Measurement of compensatory wrist joint rotation using three-dimensional motion analysis in patients with unilateral proximal congenital radioulnar synostosis

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ABSTRACT

Objective: This study aims to investigate compensatory rotational movements of the wrist joint in patients with proximal congenital radioulnar synostosis (CRUS), using a valid and reliable three-dimensional (3D) motion analysis technique.

Methods: A total of 26 patients (6 females, 14 males; mean age=15.3 years; and age range=6-32 years) who were diagnosed with unilateral proximal CRUS but were not operated were enrolled in this study. Patients were then categorized into 2 groups: Group I included 5 patients younger than 10 years, and Group II included 15 patients older than 10 years. Eighteen light-reflective skin markers were placed on the bony landmarks of both upper limbs, and both distal forearms were fixed using a U-shaped device to minimize forearm rotation. Each patient grasped the handle of an instrument that used a goniometer to measure wrist rotation; maximal passive pronation and supination angles of the wrist were measured in this manner and also using 3D motion analysis.

Results: There was a significant correlation between measurements by the goniometer and 3D motion analysis (r=0.965, p<0.001). The test-retest reliability of the 3D motion analysis was acceptable for both the affected side (ICC=0.992) and the contralateral normal side (ICC=0.997) with low standard measurement errors (1.3° and 0.8°, respectively). Although no significant difference was observed in the range of the wrist rotation between the affected and contralateral sides in Group I (p=0.686), there was a significant difference in the wrist rotation between the affected and contralateral sides in Group II (p<0.001). Further, the pronation angle of the wrist joint was significantly larger in the affected side than that in the contralateral normal side in Group II (p<0.001).

Conclusion: The 3D motion analysis technique seems to be a valid and reliable method to measure the rotation of the wrist joint. Unilateral proximal CRUS patients older than 10 years of age may develop rotational hypermobility of the wrist joint compared to the contralateral normal side as a compensatory phenomenon.

Level of Evidence: Level III, Diagnostic Study

Introduction

Proximal congenital radioulnar synostosis (CRUS) is a rare congenital anomaly in which the proximal portions of the radius and ulna are fused, thereby limiting forearm rotation (1). In most cases, the forearms are fixed in a position ranging from neutral rotation to considerable pronation. The frequency of bilateral involvement is 50% (2, 3). When the affected forearm is fixed at greater than 60° of pronation in patients with bilateral involvement or at extreme pronation in unilateral involvement, difficulty in the performance of daily activities is expected (1, 2). Thus, in cases with forearm fixed at pronation exceeding 60° and disabilities in activities of daily life, derotational osteotomy is recommended to place the forearm in a more functional position (1, 4-7). However, disability is seldom evident in cases with mild deformities (8-10). The ipsilateral shoulder and wrist, combined with the opposite limb, can compensate for the fixed pronation position of the affected forearm. Ogino and Hikino (8) measured compensatory movements around the affected wrist using a goniometer and found that compensatory movements around the wrist were evident in almost all cases of total ankylosis. They measured the compensatory motion around the wrist using the palm position, however, they did not measure the true rotational angle of the wrist joint. Kasten et al. performed a three-dimensional (3D) motion analysis of compensatory shoulder and elbow movements in patients with radioulnar synostosis engaged in activities of daily living (ADL) (10). However, they did not evaluate the compensatory wrist rotational movements in those patients. The aim of this study is to measure the rotational angle of the wrist joint in patients with unilateral proximal CRUS using a valid and reliable 3D motion analysis technique.

Materials and methods

We reviewed all patients who were diagnosed with unilateral proximal CRUS, from January 2014 to December 2018, but were not operated because of this condition. The Institutional Review Board of our hospital reviewed and approved the study (IRB no. 1604-175-944). The inclusion criteria were as follows: (1) diagnosis of unilateral proximal CRUS, (2) no history of operation on the CRUS, and (3) provision of written informed consent to participate in this study. Patients with a history of upper extremity trauma on
the same side and those with associated other congenital upper extremity anomalies or syndromic conditions, which have generalized ligament laxity, were excluded. Ultimately, 20 patients with unilateral proximal CRUS (6 females, 14 males; mean age: 15.3 years [range: 6–32 years]) were enrolled (Table 1). The right side was affected in 11 patients, and the left in 9. The average fixed forearm position was 21° of pronation (range: 75° of pronation to 20° of supination). According to Cleary and Omer classification (11), there were 12 patients with type III (Figure 1), 6 with type II, and 2 with type IV. The patients were divided into 2 groups: Group I included 5 patients younger than 10 years, and Group II included 15 patients older than 10 years.

