Madelung's deformity: distal radioulnar joint re-optimization

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Background
Although Madelung's deformity (MD) is believed to be a rare congenital anomaly of the wrist, the symptoms do not manifest till late childhood. It is caused by asymmetric growth at the distal radial physis secondary to premature and partial volar–ulnar-sided arrest, and a thickened radiolunate volar ligament (Vickers ligament) that creates a tether across that physis segment, restricting growth across it. Radial growth disturbance causes the distal radius to deform in the coronal (increasing radial inclination) and sagittal (increasing volar tilt) planes, and eventually lead to a progressive, three-dimensional wrist deformity including radiocarpal and distal radioulnar joint (DRUJ) malalignment. Various techniques for the surgical management of MD have been described, but clear evidence to support the use of any single approach is lacking.
Patients and methods
Eight wrists in six patients with MD were included in this study. All patients underwent combined radial biplane closing wedge osteotomy above Vickers' ligament radial origin, which allowed for three-dimensional correction of the deformity and reposing of the ligament, making it easy to achieve more congruity of the DRUJ, and temporary fixation with dual percutaneous K-wires of the DRUJ in the achieved re-optimized new components' correlation.
Results
At 22 months of an average follow-up, patients were satisfied with the appearance of their wrists and forearms. The range of active pain-free wrist movements improved notably in supination and extension, correction toward normal values for the mean measurements of radiographic parameters was reported, and the DRUJ was congruent with sustained improvement of its mean gap distance at the latest follow-up radiographs.
Conclusion
This technique seems to be safe and can reliably restore hand–forearm alignment with respect to DRUJ re-optimization through the redesigned distal radial alignment in both coronal and sagittal planes, and improve joint congruity.

Keywords:
biplane, osteotomy, K-wire, Madelung', s deformity, vickers ligament

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Introduction

Madelung's deformity (MD) is a rare condition, which is found in only 1.7% of hand deformities [1], showing female predominance [2], occurring between the age of 8 and 13 years, often bilateral [3] and is frequently associated with Leri–Weill dyscondrosteosis, a form of mesomelic dwarfism [1,3]. This was first reported by Malgaigne in 1855 [4]; then, in 1878, Madelung described this congenital painful wrist deformity [5,6]. It is caused by premature partial closure of the volar–ulnar part of the distal growth plate of the radius [7]. Subsequently, Madelung-like deformities have been reported in a variety of acquired forms [5,6,8].

Despite being a congenital disorder, the symptoms are absent till late childhood. MD is characterized by the presence of an abnormal volar structure, Vickers ligament [1], originating from the ulnar border of the radius in a flame-shaped notch and inserted into the palmar pole of the lunate [9,10] that tethers the lunate to the volar distal radius, and is believed to impede the growth by compressing the growth plate [10–12]. Vickers ligament is not seen in Madelung-like deformities and serves to distinguish between the two entities [8].

As the remaining physis grows, the radius deforms with increasing radial inclination and volar tilt [8], while its segmental slowing of growth induces a helical movement around the longitudinal axis of the forearm [2]. The unaffected ulna continues to elongate, yielding a progressive ulnar positive variance. Ultimately, the distal radioulnar joint

(DRUJ) fails to form normally and the distal ulna subluxates or dislocates in a dorsal direction [8].

These anatomical changes alter the kinematics of the wrist and may result in decreased range of movements, particularly supination and extension, decreased grip strength and pain [5], and esthetic considerations if the deformity is severe [3]. Although its rarity hinders the assessment of the various surgical options for improving esthetics and function [2], various combinations of opening or closing wedge osteotomies were developed and long-term data are available for some procedures [13,14]. Recently, preservation of the distal ulna has become a priority [15], and several researchers have focused on the importance of preserving the DRUJ during MD correction [15–18], a principle that is incorporated into our technique, which comprises of radial biplane closing wedge osteotomy (BCWO) correction of the deformity, and DRUJ re-optimization through a redesigned distal radial alignment in both coronal and sagittal planes, plus temporary fixation with dual percutaneous K-wires of the DRUJ in the new component correlation achieved.

