Pathology and Therapeutic Results in Patients with Intraosseous Ganglia of the Carpal Bone

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ABSTRACT

This study evaluated the pathology and therapeutic results of seven patients with intraosseous ganglia of the carpal bone. The mean age at the time of surgery was 27.6 years. The lesions were localized in the proximal carpal row in six patients and in the distal carpal row in only one. Surgical treatment was performed in all patients with good bone union.

None had pain during activity or at rest and no recurrence had occurred. The intraosseous ganglia in four patients was of the idiopathic type, and in the other three patients was of the penetrating type. Although intraosseous ganglia of the carpal bone is reported as a rare disease, there were 159 cases in the literature. The pathology was intra- or extraosseous development, showing variation, but most cases were localized in the proximal carpal row.

Keywords: Intraosseous ganglia, scaphoid, lunate, hamate

Patología y Resultados Terapéuticos en los Pacientes con Ganglión Intraóseo del Hueso Carpiano

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RESUMEN

Este estudio evaluó la patología y resultados terapéuticos de siete pacientes con ganglión intraóseo del hueso carpiano. La edad promedio en el momento de la cirugía fue 27.6 años. Las lesiones se localizaban en la fila proximal del carpo en seis pacientes y en la fila distal carpiana solamente en uno. El tratamiento quirúrgico se realizó en todos los pacientes con buena unión ósea. Ninguno tuvo dolor durante la actividad o el reposo, y no había tenido lugar recurrencia alguna. El ganglión intraóseo en cuatro pacientes fue de tipo idiopático, y en los otros tres pacientes fue de tipo penetrante. Aunque el ganglión intraóseo del hueso carpiano se informa como una enfermedad rara, se reportaban 159 casos en la literatura. La patología consistía en un desarrollo intra- o extraóseo, con variación, pero la mayoría de los casos se localizaban en la fila proximal carpiana.

Palabras claves: Gangliones intraóseos, escafoide, semilunar, unciforme

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INTRODUCTION

Intraosseous ganglia cysts of the carpal bone is an unusual disease. Regarding the cause of its development, there is the endogenous hypothesis suggesting a synovial membrane-like degeneration of intraosseous connective tissue, and the exogenous hypothesis, related to intraosseous invasion by synovial fluid or soft tissue ganglia, but no theory has been established. In this study, we report seven patients with intra-

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osseous ganglia cysts of the carpal bone with a literature review.

SUBJECTS AND METHODS

Postero-anterior and lateral radiographs were systematically performed on the patients with wrist pain. There were seven patients whose radiographic findings showed cystic radiolucent lesions surrounded by a sclerotic zone in the carpal bone. None of them was bilateral. There were three male and four female patients and the affected side was right in three and left in four. The age of the patients at the time of surgery was 16 to 47 years (mean: 27.6 years) and the post-operative follow-up period was six to 28 months (mean: 11.4

months). The preoperative symptoms were pain of the wrist joint on active motion in all patients but no resting pain, severe limitation in the range of motion of the wrist joint, sign of local infection or findings suggesting neurovascular disorder were noticed in any patient. These lesions were localized in the proximal carpal row in six of the seven patients: the scaphoid in five and the lunate in 1, and in the distal carpal row (the hamate) in only one patient. No apparent traumatic history was noted in any patient. However, only a 16-year old student with pathological lesion in the hamate had participated in Japanese kendo from 10 years old. Plain radiographic findings detected cystic radiolucent lesions surrounded by a sclerotic zone in the hamate (Fig. 1),







Fig. 1: Plain radiographs of patient No. 2. These findings showed a cystic radiolucent lesion (white arrow) surrounded by a sclerotic zone in the hamate

thus diagnosing a rare case of intraosseous ganglia in the hamate. Also, a 35-year old man with a pathological lesion in the scaphoid was engaged in manual labour. Plain radiographic findings detected a cystic radiolucent lesion surrounded by a sclerotic zone in the proximal third region of the scaphoid (Fig. 2). On T1-weighted MRI, a low-intensity





Fig. 2: Plain radiographs of patient No. 4. Plain radiographic findings detected a cystic radiolucent lesion (white dotted arrow) surrounded by a sclerotic zone in the 1/3 proximal region of the scaphoid.

region was present in the scaphoid. No apparent connection with an extraosseous region was noted on plain radiographic findings or MRI. But 3DCT detected a cortical defect of the scaphoid, and it was suspected to penetrate the cortex of the scaphoid on preoperative diagnosis (Fig. 3). Meanwhile, the





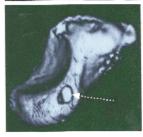


Fig. 3: MR images and 3DCT of Patient 4. On T1-weighted MR images, a low-intensity region was present in the scaphoid (white arrow). No apparent connection with an extraosseous region was noted on plain radiographic findings or MRI, but 3DCT detected a cortical defect of the scaphoid(white dotted arrow).

duration of pain of the wrist joint varied from 2 months to 5 years including pain-free periods.

Surgical treatment was performed in all patients, and comprised curettage of the pathological lesion followed by filling of the bone defect. Almost all surgical approach was performed using the palmal side incision. However, the two patients with the pathological lesion within the hamate and the proximal third of the scaphoid had an approach using the dorsal side incision. A jelly-like substance was noted in the lesion during surgery in all patients and the lesion was covered with membrane-like tissue. The membrane-like tissue consisted of fibrocytes without synovial lining cell. The iliac crest, distal radius and artificial bone were used for bone grafting without internal fixation in four patients respectively. Postoperative immobilization was applied for three weeks to patients with the intraosseous carpal lesion alone, and 4 weeks to those with connection outside of the carpal bone cavity.

