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Original article

Biplanar carpal wedge osteotomy in the treatment of the arthrogrypotic patients☆

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ABSTRACT

Objective: To describe the results of the surgical treatment of patients with congenital arthrogryposis with wrist deformity through biplanar carpal wedge osteotomy.
Method: This study analyzed nine patients through a retrospective evaluation with severe deformity in flexion and ulnar deviation of the wrist in the period between January 2004 and December 2009. They were submitted to carpal osteotomy with a biplanar dorsal resection wedge, with a minimum evaluation of 48 months of postoperative evolution. In three patients the osteotomy was bilateral, totalling 12 cuffs analyzed. The indication for the technique described was deformity and stiffness for over six months, without improvement with the conservative treatment.
Results: The mean age of the patients on the day of surgery was five years and eight months. The initial mean wrist mobility was 35°, and the joints presented a mean flexion of 72.5° in a resting position. Osteotomy union occurred in all patients at an average period of 5.7 weeks. The final position of the resting wrist was 12° of flexion and the mean mobility was 26.6°, slightly lower than preoperatively but in a much better position. No serious complications arising from surgery or in the immediate postoperative period were observed.
Conclusions: Carpal osteotomy with biplanar dorsal resection wedge was useful and effective in helping to correct the deformities in flexion and ulnar deviation of the wrist, maintaining a reasonable mobility. It is a preservation surgery, which has low morbidity and avoids the progression of deformity and future degenerative changes.

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Osteotomia intracárpica biplanar no tratamento de pacientes com artrogripose

Palavras-chave:
Artrogripose/terapia
Artrogripose/cirurgia
Procedimentos ortopédicos/métodos
Osteotomia/uso

RESUMO

Objetivo: Descrever os resultados do tratamento cirúrgico de pacientes portadores de artrogripose congênita com deformidade do punho através da osteotomia intracárpica biplanar.
Método: Por meio de uma avaliação retrospectiva, entre janeiro de 2004 e dezembro de 2009, o estudo avaliou nove pacientes com grave deformidade em flexão e desvio ulnar do punho por artrogripose submetidos a osteotomia intracárpica com cunha de ressecção dorsal biplanar, com avaliação mínima de 60 meses de evolução pós-operatória. Em três pacientes, a osteotomia foi bilateral, perfez 12 punhos analisados. A indicação da técnica descrita foi deformidade e rigidez havia mais de seis meses, sem melhoria com tratamento conservador.
Resultados: A média de idade dos pacientes no dia da cirurgia foi de cinco anos e oito meses. A média de mobilidade inicial do punho foi de 35° e as articulações apresentavam 72,5° de flexão média em posição de repouso. Todas osteotomias consolidaram em um período médio de 5,7 semanas. A média da posição final do punho em repouso foi de 12° de flexão e a mobilidade média foi de 26,6°, ligeiramente inferior ao pré-operatório, porém mais bem posicionado. Não foram observadas complicações graves decorrentes da cirurgia ou no pós-operatório imediato.
Conclusões: A osteotomia intracárpica com cunha de ressecção dorsal biplanar se mostrou útil e eficaz no auxílio da correção da deformidade em flexão e desvio ulnar do punho, com manutenção de uma mobilidade razoável. É uma cirurgia preservadora, com baixa morbidade e que evita a progressão da deformidade e alterações degenerativas futuras.

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Introduction

A word of Greek origin, arthrogryposis means rigid articulation. It is not a single diagnosis, but rather a syndrome characterized by severe deformities and multiple joint contractures associated with muscular atrophy. Even in the presence of gross deformities, patients are characterized by an excellent adaptation to difficulties due to an above-average intelligence.[1–4]

The challenge is in deformity correction, which is difficult and complex, with frequent postoperative recurrences.[5–10] Several therapeutic procedures have already been described for the treatment of the wrist deformities, among them proximal carpectomy,[11] use of external fixator,[4] arthrodesis,[3] and distal osteotomies of the forearm.[5,12]

Joint alterations of arthrogryposis may affect the four limbs; the upper limb involvement is usually characterized by symmetrical, multiple involvements with internal rotation of the shoulders, elbow extension, forearm pronation, flexion and ulnar deviation of the wrist, and rigid fingers in flexion with adduction contracture of the thumb.[2,4] As lower limb alterations are frequently observed, it is not uncommon for these patients to require the upper limbs to aid in locomotion. In the presence of a flexion deformity of the wrist, patients may use the back of the wrist and hand to support themselves on the ground, which is evidenced by the presence of local calluses (Fig. 1). Such practice, in addition to increasing the wrist deformity, can lead to radiocarpal subluxation and a degenerative joint process.