Patients and methods

After IRB approval was obtained, eight wrists in five female patients and one male patient were studied; they were operated on between 2010 and 2015 at Mansoura University Hospital. Two patients were operated on bilaterally, while the other four (two right-dominant and two left-nondominant) were operated on unilaterally. Patient demographics, presenting complaints and any associated history for all patients enrolled in this study were documented. The average age at presentation was 12 years, and the average age at the time of the surgery was 12.3 years (range: 10–14 y). The mean follow-up time was 22 months (range: 17–30 months). Pain was the chief complaint at presentation in three patients (five wrists) and a secondary concern in two unilaterally affected patients. The average duration of pain was 18 months. Concern about increasing deformity was the most common complaint in all patients. Decreased range of motion was the presenting complaint for the bilaterally affected patients and a secondary concern in two others.

All patients underwent periodic serial radiographic examinations including standardized posteroanterior (PA) and lateral (Lat) views of their wrist both preoperatively and postoperatively. Parameters analyzed were digitally measured using Weasis v2.5.2 medical viewer software for radiograph files and included ulnar tilt (radial slope), Lunate fossa angle, palmar carpal displacement (mm) and Lunate subsidence (mm) as described by McCarroll *et al*. [19]. Also, the lunate-covering ratio [4], radial inclination angle [15] and the DRUJ gap (our measure to evaluate the results of temporary fixation of the DRUJ) were determined. Joint range of motion (in degrees) on a goniometer (flexion/extension and pronation/supination) and subjective esthetic discomfort were also assessed.

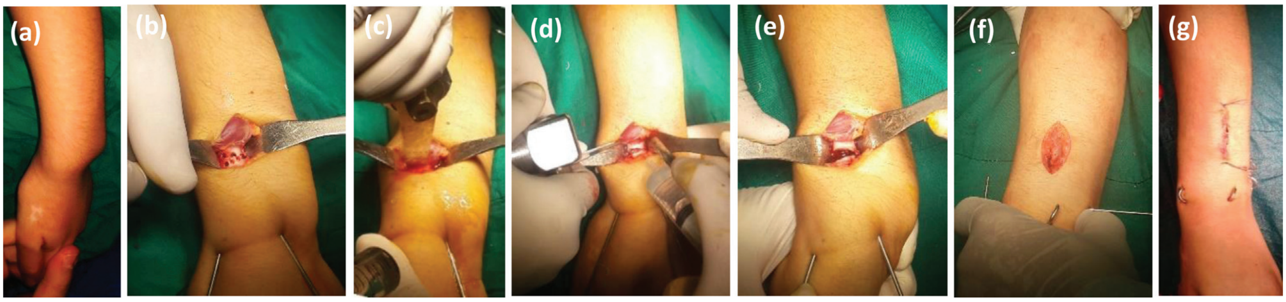
Surgical technique

The wedge angle and dimensions were estimated preoperatively as two triangles, dorsal and radial with a shared base, calculated to reoptimize the radial inclination and volar tilt radiologically using Weasis 2.5.2 medical viewer Software (Weasis DICOM viewer is cross-platform, free/libre and open source software (FLOSS), multi-language and allows a flexible integration to PACS, RIS, HIS or PHR. This multi-platform DICOM viewer runs on Windows, Linux, and Mac OS X. It allows high-quality renderings with high performance through the library) (Fig. 4a). Surgery was performed under general anesthesia with a tourniquet. Under control of an image intensifier, a landmark with an 18-gauge needle was performed just proximal to the flame-shaped notch of Vickers ligament radial origin (Fig. 3a, f), which was considered the site of osteotomy, and used as a radio-opaque target for two 1.6 mm K-wires that were percutaneously driven dorsally: the first one from the radial styloid process (radial side K-wire) and the second from the ulnar corner of the distal radius (ulnar side K-wire), drilled parallel to the radial and ulnar borders of the distal radius on the anteroposterior (AP) view, respectively, and directed centrally on the lateral (Lat) view aiming toward the needle (Fig. 2a).

A small dorsal skin incision was made centered upon the needle landmark, followed by blunt soft tissue dissection and sharp incision of the periosteum.