Experimental setup
Eighteen light-reflective skin markers (Motion Analysis, Santa Rosa, CA, USA) were placed on the bony landmarks of the upper limbs (Table 2, Figure 2). We manufactured a device that could measure the passive wrist rotational angle. Each patient was asked to sit comfortably on a height-adjustable stool with the hips and knees flexed at 90° and both feet placed flat on the floor. Initially, the shoulder was in 0° of abduction, flexion, and rotation, and the elbow was in 90° of flexion. To prevent forearm rotation, the distal forearm was fixed using the device. Each patient was asked to grasp the handle while the examiner grasped the opposite handle (Figure 3). The examiner rotated the handle clockwise and counterclockwise until the point where the resistance against the rotation was felt. We measured the maximal passive pronation and supination angles using the goniometer that was attached to the device. During the measurements, we evaluated the positions of hand segment with respect to the upper arm segment using the 3D motion analysis. Although we fixed the forearm during the measurements, the movement of the forearm was most likely present. Thus, the angles measured using the goniometer and the values of hand segment with respect to the upper arm segment using the 3D motion analysis were noted as the sum of forearm and wrist rotations. To assess the criterion-related validity of the 3D analysis of wrist rotational motion, we compared the goniometric measurements with the rotational angles of the hand segment with respect to the upper arm segment through 3D motion analysis. We defined the rotational angle of the wrist joint as the rotational angles of the hand segment with respect to the forearm segment. The maximal passive pronation and supination angles in the wrist joint were measured using 3D motion analysis. These measurements were repeated 3 times. The length of the test-retest interval was 1 week. The test-retest reliability of the 3D motion analysis for the measurement of wrist rotation was evaluated.

Upper limb model
To evaluate the relative movement between the forearm and hand segment (wrist motion), we modified the biomechanical upper limb model of Rab et al., which has 3 segments (upper arm, forearm, and hand) on each arm (12). In the model of Rab et al., hand and forearm segments shared the same two markers which were attached on the distal radial and ulnar styloid processes (12). In this study, to accurately measure the wrist rotation angle, we added wrist wand markers and separated the hand segment from the forearm segment. The forearm segment was composed of 3 markers: the olecranon tip, radial, and ulnar styloid processes. Hand segment consisted of 3 markers: the 3rd metacarpal head, Mid-carpal center (MC), and 3CP markers (Figure 4). The MC marker was attached on the 3rd metacarpal base. The 2CP marker was the most superior marker, the 3CP marker was the most lateral marker, and the 4CP marker was the most inferior marker. The kinematic evaluation of the wrist rotation was defined as the rotation of the hand segment relative to the forearm segment. As in the model of Rab et al., the axes were defined in an anatomically neutral position, as follows: The X-axis directed laterally to the right, the Y-axis directed anteriorly, and the Z-axis directed superiorly (12). Motion was measured in the order of the sagittal, coronal, and transverse planes (X-Y-Z; the Eulerian sequence). The starting point was the initial fixed position, not the anatomically forearm neutral

![Figure 1. a, b. Case no. 1. Left elbow an teroposterior (AP) and lateral radiographs of a 6-year-old boy, showing the proximal radioulnar synostosis with posterior dislocation of the hypoplastic radial head](image-url)

### HIGHLIGHTS

- The ipsilateral shoulder and wrist, combined with the opposite limb, can compensate for the fixed position of the affected forearm: functional tolerance is excellent, especially when the forearm is fixed in a neutral position.
- Three-dimensional motion analysis technique is a valid and reliable method to measure the rotation of the wrist joint.
- The rotational hypermobility of the wrist joint compared to the contralateral normal side was observed in unilateral proximal CRUS patients, who were older than 10 years.