The aim of upper limb treatment in patients with arthrogryposis is to assist in independent walking and the use of both hands in daily tasks, allowing them to perform feeding and hygiene tasks independently.[3,4,13] The aim is to reduce the deformity, increasing or maintaining the range of active and passive movements.

This study is aimed at describing the results of the surgical treatment of patients with congenital arthrogryposis with severe deformity in flexion and ulnar deviation of the wrist through biplanar intracarpal osteotomy, preserving the distal radial physis with the resection of a biplanar wedge, with a dorsal and radial base, according to the technique described by Ezaki[14] (Fig. 2).

Material and methods

This is a retrospective study in which patients with a diagnosis of arthrogryposis who presented severe deformity in flexion and ulnar deviation of the wrist underwent biplanar dorsal carpal wedge osteotomy, performed between January 2004 and December 2009. Nine patients were selected; three had bilateral deformities and were operated on both wrists. Therefore, a total of 12 wrists were analyzed. Only patients with a complete evaluation and at least 48 months postoperative evolution were included in the study. The study was authorized by the research ethics committee of this hospital under protocol No. 63948217.1.0000.5683, opinion No. 2.057.952.

Fig. 1 – Male patient, 4 years, with arthrogryposis and severe deformity in flexion and ulnar deviation of the wrist (A). He uses the back of the wrist and hand to help in locomotion and presents a local callus (white arrow) (B and C). Radiograph of the wrist shows no bone changes (D). Preoperative mobility of 35° (E and F).

The criteria for indication of surgical treatment were rigidity, ulnar deviation and wrist flexion above 40°, and no improvement after over six months of conservative treatment (physical therapy and orthosis use). This technique is used for pediatric patients (open distal radius physis). This retrospective review examined the clinical and radiological findings and possible complications arising from the surgery. As the study comprised a pediatric population with multiple joints involved in the pathology, the authors decided not to use quality of life or function questionnaires. The wrist joint mobility angles (flexion and extension) were measured clinically with a goniometer. Through posteroanterior and lateral wrist radiographs, the time of union and the presence of intracarpal synostosis were analyzed. The parents/guardians satisfaction with the surgical procedure was evaluated on a simple scale from 0 to 10, in which 0 corresponded to very dissatisfied and 10 to very satisfied.

Surgical technique

The patient is positioned in a supine position under general anesthesia. A pneumatic tourniquet is applied at the base of the upper limb.

Firstly, a longitudinal volar incision is made, allowing the release of contracted soft tissue, such as the antebrachial fascia and the joint capsule. Subsequently, the flexor tendons are assessed; if fibrotic and without an excursion, they must be sectioned. If they appear to be healthy and functional but contracted, they are isolated and subsequently elongated by Z-plasty or intramuscularly.

Then, a longitudinal dorsal incision centered on the wrist is made. The authors modified Ezaki's original technique, which includes a transverse dorsal incision and sometimes a second incision in the distal part of the forearm.[15] By using a dorsal longitudinal incision, the cutaneous sensory branches