Using the preprepared dorsal and radial triangle templates, a few near-cortex drills with a K-wire were performed to outline the dorsal wedge ulnar angle with its distal limb at the needle landmark, and the shared base with the radial wedge at the dorso-radial border (Fig. 1b). With a pneumatic saw, the distal limb cut of the dorsal and radial wedge was performed, preserving the volar cortex and then the dorsal cortex wedge cut

Figure 1



Operative technique. (a) Preoperative view. (b) A few near-cortex drills were performed to outline the pre-planned wedge angle. (c) With a pneumatic saw, the dorsal wedge osteotomy was performed preserving the volar cortex. (d) With a pneumatic saw, the radial wedge was accomplished. (e) After removing the two triangles (dorsal and radial triangular components of biplane closing wedge osteotomy), (f) drilling of a K-wire under control of an image intensifier was performed from the distal ulna to the distal radius for closing the distal radioulnar joint leisure. (g) Skin closure.

Figure 2

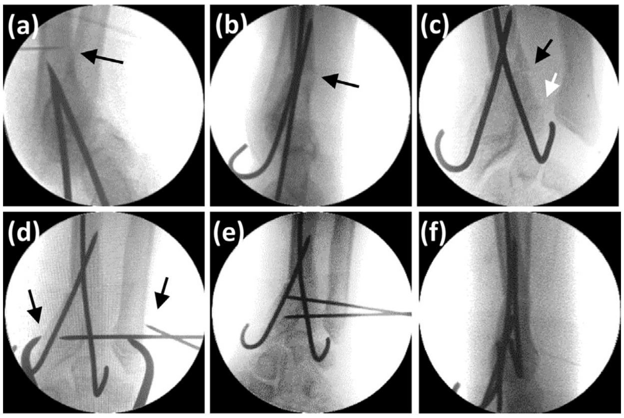


Image intensifier radiographs. (a) Radio-opaque landmark with an 18-gauge needle (black arrow) at Vickers ligament radial origin and two K-wires driven dorsally and aimed at the needle entry point. (b) Volar angulation (black arrow), correcting the volar tilt, after closing the biplane wedge. (c) Ulnar angulation (black arrow), correcting the ulnar tilt, after closing the biplane wedge, distal radioulnar joint leisure (white arrow). (d) Two K-wires were drilled from the distal ulna to the distal radius, while applying compression with a clamp (black arrows). (e) Intraoperative final anterioposterior position; note the minimized distal radioulnar joint gap. (f) Final lateral view after biplane closing wedge osteotomy and distal radioulnar joint K-wire fixation.

was completed (Fig. 1c); the same was done for the radial cortex wedge (Fig. 1d).

After removing the two triangles (dorsal and radial triangular components of BCWO) (Fig. 1e), the volar inner cortex was meticulously attenuated with the saw, and then joystick manipulation with both K-wires was carried out to the distal radial fragment while closing the biplane wedge space dorsoradially (Fig. 1f). Meanwhile, the ulnar side K-wire was impelled to the proximal medulla and when we reached an optimum radial inclination on the PA view, it was introduced medullary. Simultaneously, the radial side K-wire was used to uplift the distal radius dorsally to adjust the volar tilt on the Lat view and gain more closure of the wedge space dorsally, and was then driven to the proximal medulla (Fig. 2b, c).

Now, the distal radial articular surface was correctly repositioned in both coronal and sagittal planes, since the radial triangular component of BCWO produced volar angulation accompanied by dorsal shift of the distal radial fragment and corrected the volar tilt (Fig. 2b). Also, the dorsal triangular component created ulnar angulation proximal to Vickers ligament radial origin, which relaxes the ligament by distal and ulnar repositioning of its radial origin, and hence released the lunate from impingement against medioradial metaphysis and triangular fibrocartilage complex (TFCC), providing ease to gain nearer between the distal radial fragment and ulna, and permit for closing DRUJ leisure (Fig. 2c).

Then, dual 1.6 mm K-wires were drilled under control of an image intensifier from the distal ulna to the distal radius, distal to the osteotomy line and proximal to the distal radial epiphysis, while applying compression with a clamp on the distal ulna and radius (Fig. 2d–f). After skin closure (Fig. 1g), an above-elbow cast/splint was applied up to 12 weeks (Figs. 3 and 4).

