Carpal tunnel syndrome associated with Kienböck disease

Takaaki Shinohara¹, Ryogo Nakamura¹, Etsuhiro Nakao¹ and Hitoshi Hirata²

¹Nagoya Hand Surgery Center, Chunichi Hospital, Nagoya, Japan
²Department of Hand Surgery, Nagoya University Graduate School of Medicine, Nagoya, Japan

ABSTRACT

We retrospectively reviewed 12 patients (3 men and 9 women, with a mean age of 72 years) who were surgically treated for carpal tunnel syndrome associated with Kienböck disease. All patients except 1 were incidentally diagnosed with Kienböck disease and had little or no wrist pain. Radiographic tests revealed advanced Kienböck disease in all patients. Intraoperative findings indicated that the site of maximum compression on the median nerve was located at the level of the carpal tunnel inlet in 11 patients, and the volar dislocated fragment of the lunate was located proximally adjacent to the floor of the carpal tunnel inlet. This disorder is most prevalent in elderly women, and even advanced Kienböck disease can present without wrist pain. Our findings suggest that palmar protrusion of the lunate may be the primary cause of carpal tunnel syndrome associated with Kienböck disease.

Key Words: Kienböck disease; carpal tunnel syndrome; lunate; median nerve; elderly

This is an Open Access article distributed under the Creative Commons Attribution-NonCommercial-NoDerivatives 4.0 International License. To view the details of this license, please visit (http://creativecommons.org/licenses/by-nc-nd/4.0/).

INTRODUCTION

Kienböck disease involves idiopathic avascular necrosis of the carpal lunate, and was first reported by Robert Kienböck in 1910.¹ The etiology and natural history of this condition is poorly understood, and it is generally accepted that initial sclerosis of the lunate ultimately leads to collapse and osteoarthritis as the disease progresses. The notion that carpal tunnel syndrome (CTS) occurs in conjunction with Kienböck disease has previously been proposed.² However, there are few reports about CTS associated with Kienböck disease,³⁴ and the clinical conditions for such an association have not been defined. Moreover, it remains unclear how the volar dislocated fragment of the lunate influences the median nerve.

The purpose of this study was to investigate the clinical characteristics of the lunate, median nerve, and transverse carpal ligament, as well as the relationships between them, in CTS associated with Kienböck disease.

METHODS

This study included 12 patients (3 men and 9 women) who had been surgically treated for...
Takaaki Shinohara et al.

Table 1  Patient characteristics

| Patient no. | Sex | Age (years) | The stage of Kienböck’s disease | Wrist pain | The duration of CTS (months) | DML of the affected side (ms) | DML of the unaffected side (ms) | LPR | Surgical outcome |
|-------------|-----|-------------|---------------------------------|------------|-----------------------------|-----------------------------|-----------------------------|-----|-----------------|
| 1           | M   | 67          | IV                              | None       | 60                          | NE                          | 5.6                         | –   | Fair            |
| 2           | F   | 62          | IIIb                            | Mild       | 8                           | NE                          | 3.7                         | 0.42 | Excellent       |
| 3           | M   | 67          | IV                              | None       | 5                           | NE                          | 5.7                         | 0.73 | Good            |
| 4           | F   | 77          | IV                              | None       | 6                           | NE                          | 5.7                         | 0.60 | Excellent       |
| 5           | F   | 82          | IIIb                            | None       | 6                           | NE                          | NE                          | 0.52 | Excellent       |
| 6           | F   | 63          | IV                              | None       | 36                          | NE                          | 5.4                         | 0.51 | Excellent       |
| 7           | F   | 69          | IV                              | None       | 36                          | NE                          | 4.4                         | 0.72 | Good            |
| 8           | F   | 67          | IV                              | Moderate   | 120                         | 5.8                         | 5.1                         | 0.71 | Fair            |
| 9           | F   | 87          | IIIb                            | None       | 2                           | 8.0                         | 4.1                         | 0.54 | Good            |
| 10          | F   | 78          | IV                              | None       | 18                          | 6.1                         | 4.2                         | 0.61 | Good            |
| 11          | M   | 76          | IIIb                            | None       | 12                          | NE                          | 4.4                         | 0.62 | Excellent       |
| 12          | F   | 67          | IIIb                            | None       | 24                          | 6.2                         | 4.7                         | 0.52 | Excellent       |

CTS: carpal tunnel syndrome, DML: distal motor latency, NE: not evoked, LPR: lunate protrusion ratio

CTS associated with Kienböck disease at our 2 hospitals between 1996 and 2012 (Table 1). All protocols for this retrospective study were approved by the Institutional Review Board of each participating hospital. The mean age ± standard deviation (SD) at the time of surgery was 72 ± 8 years (range, 62–87 years); the left and right hands were affected in 4 and 8 patients, respectively. The mean duration of post-operative follow-up was 35 months (range, 10–84 months). All 12 patients had numbness and thenar muscle atrophy in the affected hand. Radiographic evaluations were performed on the anteroposterior and lateral wrists in order to diagnose bone abnormalities that may cause CTS. There was no history of Kienböck disease in 11 of the patients; the 12th received surgical treatment 40 years ago although the details of the procedure were unclear. Hence, 11 patients were incidentally diagnosed with Kienböck disease. Motor nerve conduction studies were carried out in all patients to confirm the diagnosis of CTS, which was then treated with open carpal tunnel release in all patients. Lunate resection and vascularized pisiform transfer for Kienböck disease was performed simultaneously in one patient who had moderate wrist pain.