RESULTS

The therapeutic results on the final examination are shown in Table 1. Bone union was achieved in all seven patients. None of these patients experienced pain during activity or at rest and no recurrence of ganglia had occurred. The patients could return to their original work. However, in the case of

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Table 1: Final therapeutic results

No.	Location	Age (years)	Grip (kg) (other side)	ROM*	OA**	Return to work (months)
1	Scaphoid	29	19.9 (17.0)	80–80°	None	1
2	Hamate	16	40.0 (43.7)	90–90°	None	1
3	Scaphoid	47	36.8 (34.1)	60-50°	Yes	2
4	Scaphoid	35	19.9 (17.0)	70–60°	None	2
5	Lunate	24	21.2 (19.9)	80–70°	None	1
6	Scaphoid	21	21.3 (22.5)	90–80°	None	1
7	Scaphoid	34	17.8 (20.2)	80–80°	None	1

*ROM: Degree of dorsiflexion and palmarflexion

one patient (No 3), osteoarthritic changes of the radiocarpal joint were observed on radiographic examinations. The grip strength and range of motion of the wrist joint was restored to the same level as that of the non-affected side. On the basis of the connection outside of the carpal bone cavity observed during surgery, we confirmed that the intraosseous ganglia in four patients were of the idiopathic type which mean no connection to outside of that bone, and that in the other three patients were of the penetrating type that is the cyst and stalk penetrated the cortex of the bone.

DISCUSSION

An intraosseous ganglia was termed a juxta-articular bone cyst by the WHO (1) and defined as a tumour that is localized in the subchondral bone adjacent to a joint, showing a cystic radiolucent lesion surrounded by a sclerotic zone in the bone on plain radiograph and histologically lacks synovial lining cell. In 1979, Schajowitz et al (2) investigated 88 cases of intraosseous ganglia regardless of the development site. All cases were located in the subchondral bone adjacent to a joint, and the patients ages ranged widely from 14 to 73 years. They classified the development mechanism into endogenous (idiopathic) and exogenous (penetrating) types based on the presence or absence of extraosseous connection, and found that the former and latter types accounted for 84% and 16% of cases, respectively. Hicks et al (3) and Byers et al (4) reported endogenous hypotheses, while Crabbe et al (5) and Crane et al (6) reported exogenous hypotheses: tumour is caused by invasion by extraosseuous soft tissue ganglia or synovial fluid, but no theory of intraosseous ganglia has been established. In intraoperative findings of our patients, the idiopathic type localized within the carpal bone was noted in four patients (scaphoid: 3, hamate: 1) and the penetrating type with apparent connection with the outside of the bone cavity was noted in three (scaphoid: 2, lunate: 1). There was no apparent difference in the preoperative findings between the two types, but the bone defect was large in one case of a penetrating type scaphoid lesion, and finally resulted in osteoarthritic changes.

In 1999, Uriburu *et al* (7) investigated 15 cases of intraosseous ganglia in the carpal bone (scaphoid: 6, lunate: 9), and found that 11 cases were of the idiopathic type with cortical bone defects, 6 were of penetrating type with communication with soft tissue ganglia, and postoperative recurrence occurred in one. In 2005, Van den Dungen *et al.* (8) investigated 51 cases of soft tissue ganglia that developed in the dorsum of the hand, and found intraosseous ganglia of the carpal bone communicating with the soft tissue ganglia in 24 cases. In 1995, Magee *et al.* (9) examined 400 patients with pain in the wrist joint with unknown pathology using multidirectional plain radiographs, bone scintigraphy, and MRI, and identified intraosseous ganglia in 15 cases as the cause of pain.

To our knowledge after 1972, intraosseous ganglia of the carpal bone reported in detail in Japan and abroad are shown in Figs. 4 and 5. There were five bilateral cases in

Japanese literatures

Bilateral: 5

Lunate 3, Scaphoid 1, Trapezium 1

Unilateral: 62

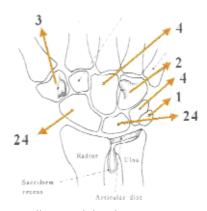


Fig. 4: Carpal intraosseous ganglia reported abroad.

Foreign literatures

Bilateral: 5

Lunate 3, Scaphoid 1, Hamate 1

Unilateral: 77

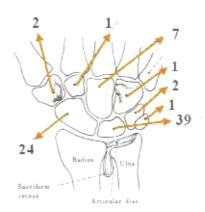


Fig. 5: Carpal intraosseous ganglia cases reported in Japan.

^{**}OA: Osteoarthritis

Foreign literatures

Bilateral: 5

Lunate 3, Scaphoid 1, Hamate 1

Unilateral: 77

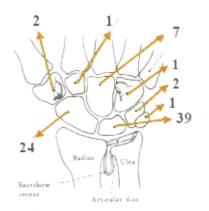


Fig. 5: Carpal intraosseous ganglia cases reported abroad.

each series, with 77 cases of unilateral cysts reported abroad, showing that the disease is not rare. Regarding the localization, the ganglia developed in the proximal carpal row, mainly in the lunate and the scaphoid, in more than 80% of all cases reported abroad and in Japan, while it rarely localized in the trapezoid, pisiform, and hamate. Intra-osseous ganglia are more likely to localize in the proximal row than in distal carpal row, which may be due to relatively vulnerable ligament connection between the carpal bones, which allow a wide range of motion during wrist joint

movement, and easily receives directly mechanical stress from the radius. In fact, the lesions were localized in the proximal carpal row in 6 of the 7 cases in our study.

CONCLUSION

Although intraosseous ganglia of the carpal bone are reported as a rare disease, there were 159 reported cases in the literature. For painful intraosseous ganglia, curettage of the lesion and bone grafting achieved satisfactory stable therapeutic results. The pathology was intra- or extraosseous development, but most cases were localized in the proximal carpal row.

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