CASE REPORT

Early surgical anterior release for congenital and isolated elbow contracture in flexion: A case report of a 16-month-old child

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Summary
Isolated congenital elbow contracture is a rare upper-extremity disorder and there are few data about management of this condition. Authors report their experience after aggressive management of children with isolated congenital elbow contracture in flexion. Because of total absence of range of motion (ROM) improvement despite physical therapy (ROM 90°–120°) and bone deformity, an anterior surgical release of the elbow was performed through an extensive lateral approach, at sixteen months of age. After surgery, this child was treated by three casts at maximal gained extension followed by sequential Turnbuckles splints. After five years of follow-up, the result was excellent with ROM 5°–135°, normal function and absence of growth disturbance. The limiting factor of this protocol was excessive traction in elbow extension on the neurovascular structures, especially the radial nerve. This treatment represents an aggressive management with multiple general anaesthesia, but was found to be a valid option.

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Introduction

Congenital elbow contracture is a rare upper-extremity disorder. Arthrogryposis is the main etiology of congenital elbow stiffness. It can represent a severe disability and surgical treatment is often used to treat extension contracture but rarely used for flexion contracture [1]. The timing for arthrogryposis elbow surgery is still controversial [2,3]. The results of surgical release in elbow joint contracture in children are mainly reported for traumatic stiffness [4–8]. To our knowledge, there are no data about management of isolated congenital elbow contracture in flexion. This study reports the result of aggressive management of a child with isolated congenital elbow contracture in flexion treated by early surgical anterior release and repeat casting series.

Case report

Hani was the third boy of the family; he was born after a normal delivery. There was no history of trauma or
infection. No family pathologic history was related but the parents have a between blood relation (cousin). They noticed a painless contracture ten days after Hani’s birth by tears during left elbow mobilisation. The boy presented a severe restriction in extension of the left elbow with a range of motion (ROM) of 0–90–120° but normal range of pronosupination (0–0–170° ). Radiographs showed a distal diaphyso-metaphyseal incurvation but no congenital proximal radio-ulnar synostosis, no heterotopic calcifications, and no radial head dislocation were detected (Fig. 1).

Because of failure of physical therapy and splinting, the baby was referenced in our institution at 8 months old (Fig. 2). Others articulations were normal, no muscular disease was detected, and no hypotony or hyperlaxity were found. Genetic assessment did not conduct to any diagnosis or arthrogryposis syndrom.

Magnetic resonance imaging (MRI) was performed and detected a congenital hypoplastic anterior articular capsule with normality of joint surfaces and of epiphyseal structures and the presence of olecrania fossa (Fig. 3). EMG exploration of triceps did not relate abnormality of contraction.

An elbow examination and an arthrography were performed under general anaesthesia. It showed no amelioration of the ROM after muscular decontraction by manual mobilisation, no incongruity of the joint surface between the humerus and the ulna. An absence of anterior articular capsular recessus was confirmed (Fig. 1D).

Because of total absence of ROM improvement despite physical therapy, and bone deformity on X-rays, an anterior surgical release of the elbow was performed at sixteen months of age.

A lateral approach along the lateral supracondylar ridge of the humerus was used under general anaesthesia without tourniquet. The brachioradialis and the extensor carpi radialis longus were mobilised off the humerus allowing exposure of the anterior capsule. No clear anterior articular capsule was identified and strong fibrous adherences were noticed on chondroepiphysis. All adhesions were extensively released with extraperiosted approach and the dissection was carried out as far as the medial side of the joint. Z musculo-tendinous lengthening of the brachialis and the biceps brachialis were also performed.

The anterior release was limited by neurovascular anterior structures (radial nerve and vascular humeral bundle), and the residual deficit of extension was 45° at the end of the procedure. No other approach and no posterior release were associated.

After surgery, this child was treated by three casts in maximal extension gained, made every ten days under general anaesthesia to increase the ROM and to stretch neurovascular bundle. Tolerance of this postoperative treatment was excellent particularly for neurological structures. After series of cast (1 month after surgery), the deficit of extension was reduced to 15°. Turnbuckles splints (one for extension, one for flexion) were subsequently used in

Figure 1  X-ray lateral views before surgery. A: 12 days after birth. B, C: at 8 months of age; note the incurvation variation according to different obliquity of the lateral view. D: arthrography just before surgery (16 months of age) showing absence of anterior capsular recessus.

Figure 2  Spontaneous attitude at time of refer in our institution for left congenital isolated elbow contracture with limited range of motion (ROM) to 0–90–120°.
order to preserve extension and flexion. These two different splints were alternated every two hours during three months and replaced with night splints. Passive soft mobilisations by parents were performed. Because of 20° deficit of extension noticed one year after surgery, a new series of three casts in extension were performed (without anaesthesia). Physiotherapy and splinting were stopped before 2 years of age. The ROM during management is resumed in Fig. 4. Pronosupination was always noticed normal: 0°–0°–170°.