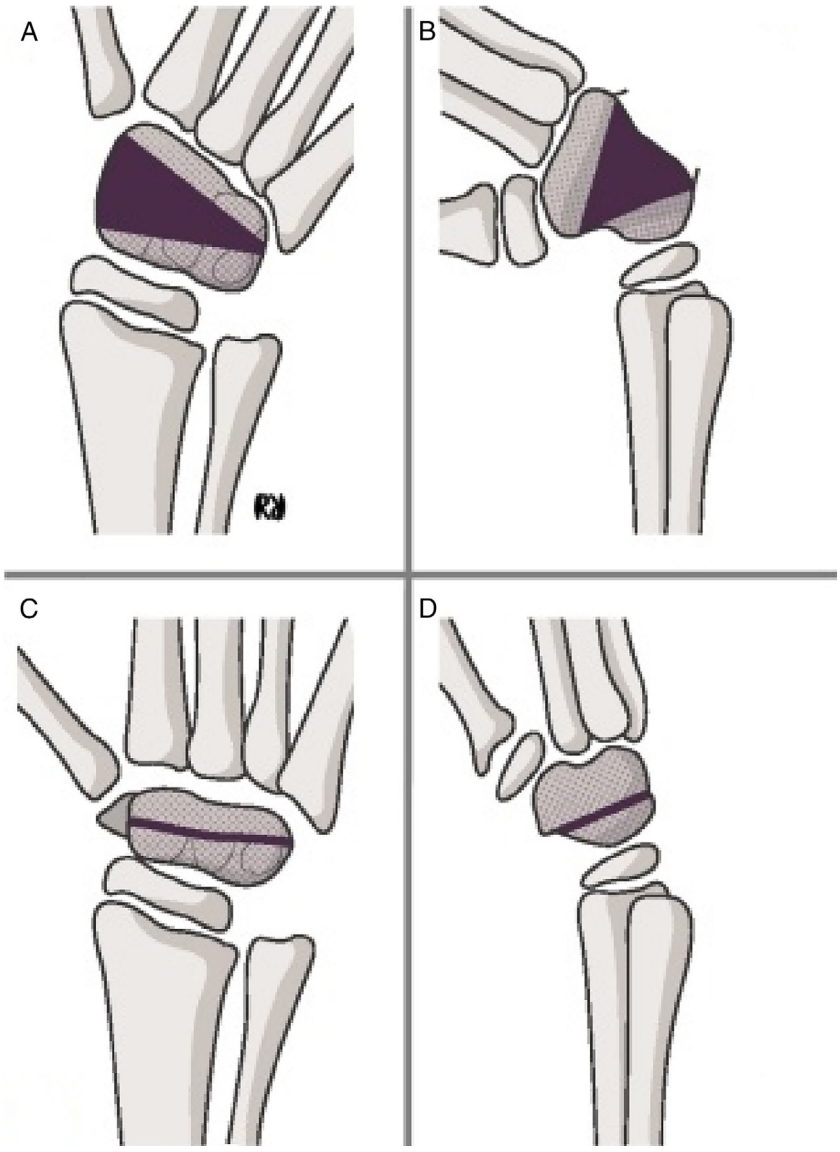

Fig. 2 – Intracarpal osteotomy planning for correction of the deformity in flexion and ulnar deviation of the wrist in arthrogryposis. The first cut is made on the proximal row of the carpus, immediately distal to the radiocarpal joint and perpendicular to the two axes of the forearm. The second, more distal cut is made on the distal row perpendicular to the two long axes of the metacarpals (A and B). After the bone wedge is resected, the defect is closed and the hand is repositioned radially and dorsally, achieving the position planned preoperatively (C and D).

and the dorsal veins are more easily protected. Furthermore, it allows a better release of the extensor tendons, without the risk of causing any complications. Care should be taken to maintain intact venous return and the cutaneous sensory branches. Each extensor compartment (third to fifth extensor tendon compartment of the wrist) must be mobilized and individualized. Subsequently, the extensor carpi radialis tendons should be isolated and released proximally (they usually are not easily distinguishable from the dorsal capsule and the distal radius); they should be dissected carefully and released as proximally as possible, preparing to receive the transfer of the extensor carpi ulnaris (ECU). The ECU is then identified, dissected proximally, and sectioned at its insertion on the base of the fifth metacarpal so it can be transferred to the radial side. The next step is the arthrotomy. Initially, space should be created between the capsule and the first extensor compartment, so that a retractor that will protect the dorsal branch of the radial artery can be placed. The radiocarpal joint is then easily palpable, and a transverse incision is made centered on the lunate, scaphoid, and pyramidal. A rectangular dorsal based capsule flap is then made, allowing a complete exposure of the distal portion of the carpus. For the osteotomy, two retractors are positioned in the capsule, exposing the proximal and distal carpal rows. The wrist is then positioned in maximum passive extension. The first osteotomy cut (the most proximal) is made on the proximal carpal row, distally to the radiocarpal joint and perpendicular to the two axes of the forearm. At this stage, maximum care must be taken not to