Results

The results of our eight procedures have been satisfactory; pain was relieved and patients were satisfied with the appearance of the wrist alignment. Hence, they were able to resume activities that were prohibited by pain. Patients with bilateral deformity had requested the operation on the other wrist shortly after removing the cast and wires of the first one. Our experience mimics that of other researchers, who find

Figure 3

Radiological follow-up. Bilaterally affected patient. (a) Posterioanterior views, showing the characteristic flame-shaped notch at the radial origin of the Vickers ligament (white arrows). (b) Lateral view, showing dorsal ulnar displacement. (c) Postoperative follow-up at osteotomy union (right wrist). (d) Postoperative last follow-up for right and left wrists at 26 and 22 months, respectively. Unilaterally affected patient (e) posterioanterior and lateral views; note the radial notch at the origin of Vickers ligament (black arrow), and a bony bar bridges the epiphysis and metaphysis [20] (two thin arrows). (f) Postoperative follow-up at osteotomy union. (g) Postoperative last follow-up at 30 months.

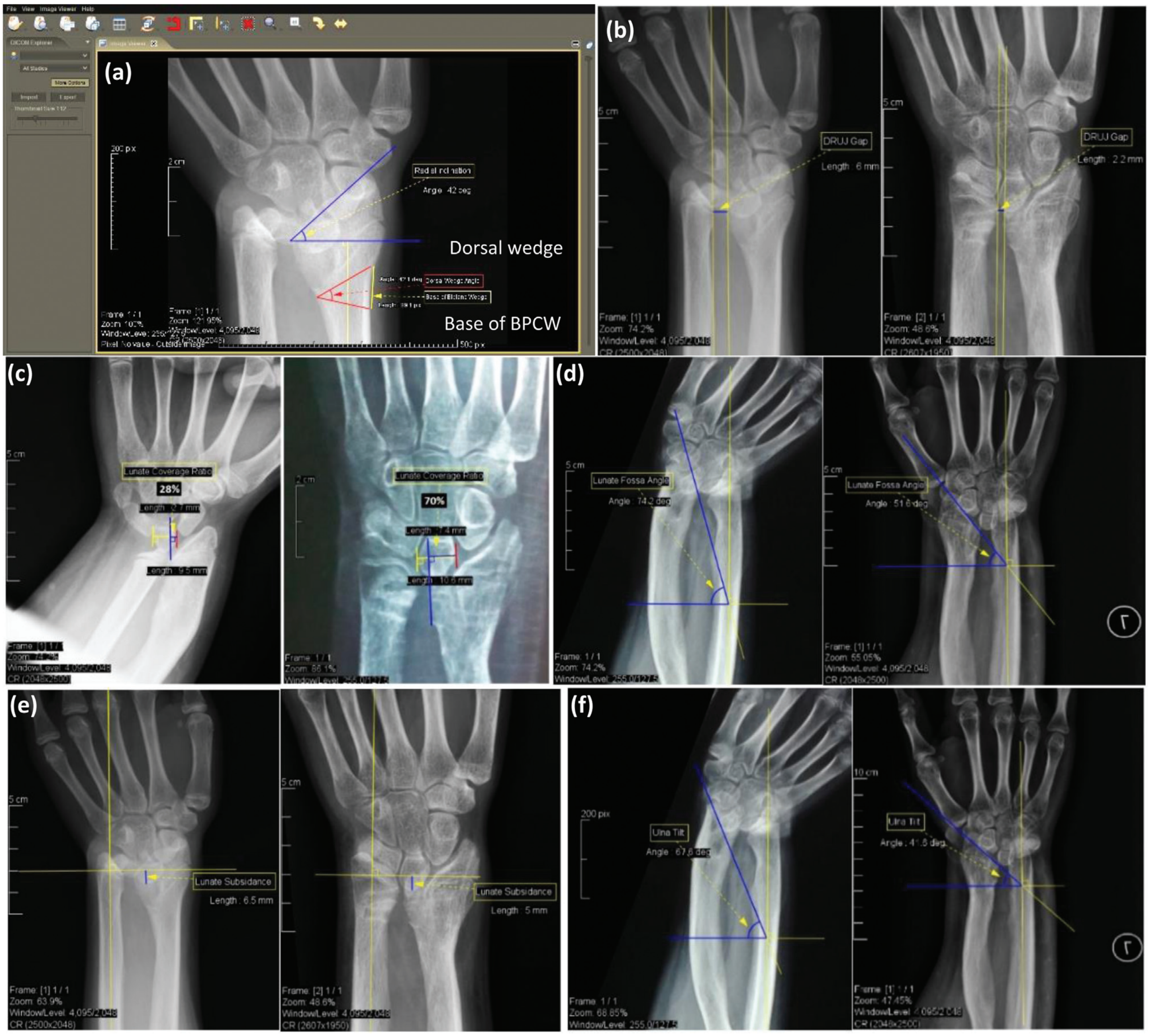
that it is possible to preserve a functional DRUJ while correcting wrists with MD. Although in a dominant wrist of one bilaterally affected patient the distal ulna sublaxed dorsally with passive manipulation but rested in normal alignment with the radius in the absence of external stress, this did not result in symptoms.