After five years of follow-up, the result was still excellent with good passive and active ROM, no growth disturbance and no joint elbow instability (Fig. 5). The improvement of total arc of elbow motion was 430% (from 30° to 130°). No pain or no discomfort was described in daily living activities.

Hani used his left arm in all activities like his right arm. X-rays after last follow-up showed no abnormality (Fig. 6).

Discussion

Early anterior elbow surgical release following by series of cast was successful in management of isolated congenital contracture in flexion in our case. The total arc of motion increased from 30 to 130° at 5 years after surgery (430% of improvement motion).

There are few data in literature about surgical elbow release in children. Series mainly included post-traumatic elbow contracture [6,9]. In 1994, Mih et al. reported on nine patients (average age 12 years) with significant loss of

Figure 3  Elbow magnetic resonance imaging (MRI) before surgery suggested a congenital hypoplastic anterior articular capsule with normality of joint surfaces.

Figure 4  Evolution of elbow range of motion during main steps of management of the flexion elbow contracture.
Figure 5  Five years follow-up after anterior surgical release, the range of motion (ROM) was $0^\circ$–$5^\circ$–$135^\circ$, pronosupination was normal and no growth disturbance was noticed.

functional elbow ROM despite preoperative physical therapy and splintings [6]. Surgical release was performed and the ROM was approximately improved of 100%. A lot of authors suggest that good results of surgical treatment can be expected in most patients with sequel of traumatic elbow [4,5,8]. On the opposite side, Stans et al. found that surgical release of elbow stiffness in children were less favourable and less predictable than in adults [7]. But congenital and post-traumatic contracture are not similar disease with different age at time of management and prognosis. Post-burn elbow stiffness could be a closer problem to congenital disease, and in such condition better results have been reported when release surgery was performed early [10]. Our study could reach to similar conclusion.

Congenital contracture reported in this study could be assimilated to other congenital stiffness like arthrogryposis elbow but the problematic is clearly different. In arthrogryposis multiplex congenita, the extension contracture of the elbow joint especially is the main concern as it makes it impossible to reach the mouth or to perform hygienic necessities [2]. Therefore, the rehabilitation program includes an improvement of passive elbow flexion by capsulotomy [1] or of active flexion by triceps transfer if possible or both [2]. Williams et al. recommended surgical treatment after

Figure 6  X-ray lateral views after surgery. A: 45 days following surgery; note that humeral incurvation is still present. B, C: bone modelage after 3 and 5 years follow-up.
the age of 5 [3]. According to Axt et al., surgical treatment is indicated much earlier in condition of progressive bony changes of the elbow joints, or in condition of the possibility of advanced mental development and of independence [2].

At the contrary to extension contracture, the management of flexion contracture in arthrogryposis is rarely surgical despite of the stiffness because of a useful range of motion around the right angle position and an acceptable function [3].

The management of the case reported in this study did take into account the problematic of arthrogryposis elbow stiffness but no active muscular deficit was present in our case. Anterior capsular malformation or contracture could have been the primary disease and could have led to secondary anterior muscular and neurovascular bundle retraction. Because of humerocorondylar angle increasing at birth, it could also be argued that bony deformity has participated to mobility restriction (Fig. 1). We believe that a such humerocorondylar angle is not compatible with 90° deficit in extension; Simanovsky et al. found extreme variants in humerocorondylar angle up to 70° in children without any history of previous trauma and having a normal range of elbow motion [11]. Moreover, this bony deformation was still present few months after soft tissue surgical release (late modelage) suggesting that bone deformity was not contributed to the lack of extension (Figs. 1 and 6).

A lot of surgical approaches have been proposed for elbow release [12,13]. In our patient, anterior capsulotomy from lateral to medial structures was done by lateral approach alone, but careful attention must be paid to tension on the anterior neurovascular structures. We believe that release should be performed by extraperiosteal approach in order to reduce growth disturbance risk. It is usually admitted that range of motion obtained at the end of release will be the definitive outcomes. A different observation was noticed in our case, as flexion was reduced from 45° after surgery to 5° at the last follow-up. In order to increase extension, progressive corrections with successive casts under general anaesthesia were performed. The limiting factor was excessive traction in elbow extension on the neurovascular structures, particularly the radial nerve. This treatment represent an aggressive management with multiple general anaesthesiae, but was find to be a valid option to stretch progressively and safely the neurovascular bundle.

Passive motion, physical therapy and splinting were an important component of our postoperative protocol [14—16]. The disadvantage of surgery over 2 years old is the absence of cooperation in the postoperative physiotherapy program. Supportive parents are the key to success in a very young child; they must provide the time consuming and extensive postoperative care.

Conclusion

Aggressive management of isolated elbow congenital contracture in flexion by early anterior surgical release and repeat series of casts could be a valid option. It conducted to a very good result in our case. This good outcome will need to be confirmed with longer follow-up and the strategy to be validated in others cases.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

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