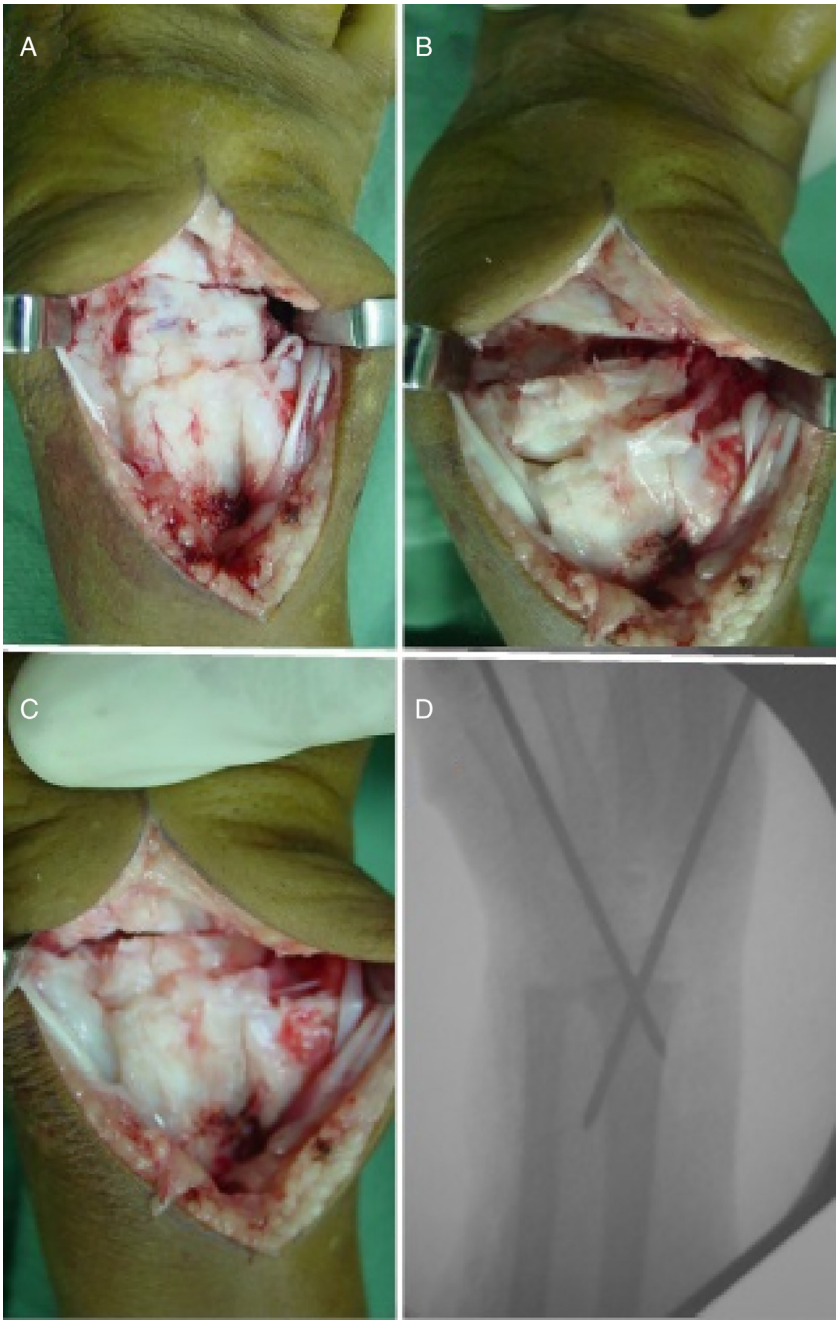

Fig. 3 – Intracarpal osteotomy of the wrist in arthrogryposis. Proximal and distal cuts (A). Wedge resection(B). Correction of deformity (C). Fixation using retrograde cross-linked Kirschner wires (D).