Detailed information regarding the status of wrist pain on the first visit, the interval between onset of numbness and consultation, distal motor latency (DML) of the median nerve in both wrists, intraoperative findings, and surgical results for CTS were extracted from medical records. Pain was classified into four levels of severity: no pain, mild occasional, moderate (tolerable), and severe (intolerable) in accordance with the Mayo Modified Wrist Score.5) The DML was diagnosed as abnormal if it exceeded 4.5 ms. CTS was diagnosed based on clinical findings, physical examination, and nerve conduction studies. The outcome of surgery for CTS was graded according to the classification system of Kelly et al.6) as follows: Excellent (complete relief of symptoms), good (persistence of occasional minor symptoms), fair (some constant or annoying symptoms), and poor (symptoms unchanged or worse).

Posteroanterior and lateral radiographic images were acquired for radiographic evaluations. Radiographic staging of Kienböck disease was performed according to Lichtman’s classification.7)
CTS associated with Kienböck disease

On the lateral radiographic image, two longitudinal lines along the long axis of the radius that passed through the volar margin of the lunate (X1) and the capitate head (X2) were drawn. The anteroposterior diameter (a) of the capitate head and the orthogonal distance (b) between X1 and X2 were measured (Fig. 1). The lunate protrusion ratio (LPR) was determined by the ratio of b to a. We selected 100 patients (25 men and 75 women; mean age ± SD = 72 ± 8, range, 62–87 years) with idiopathic CTS who did not have Kienböck disease and who underwent carpal tunnel release by matching based on age and sex. The LPR was calculated for patients with CTS associated with Kienböck disease (the Kienböck group) and for idiopathic CTS patients (the idiopathic group); LPR values were compared between these 2 groups by using the Mann-Whitney’s U test. A p-value <0.05 was considered significant.

RESULTS

The clinical data and results are shown in Table 1. There were 2 patients who noted wrist pain at their initial consultation for CTS (one with mild pain and the other with moderate pain). The mean duration of CTS symptoms was 28 months (range, 2–120 months). Eight patients showed absence of compound muscle action potential and the other 4 showed a delayed DML of greater
than 5.8 ms in the affected wrist. For unaffected wrists, one patient showed absence of compound muscle action potential and 6 showed a delay in DML greater than 4.5 ms. Intraoperative findings indicated that the site of maximum nerve compression was located at the level of the carpal tunnel inlet in 11 patients and the central part of the carpal tunnel in 1 patient. The volar capsule was not ruptured in any of the wrists, and bone prominence was located proximally, adjacent to the floor of the carpal tunnel inlet (Fig. 2). In 5 of 12 patients, radiographic evaluations were performed by placing a marker on the site of maximum compression during surgery in order to investigate the relationship between the lunate and the median nerve. The marker was located distally adjacent to the lunate in all 5 patients (Fig. 3).

The surgical outcomes were excellent in 6 patients, good in 4, and fair in 2. Two patients with fair results had experienced CTS symptoms for more than 5 years.

Radiographic staging of Kienböck disease were stage IIIB in 5 and stage IV in 7 patients. In 1 patient of the Kienböck group, LPR was not measured because the accurate lateral radiograph was missing. The mean LPR ± SD was 0.59 ± 0.10 (range, 0.42–0.73) in the Kienböck group and 0.21 ± 0.07 (range, 0.12–0.53) in the idiopathic group. The LPR of the Kienböck group was significantly higher than that of the idiopathic group (p < 0.001).

**DISCUSSION**

Beckenbaugh et al.\textsuperscript{2} described the natural history of 46 patients with Kienböck disease; 5 of these patients had complained of CTS symptoms. Otherwise, there are few reports of CTS associated with Kienböck disease. Taniguchi et al.\textsuperscript{3} reported 14 patients who were incidentally diagnosed with Kienböck disease on radiographic examinations obtained for other reasons such as CTS (4 patients), Colles’ fracture, and pseudogout attack. All 14 patients had stage IV disease and were not experiencing wrist pain. Four patients with CTS were elderly women (62–72 years). Hayashi et al.\textsuperscript{4} noted 17 patients with CTS who were incidentally diagnosed with Kienböck disease; their mean age was 69 years and 70.6% were female. All patients showed a delayed DML in the affected wrist, and 8 patients had a delayed DML in the unaffected wrist. Comparison of the clinical data for Kienböck disease patients with and without CTS revealed that mean age, frequency of female sex, frequency of mild or no wrist pain, and frequency of stage IV disease were all significantly greater in CTS compared to non-CTS patients. In our study, women were predominant (75%), and all but 2 patients had no wrist pain. Radiographic findings revealed advanced Kienböck disease (Lichtman stage IIIb and IV) in all patients. Seven of 12 patients showed a delayed DML greater than 4.5 ms on the unaffected side. Our data had similar characteristics to those reported by Taniguchi et al.\textsuperscript{3} and Hayashi et al.\textsuperscript{4} Based on the above results, there is a possibility that these patients are related to the underlying subclinical CTS.

