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1 *Case Report*

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3 **Postoperative early load-bearing walking by an adult with**
4 **painful bilateral os intermetatarsea treated surgically: A Case Report**

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15 **Conflict of Interest**

16 The authors did not receive and will not receive any benefits and funding from any

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18

1 INTRODUCTION

2

3 The os intermetatarsium is a rare accessory bone of the foot that is usually
4 located between the bases of the first and second metatarsals. Most cases of os
5 intermetatarsia are symptomless [1]. Good results were reported in cases of painful
6 unilateral os intermetatarsium that were treated with surgery [2, 3]. By contrast, to the
7 best of our knowledge there have been no reports of simultaneous bilateral surgical
8 treatment in cases of bilateral os intermetatarsia, and there is a lack of knowledge
9 regarding how to approach postoperative therapy in such cases. The present case
10 report appears to be the first on the surgical treatment of bilateral os intermetatarsia
11 followed by successful postoperative early walking training.

12

13 **CASE REPORT**

14

15 A 22-year-old Japan Ground Self-Defense Force member with no remarkable
16 medical history complained of pain over the dorsum on both of his feet. From
17 childhood, he was aware of ridges on the dorsum on both feet and occasionally had
18 pain at these sites, but the symptoms had always been temporary. However, prior to
19 his seeking medical treatment, the symptoms had recurred for 6 months without an
20 obvious cause. He went to a clinic and received symptomatic treatment over a period
21 of 3 months, but because his symptoms did not improve, he came to our hospital.

22 A physical examination revealed bilateral bony prominences between the first
23 and second metatarsal bases and swelling around the bony prominences. Tenderness
24 and hypesthesia were present in the deep peroneal nerve domains on both feet, with a
25 Tinel-like sign on the right foot. The Japanese Society for Surgery of the Foot (JSSF)
26 midfoot scale [4] scores were 67 points bilaterally.

27 Weight-bearing plain radiographs revealed ossicles between the bases of the
28 first and second metatarsals in both feet (Fig. 1A, B). There was a difference in the
29 distance between the base of the second metatarsal and the medial cuneiform
30 bilaterally, but the patient had no symptoms and no history of injury. Three-dimensional

31 computed tomography (3D-CT) showed that the ossicle on the right side was 7.3 mm ×
32 9.9 mm × 12.0 mm in size, and that on the left side was 6.1 mm × 7.7 mm × 10.7 mm
33 (Fig. 2). No osseous fusion or articular surface formation with the surrounding bone
34 was observed on either side. Magnetic resonance imaging (MRI) showed a bone
35 fragment deep in the dorsal Lisfranc ligament (Fig. 3A,B). Based on these findings, we
36 diagnosed this case as painful bilateral os intermetatarsea.

37 For 2 months we administered conservative treatments such as medication with
38 non-steroidal anti-inflammatory drugs and rest with local decompression by changing
39 the patient's shoes, but the pain persisted. We thus performed surgical excision of the
40 ossicles on both sides in a single surgical procedure.

41 Under lumbar anesthesia, an approximately 20-mm vertical skin incision was
42 made just above the ridge of the right foot. To preserve the DLL, we approached from
43 the distal side and expanded the deep layer. The os intermetatarseum was identified
44 between the bases of the first and second metatarsals, and it compressed the deep
45 peroneal nerve dorsally. There was DLL on the dorsal side of the proximal portion of
46 the ossicle. However, the DLL was not exposed to prevent damage. The nerve was
47 carefully avoided, and the os intermetatarseum was exposed from distal to proximal to
48 protect the DLL (Figs. 4 A,B). The os intermetatarseum was loosely connected to the

49 surrounding tissue (including the DLL), so it was easy to strip between the ossicle and
50 them and perform an en-bloc resection. Therefore, the DLL itself largely untreated and
51 we determined that it is not damaged.

52 The same procedure was then performed on the left foot, and similar findings
53 were obtained. The histopathological findings showed that the bone tissue was mature,
54 and no evidence of malignancy or osteonecrosis was observed.

55 From the first day post-surgery, pain-limited weight bearing was permitted
56 without any other restrictions. No bandages or orthotics were used. One week after the
57 surgery, the patient was able to walk independently. Three weeks after the surgery, he
58 was able to return to daily life, and weight-bearing plain radiographs of the bilateral feet
59 showed no changes in the distance between the base of the second metatarsal and the
60 medial cuneiform bone compared to the distance pre-operation (Fig. 5A,B). Although it
61 did not interfere with daily life, the patient had to restrict his work for 4 months after the
62 surgery because of scarring, irritation, and pain caused by his military-issue boots.