invade and injure the radiocarpal joint, as this is the location of the residual mobility of the wrist. The second, more distal cut is made on the distal row perpendicular to the two long axes of the metacarpals. This will create a wider bone wedge on the dorsal and radial sides with the palmar and ulnar apexes. Osteotomy cuts can be made with scalpels or with delicate osteotomes, without the need of an oscillating saw. After the bone wedge is resected, the defect is closed radially and dorsally, achieving the position planned preoperatively. Carpal osteotomy can be stabilized with intraosseous unabsorbable sutures, connecting the proximal to the distal row. For patients with a severe deformity, the wrist can be stabilized with 2.5 mm Kirschner wires, inserted retrograde and crossed from the base of the metacarpals to the distal portion of the radius; they fixate the osteotomy to the radiocarpal joint, allowing greater patient comfort and rest (Fig. 3). The flap with the dorsal capsule is then closed; it may be necessary to resect the excess. At this time, the ECU tendon should be transferred to the radial side. It may be sutured to the insertion of the ECRB or the ECRL with moderate tension. If necessary, the tension of the flexor tendons and their elongation are revised. A skin is closed with monocryl 4.0® absorbable sutures. A thick bandage and non-adherent gauze (Adaptic®) is used. The upper limb is then immobilized with an above-elbow

Table 1 – Series of patients who underwent intracarpal osteotomy for the correction of flexion deformity and ulnar deviation of the wrist in arthrogryposis.

	Gender	Age	Side	Initial position Flexion at rest	Final position Flexion at rest	Type of fixation	Support on the back of the hand	Follow-up (months)	Time until union (weeks)	Pre-operative active mobility	Post-operative active mobility
1	M	3 y + 3 m	L	45°	5°	Kirschner wires	No	132	7	40°	30°
2	M	4 y + 5 m	R	110°	20°	Kirschner wires	Yes	120	6	25°	20°
3	M	5 y + 10 m	L	85°	15°	Kirschner wires	Yes	96	6	30°	25°
4	M	4 y + 1 m	R	90°	0°	Kirschner wires	Yes	120	6	35°	30°
5	F	5 y + 1 m	L	60°	5°	Kirschner wires	No	108	7	30°	20°
6	F	6 y + 2 m	R	60°	10°	Kirschner wires	No	132	5	25°	20°
7	F	7 y + 3 m	L	50°	15°	Kirschner wires	No	120	6	35°	30°
8	M	5 y + 3 m	R	105	0°	Cerclage	Yes	48	5	40°	30°
9	M	6 y + 6 m	L	85°	10°	Cerclage	Yes	60	6	35°	30°
10	F	8 y + 1 m	R	70°	50°	Kirschner wires	No	108	5	40°	25°
11	F	5 y + 6 m	L	40°	0°	Cerclage	No	60	7	45°	35°
12	F	6 y + 11 m	R	70°	15°	Cerclage	No	60	6	40°	25°
Mean	6♂ 6♀	5 y + 8 m	6 R 6 L	72.5°	12°	Kirschner wires 08 Cerclage 4	No 7 Yes 5	97	5.7	35°	26.6°

splint. Patients are advised to keep the upper limb elevated in the first few days after surgery. Dressings under sedation are not routinely made. The first dressing is made between ten and 14 days after surgery. After the first three weeks, the elbow splint can be changed to a forearm splint; it should be used until osteotomy union, which occurs an average of six weeks. At this stage, the Kirschner wires (if used) are removed. After this period, the authors recommend the use of a removable orthosis for a minimum of six months, to prevent the recurrence of deformity and to prevent the child from attempting to use the back of their hands to support themselves on the floor.