### Table 1. Demographic data of patients

| Case Number | Gender/Age (year) | Affected side | Position of affected forearm (degrees) | Radiographic Classification (Cleary & Omer classification) |
|-------------|------------------|---------------|----------------------------------------|----------------------------------------------------------|
| 1           | M/6              | Lt            | 30 pronation                           | III                                                      |
| 2           | F/6              | Lt            | 75 pronation                           | III                                                      |
| 3           | F/7              | Rt            | 20 supination                          | II                                                       |
| 4           | F/8              | Lt            | 15 pronation                           | III                                                      |
| 5           | F/9              | Rt            | 35 pronation                           | III                                                      |
| 6           | M/11             | Rt            | 20 pronation                           | II                                                       |
| 7           | M/11             | Rt            | 15 pronation                           | III                                                      |
| 8           | M/11             | Rt            | 25 pronation                           | III                                                      |
| 9           | F/12             | Rt            | 25 pronation                           | II                                                       |
| 10          | M/12             | Rt            | 10 pronation                           | III                                                      |
| 11          | M/15             | Lt            | 30 pronation                           | II                                                       |
| 12          | F/15             | Lt            | 30 pronation                           | III                                                      |
| 13          | M/20             | Lt            | 0 neutral                             | II                                                       |
| 14          | M/20             | Rt            | 45 pronation                           | IV                                                       |
| 15          | M/21             | Lt            | 0 neutral                             | II                                                       |
| 16          | M/21             | Rt            | 10 pronation                           | III                                                      |
| 17          | M/21             | Rt            | 20 pronation                           | IV                                                       |
| 18          | M/22             | Lt            | 10 pronation                           | III                                                      |
| 19          | M/26             | Rt            | 30 pronation                           | III                                                      |
| 20          | M/32             | Lt            | 15 pronation                           | III                                                      |
| Mean        |                  |               |                                        |                                                          |

M: male; F: female; Rt: right; Lt: left
The average pronation and supination angles from the fixed position of forearm, measured using the goniometer, were 29° and 30° on the affected side and 25° and 35° on the contralateral normal side, respectively. The average hand segment pronation and supination angles with respect to the upper arm segment, measured by the 3D motion analysis, were 27° and 27° on the affected side and 24° and 33° on the contralateral normal side, respectively. There was a significant correlation between the measurements obtained using goniometer and 3D motion analysis (r=0.985, p<0.001).

In Group I, the average pronation and supination of the wrist joint, measured by 3D motion analysis, were 26° and 29° on the affected side and 28° and 30° on the contralateral normal side, respectively (Table 3, Figure 6). In Group II, these were 18° and 17° on the affected side and 28° and 30° on the contralateral normal side, respectively. There was a significant difference in the range of the wrist rotation between the affected and contralateral sides in Group I (p=0.001). The pronation angle of the wrist joint was significantly larger in the affected side than that in the contralateral normal side in Group II (p=0.001).

The test-retest reliability (ICC) of 3 measurements of wrist rotational motion was 0.992 on the affected side and 0.997 on the contralateral normal side. The SEM of the measurement was 1.3° on the affected side.
side and 0.8° on the contralateral normal side. Therefore, the test-retest reliability of 3D motion analysis for the measurement of wrist rotation was proved to be acceptable with low measurement error.

Discussion

In this study, compensatory rotation in the wrist joint was measured using 3D motion analysis in patients with unilateral CRUS. Although Simmons et al. described the hypermobility of the wrist joint in patients with CRUS in 1983, there has been no study measuring the rotation of the wrist joint accurately (14). Among the patients enrolled in this study, wrist joint hypermobility was observed in the contralateral normal side of younger patients. Antonio and Magalhaes reported that approximately 25-50% of children younger than 10 years of age have some degree of hypermobility (15). Thus, we divided our patients into 2 groups according to their age. In 3 patients of Group I, the rotational range of the wrist joint was larger in the contralateral normal side than the affected side. In Group II, the average pronation and supination of the wrist joint were measured as 18° and 17° in the affected side and as 10° and 16° in the contralateral normal side. The pronation of the wrist joint in the affected side was 80% larger than that in the contralateral side. Although 8° may be regarded as minimal difference, it should not be overlooked considering that rotation is not the primary motion of the wrist joint. Wrist joint has been regarded as a condyloid joint with 2 degrees of freedom where the main motions are flexion-extension and radial and ulnar deviation. However, Ritt et al. found that this joint has 3 degrees of freedom, which includes at least 10 to 20° of rotation with less pronation laxity than supination (16). The results are similar to the findings of this study. We measured the rotational angles of the wrist joint while the subject grasped the handle of the instrument. When clenching the fist, the axial compressive force locks proximal carpal bones into shallow scaphoid and lunate fossa of the distal radius. Therefore, if we could measure the rotation of the wrist with all fingers extended, the rotational angles might be larger than those in this study.

