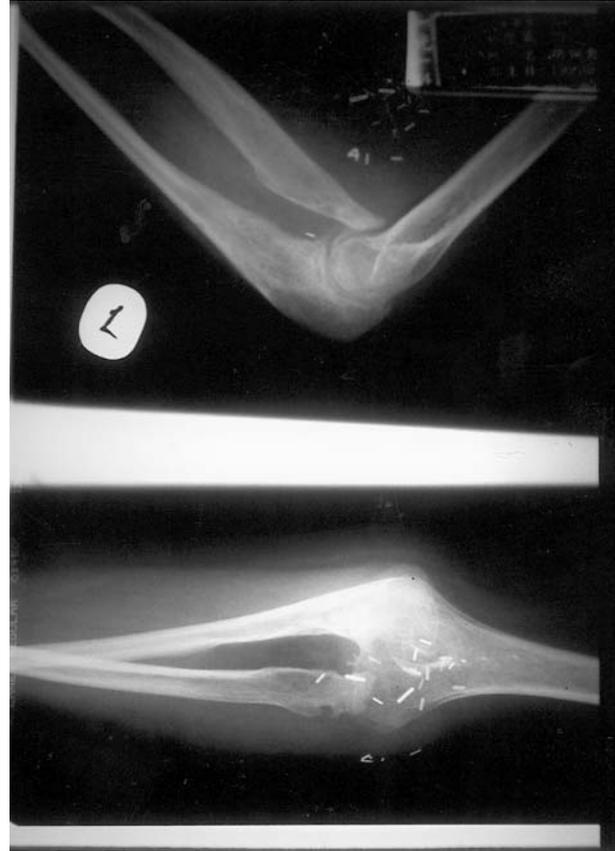


**Fig 3** 28 months after operation, the active pronation of the left forearm achieved 50 degrees.



**Fig. 4** The active supination achieved 70 degrees.

losis was found on the follow-up X- ray films (Fig. 5). For this patient, we also performed the modified Darrach procedure to release the distal radioulnar joint 7 months after the first operation because of the limited motion of wrist joint. Now, holding a bowl and accepting objects into an open palm are greatly improved on the left side.



**Fig. 5** Follow-up X-ray film showed no recurrent ankylosis.

## DISCUSSION

Congenital radioulnar synostosis is a rare problem and is thought to be a failure of longitudinal segmentation between the radius and ulna at the 7<sup>th</sup> week of development.<sup>(13)</sup> Because it is due to a defect in utero, it may be associated with some musculoskeletal anomalies and congenital syndromes such as polydactyly, syndactyly, Madelung's deformity, carpal coalition, thumb aplasia, Apert's syndrome, Carpenter's syndrome, arthrogryposis, mandibulofacial dysostosis, and William's syndrome.<sup>(14,15)</sup> This condition occurs equally in males and females with bilateral involvement in 60% to 80% of patients. In addition, positive family history has also been reported.<sup>(16)</sup> Because of the fusion of the proximal radioulnar joint, patients always presented with a fixed degree of pronation and the degree of fixed forearm pronation determines the disability and age of presentation. Although it is present at birth, it usually is

not found until early adolescence, when the patients have difficulty with grasping, holding, eating, and accepting object with an open palm.

Several classification systems based on the radiological findings have been proposed. Cleary and Omer described four types. Type I is a fibrous synostosis with a normal and reduced radial head. Type II is a visible osseous synostosis with a normal radius. In type III, a bony synostosis exists with the radial head hypoplasia and dislocated posteriorly. In type IV, the radial head is dislocated anteriorly.<sup>(17)</sup> However, there is little clinical value in this classification because the operation depends on the severity of the functional deficit and whether it is bilateral or not.