Results

The study included 12 osteotomies in nine patients, five females and four males. Six right and six left wrists were operated. Age at surgery ranged from 3 years and 9 months to 8 years and 1 month, with a mean of 5 years and 8 months. Regarding the wrist position in the preoperative period, patients had a mean deformity at rest of 72.5°, ranging from 40° to 110°. The mean active mobility in the preoperative period was 35°, ranging from 25° to 45°. Five of the 12 wrists operated on, the patient used the back of the hand as support to help with locomotion (Table 1).

The implant material used for osteotomy fixation varied according to the surgeon's choice, the severity of the deformity, and the learning curve. In eight surgeries, two 2.5 mm retrograde crossed Kirschner wires were used; in four, dorsal cerclage bone suture was applied directly at the osteotomy site.

All osteotomies healed, as evidenced by posteroanterior and lateral radiographs of the wrist. The mean union time was 5.7 weeks, ranging from five to seven. No serious complications resulting from the procedure, such as nerve or vascular injury, infection, or breakdown of the implant material, were observed intra or postoperatively. The five patients who used the back of their hand as a support to help with locomotion

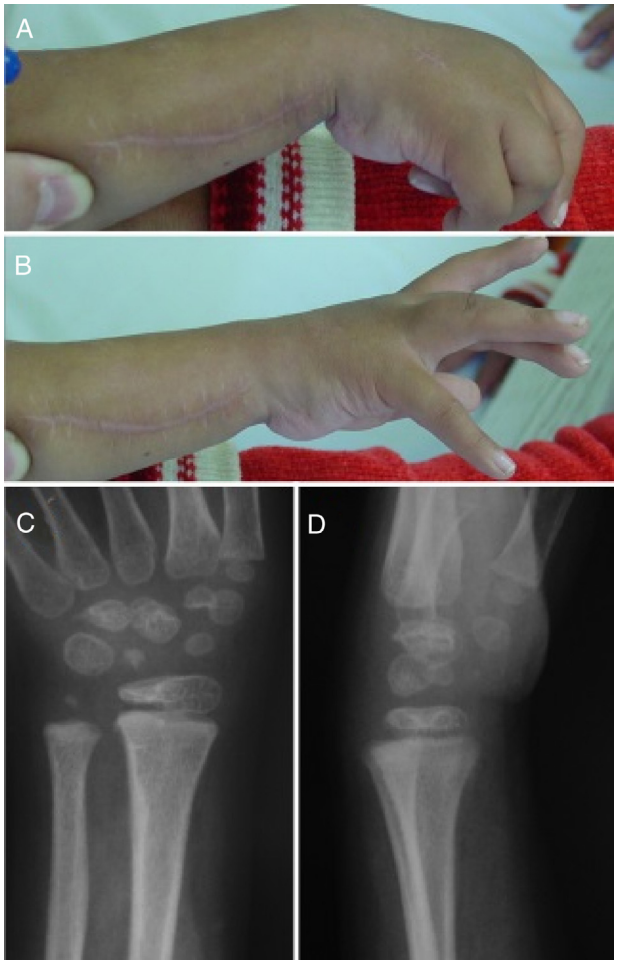

Fig. 4 – Results at 120 months of follow-up. Good mobility of the fingers and 30° of active mobility of the wrist (A and B). Radiographs show good alignment and coalitions in the mediocarpal joint at the osteotomy site (C and D).

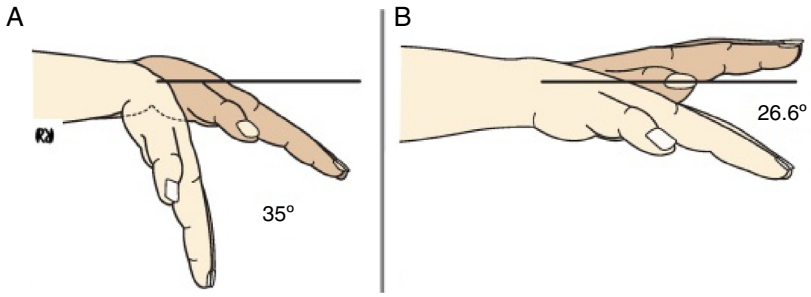

Fig. 5 – The mean active mobility in the preoperative period was 35° (A). The mean active mobility in the late postoperative period was 26.6° (B). This slight loss in active mobility is justified by the fact that the wrist became more functional, as it is closer to the neutral position.

instead began to use their palmar region (Fig. 4). On average, the final assessment was made 97 months after surgery, ranging from 48 to 132 months. On that day, the mean wrist position at rest was 12° of flexion, ranged from zero to 50°. The mean active mobility of the wrist in the final assessment was 26.6°, ranging from 20° to 35° (Fig. 5).