The osteotomy union was always achieved within 6 to 12 weeks. An above-elbow cast was used for 6 weeks and then a removable bivalve splint was used for an additional 6 weeks, allowing follow-up radiographs for osteotomy union, DRUJ and K-wire pin dressing. Removal of the immobilizing splint and wires was performed at 12 weeks. No intraoperative complications or neurovascular problems were reported. Bone union was achieved in all cases and no repeat surgery was required. Two cases of irritation of the skin occurred and were resolved after removal of the K-wires. There was no deep infection. One female patient (bilateral wrists) had a problem with a hypertrophic dorsal scar in her operated left wrist (nondominant) and was managed by scarectomy at the time of operation on the contralateral wrist.

For each clinical and radiological variable, the usual statistical tests were not carried out due to the small size of the series. There was a significant improvement in the postoperative range of motion, especially in wrist supination and extension, which increased from 50.6° ±24 to 79.3°±25 ($P=0.02$) and from 44.7°±16 to 75.3° ±17 ($P=0.004$), respectively (Table 1). There were no statistical changes in wrist pronation, flexion, or radial and ulnar deviations. Pronation increased from 70.4°±19 to 73.2°±22, while flexion increased from 69.5°±17 to 71.3°±15 on average.

The mean radiographic parameters assessed in this study showed correction toward normal measurements (Table 2). The mean ulnar tilt (radial slope) showed an average correction of 9.5°±6.7° (from 56.3°±6.8° to 46.8°±5.7°); the average lunate fossa angle correction was 8.1°±6.1° (from 61.1°±7.6° to

Figure 4



Radiographic parameter measurements. (a) Weasis 2.5.2 medical viewer software used to estimate the wedge angles and dimensions as dorsal and radial triangles with a shared base, considering the radial inclination; the angle between a line along the articular surface of the radius and another perpendicular to a line that bisects the head of the radius and passes through the radial edge of the distal radial epiphysis [15] and volar tilt. (b) distal radioulnar joint gap: measured digitally in mm between the nearest opposed distal points of the radius and ulna; wrist-1 improved from 6 to 2.2 mm, and there was an overall average improvement of 1.95±1.88 mm (*P*=0.002). (c) Lunate coverage ratio, the ratio between the lunate bone width covered by the radial joint surface and the total width of the lunate bone [4], was improved in wrist-2 from 28 to 70%, and there was an average improvement by 13% in the entire series. (d) Lunate fossa angle: between the long axis of the ulna and a line through the lunate fossa of the radius [4]; wrist-3 improved from 74.2° to 51.6°. (e) Lunate subsidence distance: between the most proximal point of lunate and a line perpendicular to the long axis of the ulna through its distal articular surface [4]; wrist-1 improved from 6.5 to 5 mm. (f) Ulnar Tilt (radial slope): angle between the longitudinal axis of ulna and a line tangent to the surfaces of scaphoid and lunate [12]; wrist-3 was corrected from 67.6° to 41.6°.