The indication for operation for treatment of congenital radioulnar synostosis is controversial. Cleary and Omer believe that the operative intervention is rarely indicated and their surgical indication is based more on functional deficits than absolute forearm position.<sup>(1)</sup> On the other hand, Simmons and Southmayd concluded that an operation is indicated in a patient with greater than 60 degrees of fixed pronation and must be individualized in patients with degrees of pronation between 15 to 60 degrees.<sup>(18)</sup> Ogino and Hikino also reported that patients with greater than 60 degrees of pronation had restrictions in daily activities.<sup>(19)</sup> Generally, surgical intervention is dependent on functional limitations, type of involvement (bilateral or unilateral, or nondominant side), social and cultural environment of the patient, and projected future activities.<sup>(10)</sup>

The goals of the operations are to restore forearm rotation and to prevent recurrent ankylosis. Many procedures have been reported but the results seem unsatisfactory. The procedures have included simple excision, resection of the synostosis with or without flap interposition, and corrective osteotomy to restore the forearm function. In most of reports, we found that either lack of long-term results or the initial results might be satisfactory but the range of motion would return to the preoperative state with time. Most recently, Kanaya and Ibaraki reported mobilization of congenital proximal radioulnar synostosis using a free vascularized fascio-fat graft.<sup>(10)</sup> In their series, all seven patients received separation of synostosis, osteotomy of radius, and free lateral arm flap to fill in the space between the separated radius and ulna. After operation, all patients had their daily

functions restored and none of the patients had recurrence. In 2003, Murase et al. demonstrated derotational osteotomy at the shafts of radius and ulna for congenital radioulnar synostosis.<sup>(6)</sup> Four patients were included and all patients' abilities to perform daily activities showed a marked improvement over a period of 45.8 months except for a 20 degree loss of correction during cast immobilization in one case.

Our patient was type II and our operative procedures were similar to the method of Kanaya and Ibaraki. The only difference was the use of the free groin flap as the spacer. Compared with the lateral arm flap, the groin flap has more abundant fat tissue and the donor site scar can be hidden within the bikini line, especially for the female patients. In addition, the two teams approach saves operation time. However, concern regarding the variable vascular origin and inconsistent caliber of vascular pedicle has made it more difficult to elevate this flap. Using the rule of two finger widths, easy and rapid intraoperative flap design is possible. Additional terminal vascular diameter can be obtained by harvesting a wedge or cuff of the femoral artery and vein with the vascular pedicle.<sup>(20)</sup> We believe that the use of free fascio-cutaneous flap is better than pedicled or local flap and the volume and size of flap are the key factors to avoiding scarring and recurrent bony bridge formation. For most patients with congenital radioulnar synostosis, the local fascial tissue is short and their fibers are abnormally directed. In addition, the interosseous membrane is narrow, and the supinator muscle may be abnormal or absent. This could explain why the results have been unsatisfactory in the previous reports, in which the local or distally based pedicle flap was used. Although it is present at birth, the patient may receive operation during their adolescence and the possibility of stiffness of distal radioulnar joint increases with age. Release procedures for stiffness of the distal radioulnar joint might be performed during the second stage; and the later the operation is performed, the better it is to combine with the release procedure.

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## 以自由筋膜脂肪皮瓣治療先天性橈尺骨性接合

高煌凱 陳宏基 陳煥棠

先天性橈尺骨性接合 (congenital radioulnar synostosis) 為一罕見的上肢畸型,其成因是由橈、尺骨在近端融合所造成。其特徵是前臂固定於不等角度的旋前 (pronation) 動作,受影響的上肢會有不等程度的功能性限制。這將會對患者在寫字、吃飯、打開手掌受物等日常活動造成影響。有許多術式來解決此一問題,但長期追蹤的結果通常令人失望。一個12歲雙側先天性橈尺骨性接合的小孩,接受左側先天性橈尺骨性接合處分隔手術合併自由腹股溝皮瓣轉移至分隔的空間,術後經28個月的追蹤,其功能性的結果令人滿意且無復發。我們認為皮瓣的大小與皮下脂肪的容積為獲得令人滿意結果的關鍵。(長庚醫誌 2005;28:117-22)

**關鍵字：**先天性橈尺骨性接合，自由筋膜脂肪皮瓣。

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