In this group of patients, the parents/guardians degree of satisfaction with the procedure was high: 8.7, on a scale ranging from zero to 10.

Discussion

Arthrogryposis refers to a group of over 150 different disorders, genetic or non-genetic, characterized by severe non-progressive joint deformities and stiffness caused by muscular fibrosis and capsuloligamentous shortening.[1,2] It is a rare pathology and occurs in 1:3000 live births.[1,2] Despite the joint deformities, these patients present no sensory or cognitive deficits.[1–3] The deformities in arthrogryposis are variable in number and severity.[3,4] In 46% of the cases, it affects the four limbs; in 43%, only the lower limbs; and in 11%, only the upper limbs.[3,4] Involvement is more common in the proximal joints; the more distal the disease, the more severe.[3,4] Most patients present the so-called amyoplasia or classical arthrogryposis, in which the deformities in the limbs are symmetrical and present at birth.[16–19]

Burgess and Robbe[12] emphasized that isolated joint correction, without checking the whole limb, may have a negative impact on function. A clear example of this fact is the case of patients with limited passive elbow mobility who use flexion wrist deformity as an aid to put the hand in the mouth.[12] Surgical correction of the wrist would impair their ability to feed themselves independently.[12]

There is no rule as to whether treatment should begin in the upper or in the lower limbs, either proximally or distally.[4–6] The goal is for the child to be able to stand up at 18 months and for the upper limbs to be correctly positioned at four years of age.[4,6] It is known that, at 5 years of age, 85% of patients with arthrogryposis are able to walk.[4,6] The wrist is usually flexed and in ulnar deviation, while the forearm is pronated, which causes an increase in the internal rotation shoulder deformity.[4–6] The volar structures are shortened and tense, and the extensors are inactive.[7] The goal is to preserve mobility and correct the hand positioning.[4–7] Sometimes, an exaggerated correction of the wrist deformity can worsen the function of the fingers.[1,3]

The priority in the treatment of the upper limb of patients with arthrogryposis is to achieve passive mobility of the elbow, maintaining active extension.[1–4] Other goals include placing the wrist in a neutral position, preserving the radiocarpal mobility, increasing the space of the first commissure between the fingers, withdrawing the thumb from the palm, and maintaining the sensitivity of the hand.[1–5] The mean wrist range of motion of patients with arthrogryposis is 45°, with flexed joints and contracted and tense volar structures.[20]

Several surgical techniques have been described to correct the flexion deformity and ulnar deviation of the wrist in patients with arthrogryposis.[1] Similarly to the treatment of congenital clubfeet, arthrogryposis treatment always starts early with therapy and use of orthoses or serial plaster casts, with the aim of improving passive mobility.[4,18] For severe lesions or those that have not improved with conservative treatment, surgical correction is recommended.[4–7] Among the surgical techniques described are tendon transposition,[1] arthrodesis,[3] the use of external fixator,[4] proximal carpectomy,[11] and distal osteotomies of the radius.[5,12] Although the contractures are not progressive, there is a high risk of deformity recurrence after surgery, regardless of the technique used. Therefore, the use of orthoses is recommended until the end of growth.[3]

Differently from other congenital pathologies, the surgical treatment for wrist deformities in arthrogryposis with soft tissue procedures, such as joint releases and tendon transfers, usually has poor results.[1,7] This is due to the severity of the contracture, as well as to inactivity and muscular atrophy.[2,18,20]

Total arthrodesis should be avoided as an initial surgical procedure for wrist deformity since arthrogryposis tends to reach multiple joints and the residual mobility of each joint must be preserved.[1]