52.9°±6.4°), whereas the improvement in the mean lunate subsidence was 1.6±2.4 mm (*P*=.004), coverage of the lunate increased from 60%±22.3 to 67.8%±25.2, with an average correction of 7.8%±22.8 (*P*=0.003), radial inclination showed a 28.1°±10.3° (59.3° ±12.6°–31.2±9.8) improvement in position toward normal (*P*=0.001) and palmar carpal displacement average of correction was 2.7±2.6 mm (from 25.2 ±3.2 to 22.5±4.8), while the DRUJ gap improved by an average of 2.0±1.9 mm (from 6.1±0.9 to 4.1±1.6) for the entire series (*P*=0.002) on obtaining the latest follow-up radiographs (Fig. 5).

Discussion

MD ensues from an abnormality of the volar–ulnar portion of the distal radial physis, and abnormal thickened radiolunate volar ligament described by

Table 1 Mean clinical values and percentage of improvement

	Supination (°)			Pronation (°)			Extension (°)			Flexion (°)		
Movement	Pre-Op	Post-Op	Improvement	Pre-Op	Post-Op	Improvement	Pre-Op	Post-Op	Improvement	Pre-Op	Post-Op	Improvement
Mean±SD	50.6±24	79.3±25	**28.7**	70.4±19	73.2±22	**2.8**	44.7±16	75.3±17	**30.6**	69.5±17	71.3±15	**1.8**
Percentage/preoperative		***56.7%**			***3.9%**			***68.5%**			***2.6%**	

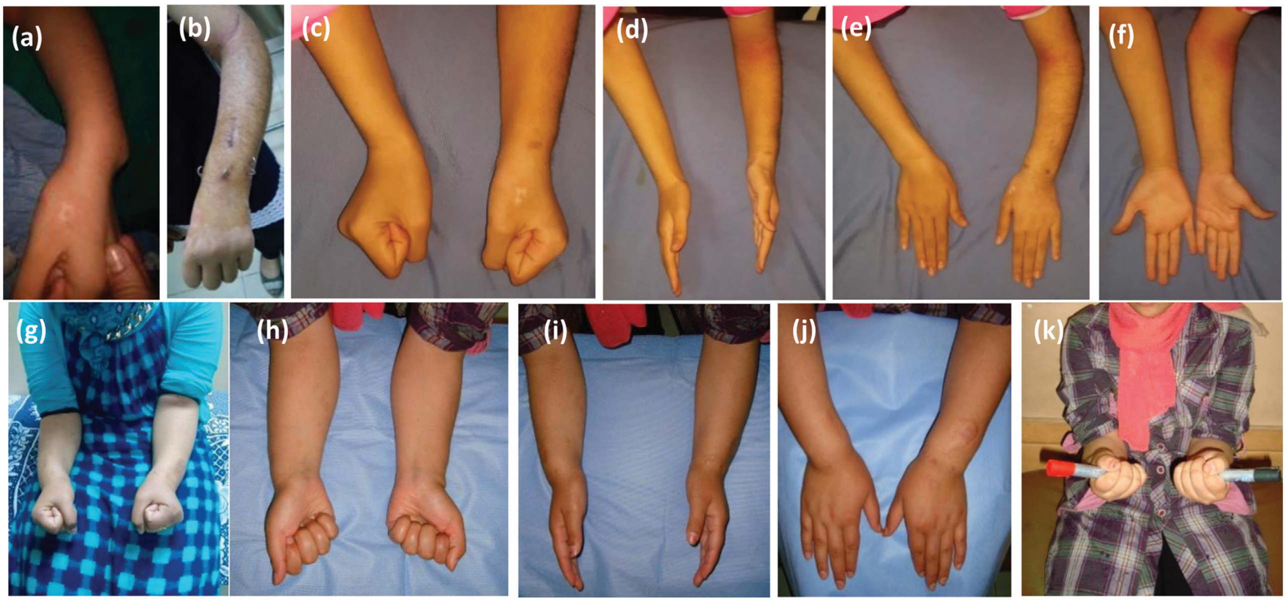
Values are given as the mean±SD. *Percentage of improvement in relation to the average preoperative value.