In idiopathic CTS patients, the site of maximum compression of the median nerve is usually located at the carpal tunnel outlet rather than the inlet.\textsuperscript{8,9} However, our intraoperative finding revealed that this site was located at the carpal tunnel inlet level in 11 of 12 patients, and bone prominence was located proximally and adjacent to the floor of the carpal tunnel inlet in all patients. The LPR was calculated to evaluate the degree of the prominence of the lunate bone after such prominence was detected, and LPR values were compared between the Kienböck group and the idiopathic group. The mean LPR of the Kienböck group was significantly higher than that of the idiopathic group. Therefore, we attribute the cause of nerve entrapment at the carpal tunnel inlet to the fact that the transverse carpal ligament is situated just distal to the site in which the median nerve is pushed against the volar side, judging by the volar prominence of the fragmented lunate bone (Fig. 4). Hence, we consider the palmar protrusion of the lunate to be the
Fig. 2 The site of maximum compression of the nerve (arrowhead) is located at the level of the carpal tunnel inlet; bone prominence of the lunate (asterisk) is present in its adjacent proximal location. Arrow: Proximal end of the transverse carpal ligament.

Fig. 3 The marker on the site of maximum compression during surgery is located distally, adjacent to the volar dislocated fragment of the lunate.
primary cause of CTS associated with Kienböck disease. Because the unaffected wrist frequently showed a delayed DML (7/12), idiopathic CTS may also influence the onset of this disorder.

All patients except 1 underwent only open carpal tunnel release because the volar capsule was not ruptured and the patients had little or no wrist pain. Our surgical procedure provided good outcomes (except in 2 patients who had experienced CTS symptoms for more than 5 years), and CTS did not recur during the follow-up period. Therefore, we recommend only open carpal tunnel release for CTS associated with Kienböck disease if there is no moderate or severe wrist pain.

There are limitations to this study, including its retrospective nature and the small number of patients investigated. Moreover, all the surgeries were not performed by the same surgeon, and the follow-up period was relatively short. Lastly, changes in carpal alignment, such as carpal height, were not examined.

Nevertheless, our findings serve to better elucidate the relationship between CTS and Kienböck disease.

CONFLICT OF INTEREST

The authors declare that they have no conflicts of interest.
CTS associated with Kienböck disease

REFERENCES

1) Kienböck R. Über traumatische malzie des mondbeins und ihre folgezustände: entartungsformen und kompressionsfrakturen. Fortschr Geb Roentgenstr, 1910; 16: 77–103.
2) Beckenbaugh RD, Shives TC, Dobyns JH, Linscheid RL. Kienböck’s disease: the natural history of Kienböck’s disease and consideration of lunate fractures. Clin Orthop Relat Res, 1980; 149: 98–106.
3) Taniguchi Y, Nakao S, Tamaki T. Incidentally diagnosed Kienböck’s disease. Clin Orthop Relat Res, 2002; 395: 121–127.
4) Hayashi M, Makoto M, Kato H. Carpal tunnel syndrome associated with underlying Kienböck’s disease. J Hand Surg Eur Vol, 2015; 40: 638–639.
5) Cooney WP, Linscheid RL, Dobyns JH. Triangular fibrocartilage tears. J Hand Surg Am, 1994; 19: 143–154.
6) Kelly CP, Pulisetti D, Jamieson AM. Early experience with endoscopic carpal tunnel release. J Hand Surg Br, 1994; 19: 18–21.
7) Lichtman DM, Degnan GG. Staging and its use in the determination of treatment modalities for Kienböck’s disease. Hand Clin, 1993; 9: 409–416.
8) Azami A, Maleki N, Anari H, Iranparvar Alamdari M, Kalantarhormozi M, Tavosi Z. The diagnostic value of ultrasound compared with nerve conduction velocity in carpal tunnel syndrome. Int J Rheum Dis, 2014; 17: 612–620.
9) Mondelli M, Filippou G, Gallo A, Frediani B. Diagnostic utility of ultrasonography versus nerve conduction studies in mild carpal tunnel syndrome. Arthritis Rheum, 2008; 59: 357–366.
10) Yazdchi M, Tarzamani MK, Mkaeili H, Ayromlu H, Ebadi H. Sensitivity and specificity of median nerve ultrasonography in diagnosis of carpal tunnel syndrome. Int J Gen Med, 2012; 5: 99–103.