The first bone procedures described for the treatment of arthrogryposis in the wrist were carpectomy and osteotomy of the distal portion of the radius and ulna.[1,2,11] Wenner and Saperia[11] performed carpectomy in five patients with an average of 11 years. They reported that, although a good intraoperative correction was observed, it was not maintained in the long-term.[11] They reported that 60% of their patients had

poor results and evolved to total arthrodesis of the wrist, and they no longer indicated the procedure.[11] Menem[13] described the results of 29 patients (44 wrists) who underwent proximal carpectomy. Followed-up for a mean period of seven years, these patients presented final active mobility of 27°.[13] Those authors advocate that the procedure should be performed when the patients are aged between 3 and 6 months, when the carpal bones have not yet calcified.[13] Esaki[14] states that, due to the presence of coalitions and synostoses in carpal bones in adult arthrogryposis patients who did not undergo surgery, a proximal carpectomy would alter the anatomy of the radiocarpal joint, the main source of residual wrist mobility in these patients.[14,20–22]

Burgess and Robbe[12] observed that osteotomies of the distal portion of the forearm lead to unsatisfactory results in the long-term. This can be explained by the great growth potential; the bones tend to remodel over time, causing deformity recurrence.

In an attempt to maintain mobility without altering bone growth potential, Ezaki[14] described that intracarpal osteotomy in dorsal and radial wedge resection, with the correction of the deformity in two planes (flexion and ulnar deviation), generates synostosis between the first and the second carpal row, an alteration expected in adults, and that does not influence the final mobility. Osteotomy should be associated with soft tissue procedures, such as volar capsule release, flexor tendon elongation, and tendon transfer (such as from the ECU to the extensor carpi radialis brevis).[14] In 2013, a series of 46 patients (75 wrists) assessed the results of this procedure.[20] The mean initial position at rest was 55° flexion and increased to 11°. The mean active mobility decreased from 32° to 22°, but the arch became more functional because it was located near the neutral position. The results were maintained even in a long follow-up period (68 months), with a satisfaction of 9.1 (ranging from 0 to 10).[20]

Van Heest and Rodriguez[23] analyzed the results of intracarpal osteotomy in 12 patients (20 wrists) followed-up for a mean period of 45 months. They found that the final mobility was very similar to the initial; however, it was located near the neutral position. They also observed that the results were better in patients older than 7 years and when the ECU was transferred during the same procedure.

The present results were similar to those obtained by other authors using this technique.[20,23] It should be noted that the lack of organization of the Brazilian public healthcare system causes a delay in the referral of patients with serious congenital pathologies to reference services. This caused the average age of the present patients to be slightly higher than that recommended in the literature (5.8 years). No studies that related the severity of the wrist deviation with the use of the back of the hand as a support for locomotion were retrieved in the literature. The authors observed that the deformity was greater in these children; for these cases, the use of transarticular Kirschner wires is recommended for stabilization, leaving intraosseous suture for the less severe procedures.

In an isolated analysis, mobility evolved with loss of active wrist range of motion from 35° to 26.6°, when compared with the preoperative and the late postoperative periods. This result is similar to those of the other published series that used this technique.[20,23] Such loss can be justified by the fact that the mobility arch is located in a more anatomical and functional position.

In one wrist (patient 10, Table 1), a moderate deformity recurrence was observed; in all other surgeries, both patients and parents/guardians noticed functional improvement and greater ease in performing manual tasks.

The present article has some limitations. It is a retrospective study with a small series of patients; therefore, there was no control group submitted to another type of procedure for comparison. Moreover, arthrogryposis is a pathology that affects several joints; the present patients underwent other surgeries in the same period, and it was difficult to isolate the results and the satisfaction with the wrist procedure alone.

Conclusion

The use of intracarpal osteotomy with biplanar dorsal wedge resection was useful and effective in helping to correct the flexion deformity and ulnar deviation of the wrist, maintaining a functional mobility that persisted over time. The present results were similar to those obtained by other authors using this technique. It is a preservation surgery, with low morbidity, that increases the child's functional capacity and quality of life.

Conflicts of interest

The authors declare no conflicts of interest.

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