Table 2 Mean radiographic parameter values

	Ulnar tilt (deg.)			Lunate fossa angle (deg.)			Palmar carpal displacement (mm)			Radial inclination angle (deg.)			Lunate coverage ratio (%)			Lunate subsidence distance (mm)			DRUJ gap (mm)		
Wrist	Pre-Op	Post-Op	Correction	Pre-Op	Post-Op	Correction	Pre-Op	Post-Op	Correction	Pre-Op	Post-Op	Correction	Pre-Op	Post-Op	Correction	Pre-Op	Post-Op	Correction	Pre-Op	Post-Op	Correction
1	53.9	46.3	7.6	61.1	54.7	6.4	21.7	19.6	2.1	42.0	37.9	4.1	90.0	100.0	10.0	6.5	5.0	1.5	6.0	2.2	3.8
2	46.8	40.2	6.6	52.5	47.1	5.4	28.9	30.4	−1.5	80.9	46.0	34.9	28.0	70.0	42.0	10.2	7.8	2.4	5.8	3.9	1.9
3	67.6	41.6	26.0	74.2	51.6	22.6	28.2	28.4	−0.2	73.4	43.9	29.5	53.0	14.5	−38.5	7.6	6.3	1.3	5.5	7.1	−1.6
4	56.7	49.5	7.2	62.4	56.8	5.6	25.4	20.9	4.5	54.0	21.0	33.0	33.0	54.0	21.0	5.0	4.0	1.0	4.9	4.2	0.7
5	49.3	42.1	7.2	59.8	52.7	7.1	20.7	18.4	2.3	50.0	24.0	26.0	68.0	76.0	8.0	2.0	−2.0	4.0	6.5	5.1	1.4
6	53.8	45.7	8.1	53.9	49.1	4.8	27.6	24.5	3.1	61.0	26.0	35.0	82.0	84.0	2.0	3.0	−2.0	5.0	5.7	2.3	3.4
7	62.4	56.2	6.2	69.4	65.8	3.6	22.1	17.4	4.7	59.0	24.0	35.0	53.0	69.0	16.0	13.0	16.0	−3.0	8.0	3.9	4.1
8	59.7	52.9	6.8	55.2	45.6	9.6	26.9	20.4	6.5	54.0	27.0	27.0	73.0	75.0	2.0	4.5	4.0	0.5	6.1	4.2	1.9
Average ±SD	56.3 ±6.8	46.8 ±5.7	9.5±6.7	61.1 ±7.6	52.9 ±6.4	8.1±6.1	25.2 ±3.2	22.5 ±4.8	2.7±2.6	59.3 ±12.6	31.2 ±9.8	28.1±10.3	60.0 ±22.3	67.8 ±25.2	7.8±22.8	6.5 ±3.7	4.9 ±5.7	1.6±2.4	6.1 ±0.9	4.1 ±1.6	2.0±1.9

Correction: is the difference value between the preoperative and postoperative values. DRUJ, distal radioulnar joint; Pre-Op., preoperative, Post-Op., postoperative.

Figure 5



Functional outcomes. (a) Preoperative left wrist. (b) Postoperative at removal of stitches. (c) Improved grip, (d, e, f) wrist movements and esthetic satisfaction. (g, h) Postoperative bilaterally operated patient, powerful fist. (i, j) Wrist movements and esthetic satisfaction improved. (k) Notable improved supination.

Vickers [10,21], thought to tether the medioradial metaphysis and TFCC to the volar surface of the lunate. The ligament restricts the volar–ulnar growth of the radius by exerting a compressive effect on the physis, hence contributing to the deformity, and resulting in the characteristic radiographic flame-shaped notch at its radial origin, and a bony bar bridges the epiphysis and metaphysis [11,20].

The volar radial growth disturbance may eventually lead to a progressive, three-dimensional wrist deformity including radiocarpal joint and DRUJ malalignment [22] since the radial deformity not only causes palmar–ulnar subluxation of the carpus but also alters the normal orientation of the sigmoid notch, resulting in DRUJ incongruity and subluxation with emergence of ulnar-sided wrist pain, deformity and limited forearm rotation [23]. There is no consensus on the best surgical treatment for symptomatic cases, with studies limited by small numbers and length of follow-up [23]. Several authors have managed MD with a radial osteotomy only (usually an open wedge osteotomy with bone graft) [2,18,24]; others advocated distal radial dome osteotomy with Vickers ligament surgical release [12].