It remains unclear why wrist pronation is increased in Group II patients. Simmons et al. described that the rotational hypermobility of the wrist was presumably due to ligamentous laxity secondary to continued rotational stress on the wrist in the absence of forearm rotation (14). Nakasone et al. evaluated the deformities of the radius and ulna using reconstructed three-dimensional images of radius and ulna (17). The radius was 18° internally rotated, and the ulna was 30° internally rotated. The rotational deformities of the radius and ulna correlated strongly with the degree of fixed pronation. These internal rotation deformities of the forearm bones imply that pronation load might be applied on the distal forearm and wrist, although we are not aware of the exact origin of the load. If any muscle acts as a supinator or pronator of the wrist joint, it originates...
from distal humerus and inserts on the metacarpal or phalangeal bones. Among the muscles, only flexor carpi radialis (FCR) can act as a pronator of the wrist joint. There is no muscle which can act as a supinator of the wrist joint. Thus, FCR might cause the pronation laxity of the wrist. Another possible reason is that pronation is more common than supination in daily and sports-related activities. Typing, eating with a knife and fork, riding a bicycle, using a computer mouse, throwing a ball, using door handles, or driving a car are all pronation activities.

There have been several reports describing compensatory motion at adjacent joints in patients with proximal CRUS. Cleary and Omer (11) reported the natural history of proximal CRUS in 23 patients who did not undergo surgery. Eight patients perceived no limitation, 14 reported slight limitation, and 1 reported moderate limitation. Although many patients exhibited increased wrist rotation to compensate for the lack of pronation and supination, the authors did not measure the rotation of the wrist joint objectively. Yammine et al. retrospectively analyzed 46 forearms of 37 CRUS patients; wrist hypermobility (flexion-extension and radial-ulnar deviation) was observed in all cases, however, the rotation angle of the wrist was not measured (9). Our study is the first to verify the presence of rotational hypermobility of the wrist joint.

Kasten et al. used 3D motion analysis to evaluate compensatory movements of the shoulder and elbow in CRUS patients performing 10 ADL (10). The principal compensatory movements were shoulder internal/external rotation during 5 ADL, shoulder abduction/adduction and elbow flexion/extension during 3 ADL, and shoulder flexion/extension during 2 ADL. Although the authors measured the radial and ulnar abduction, dorsiflexion, and palmar flexion of the wrist joint, they did not assess the compensatory rotation of the wrist joint. In this study, we focused on the rotation of wrist joint and measured the wrist rotation and forearm rotation separately via 3D motion analysis. When evaluating compensatory wrist rotation, it is not strictly necessary to model the upper arm segment. However, as we aimed to evaluate the correlation between the measurements using the goniometer and the 3D motion analysis technique, we included the upper arm segment in our model. A significant correlation was evident between the measurements obtained using the goniometer and 3D motion analysis. The test-retest reliability of the latter analysis was acceptable.

This study has several limitations. First, the patients were demographically heterogeneous, where age and sex could influence the joint flexibility. Second, we did not measure active wrist rotational angle; young patients cooperated poorly in performing active range of motion. Third, we defined the endpoints of maximal pronation and supination as the point where the resistance against the rotation was felt. However, the endpoint of wrist rotation is subjective during the measurement. Lastly, we did not evaluate compensatory motion of the elbow or shoulder joints.

In conclusion, 3D motion analysis is a valid and reliable method to measure the rotation of the wrist joint and that of the forearm separately. Rotational hypermobility of the wrist joint was observed in proximal CRUS patients who were older than 10 years.

Ethics Committee Approval: Ethics committee approval was received for this study from the Institutional Review Board of Seoul National University Hospital (IRB No. 1804-175-944).

Informed Consent: Written informed consent was obtained from patients who participated in this study.

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