63 One year after the surgery, the patient has no pain and no sensory disorder of
64 the deep peroneal nerve, and the JSSF midfoot scores were 100 points bilaterally.
65 Weight-bearing plain radiographs of the bilateral feet showed no re-ossifications and no
66 changes in the distance between the base of the second metatarsal and the medial

67 cuneiform bone compared to the distance pre-operation (Fig. 6A,B). Institutional review
68 board approval was not required for this case report of a single patient. Informed
69 consent for his case and the images to be published was obtained from the patient
70 preoperatively.
71

72 **DISCUSSION**

73

74 The os intermetatarsium is an uncommon accessory ossicle located on the
75 dorsal aspect of the mid-foot between the first and second metatarsal bases and
76 proximally by the medial cuneiform [5]. It was first described in 1856 by Gruber [6]. Os
77 intermetatarsia exhibit various shapes and sizes [5]. The reported incidence of os
78 intermetatarsium ranges from 0% to 14% [7], and diagnosis tends to be difficult using
79 plain radiographs [5]. Os intermetatarsia can be divided into three basic types:
80 freestanding, articulating, and fused [7]. The most frequently reported type of os
81 intermetatarsium is the freestanding type at 63%, followed by the articulating type at
82 30%; only 7% have been the fused type [8]. This case was of the freestanding type.
83 Most os intermetatarsia are asymptomatic [1], and most are unilateral; good treatment
84 results have been achieved with surgical excision [2,3]. Bilateral cases of os
85 intermetatarsia are very rare. Kose et al. have reported a case of conservative
86 treatment [1], and Noguchi et al. have reported a case with bilateral symptoms in which
87 surgical treatment was performed only on one side [9]. To the best of our knowledge,
88 the present report is the first to describe the bilateral simultaneous surgical treatment of
89 bilateral os intermetatarsia.

90 The Lisfranc ligament is a complex of three different ligaments: the DLL, the
91 interosseous Lisfranc ligament (ILL), and the plantar Lisfranc ligament (PLL).
92 Theoretically, excision from directly above an os intermetatarsium using the dorsal
93 approach may damage the DLL. The reported areas of the three ligaments are as
94 follows: DLL, $15.3\pm 4.0\text{ mm}^2$; PLL, $33.1\pm 12.2\text{ mm}^2$; and ILL, $74.8\pm 17.5\text{ mm}^2$ [10]. The
95 ligaments' strength values are as follows: DLL, $170\pm 33\text{ N}$; PLL, $305\pm 38\text{ N}$; and ILL,
96 $449\pm 58\text{ N}$ [11]. The DLL therefore has the highest ultimate load per unit area. The DLL
97 is located on the dorsal side of the Lisfranc joint, which is the apex of the transverse
98 arch structure of the foot. The in vivo strain pattern that occurs when the Lisfranc
99 ligament is injured is not accurately known [12], but it is expected that the ILL and PLL
100 will be overloaded when only the DLL is injured. Therefore, we consider that gentle
101 post-operative management may be required when only the DLL is injured. In fact,
102 relatively long-term load limits or activity limits were reported in previous studies [2,3].
103 Noguchi et al. performed surgical excision to treat the free-standing type of os
104 intermetatarsium, and, after a non-weight-bearing period, their patient started partial
105 weight-bearing 2 weeks post-surgery and started full weight-bearing 5 weeks post-
106 surgery [2]. Nakasa et al. reported four patients who returned to athletic competition 4–
107 6 months after surgical treatment [3].

108 In the present case, we preoperatively evaluated the positional relationship
109 between the DLL and the os intermetatarsium, and we shifted the approach distally to
110 minimize invasion of the ligament. We believe that the DLL was preserved without any
111 iatrogenic tear because it was not exposed and the bone fragments were resected
112 under the periosteum. Therefore, we permitted early weight bearing after the surgery.
113 Comparing the radiographs of the feet before and after surgery, these showed no
114 changes in the distance between the base of the second metatarsal and the medial
115 cuneiform bone. These findings suggest that there was no DLL tear associated with
116 this surgery.

117 Our patients were able to carry out their daily activities soon after surgery. In
118 young patients such as our case, a similar prognosis could likely be obtained by having
119 the patient walk on the heel while reducing or eliminating the load on the Lisfranc joint.
120 However, it is not known whether all patients with painful os intermetatarsium are
121 young and/or healthy. We believe that our report will serve as a reference when dealing
122 with a variety of cases. On the other hand, it took 4 months postoperatively for the
123 patient to fully return to work in our case. This delay was due to the patient's special
124 work environment (i.e., the need to wear cramped military shoes), and except for this
125 special circumstance, it should have been possible for the patient to return to work

126 earlier after this surgery. We believe that the value of conducting unlimited
127 rehabilitation soon after surgery is universal.

128 In conclusion, bone resection for os intermetatarsea that preserves the DLL can
129 be expected to enable early load-bearing walking.

130

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162

163 **FIGURE LEGENDS**

164

165 Fig. 1. Dorsoplantar view of the weight-bearing plain radiographs. A: Left foot. B: Right
166 foot. White arrow indicates the os intermetatarsium bilaterally. Red bar indicates the
167 distance between the base of the second metatarsal and the medial cuneiform bone,
168 and left side has 2.5mm, right side has 3.5 mm.

169

170 Fig. 2. 3D-CT of both feet demonstrating freestanding os intermetatarsia between the
171 base of the first and second metatarsals (*light blue parts*).