Hence, all structural components of the wrist should be considered to improve pain, motion and esthetic appearance, multiplanar deformity correction and restoring the DRUJ congruity (re-optimization). BCWO enables optimal repositioning of the distal radial articular surface in both coronal and sagittal planes since the radial triangular component of BCWO creates volar angulation accompanied by dorsal shift of the distal radial fragment, carpus and hand, correcting the volar tilt and radiocarpal joint, while the dorsal triangular component creates ulnar angulation proximal to Vickers ligament radial origin, correcting the radial articular surface ulnar tilt and theoretically descent the ligament origin distally and subsequently release the lunate from impingement against medioradial metaphysis and the TFCC, providing ease to gain nearer between the distal radial fragment and ulna, and permit for closing DRUJ leisure by temporary fixation with dual K-wires while gently compressing the joint in the achieved new component correlation, which would enable the re-optimization of DRUJ toward normal biomechanical orientation.

Although Vickers ligament surgical resection is used, either alone [21] or in conjunction with different surgical techniques in the literature, such as excision and physiolysis either with fat interposition [11], or with staples placed across the unaffected radial physis to temporarily curtail its growth [8], the benefits of ligament release are largely conjectural [25]. However, release of the Vickers ligament is believed to prevent thinning of the radial physis caused by compression, thereby preventing further radial deformity with continued growth [25], and may aid carpal advancement [10]. Our proposal, of relaxing and releasing the tethering effect of Vickers ligament upon the lunate indirectly by having the site of our osteotomy proximal to the ligament origin and the subsequent distal subsidence of its origin, would achieve the same goal and save its direct volar surgical excision, with possible surgical and vascular trauma to already abnormal distal radial physis.

Ligament healing does not necessarily require rigid temporary fixation since the ligament and muscle attachment healing process can continue for years after the temporary stabilization [26]. Hence, in all our patients, DRUJs were temporally fixed for 12 weeks with the aim of achieving sustained DRUJ re-optimization through healing of the ligaments within the new DRUJ component correlation, together with the process of osteotomy healing of the redesigned distal radial alignment in both coronal and sagittal planes. The DRUJ gap at the latest follow-up radiographs showed a 32.5% overall improvement, by an average of 2.0±1.9 mm for the entire series (P=0.002), with no regression during the follow-up period.

Our six patients (eight wrists) underwent an average follow-up of 22 months. Pain during daily activities decreased significantly and joint mobility improved, with a satisfactory cosmetic appearance. In one wrist, pain occurred on the ulnar side during sustained activity, predominantly while writing. Clinical improvements in supination and wrist extension toward normal were 56.7 and 68.5% from the mean preoperative values of 50.6±24 and 44.7±16, respectively (P=0.02 and 0.004, respectively), whereas the differences in the mean pronation and flexion were not remarkable. No loss of motion was observed with any wrist movement parameter. Although the functional outcomes echo the conclusions of other complicated surgical techniques of osteotomy and ligament release in a comparable follow-up period [27,28], long-term follow-up is imperative to monitor recurrence of radial deformity and DRUJ incongruity.

The standardized normal radiological values are, for instance as follows: lunate subsidence in a 15-year-old girl is 7.0 mm, radial inclination is 14° and lunate-covering ratio is 70–100% on PA view [4]. However, values in the literature remained different from normal in MD treatment radiological outcomes since radiographic improvements toward normal are the main concern. Our patients showed improvements in the ulnar tilt (Radial Slope), lunate fossa angle and lunate subsidence by an overall 17, 13, and 24.6% from preoperative mean values toward normal, respectively, while the lunate coverage ratio increased by 13% from the preoperative mean percentage (60%±22.26) and radial inclination decreased by 47% of the preoperative mean angle in all the series.

Conclusion

This combined technique seems to be safe and can reliably restore hand–forearm alignment with respect to DRUJ re-optimization through the redesigned distal radial alignment in both coronal and sagittal planes and improve joint congruity, with promising functional and esthetic outcomes.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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