172

173 Fig. 3. MRI findings of os intermetatarsium and surrounding tissues (axial view in
174 parallel to the dorsal Lisfranc ligament of a T2-weighted image). A: Left foot. B: Right
175 foot. *White arrowheads*: The dorsal Lisfranc ligaments. MC: medial cuneiform, M2: 2nd
176 metatarsal, IM: os intermetatarsium.

177

178 Fig. 4. Intraoperative findings of the right foot. A: Before resection. The os
179 intermetatarsium was compressing the deep peroneal nerve upwards from the bottom.
180 B: After resection. Avoiding the deep peroneal nerve, the os intermetatarsium was
181 resected as a mass.

182

183 Fig. 5. Three weeks post-surgery, anteroposterior plane radiographs of both feet. A:
184 Left foot. B: Right foot.

185

186 Fig. 6. One year post-surgery, anteroposterior plane radiographs of both feet. A: Left
187 foot. B: Right foot. Red bar indicates the distance between the base of the second
188 metatarsal and the medial cuneiform bone, and these are same distances at pre-
189 operation.

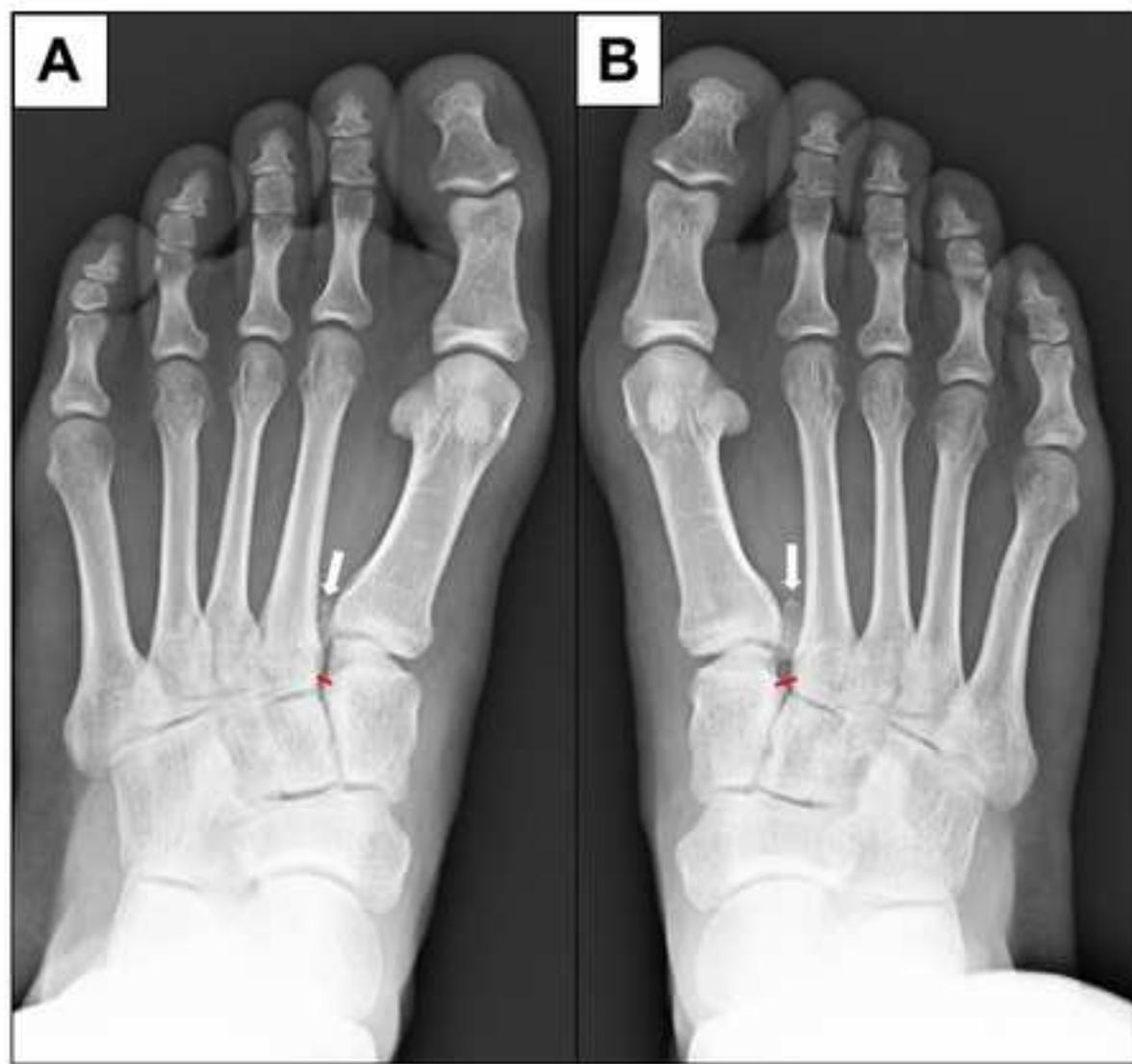


Figure 1

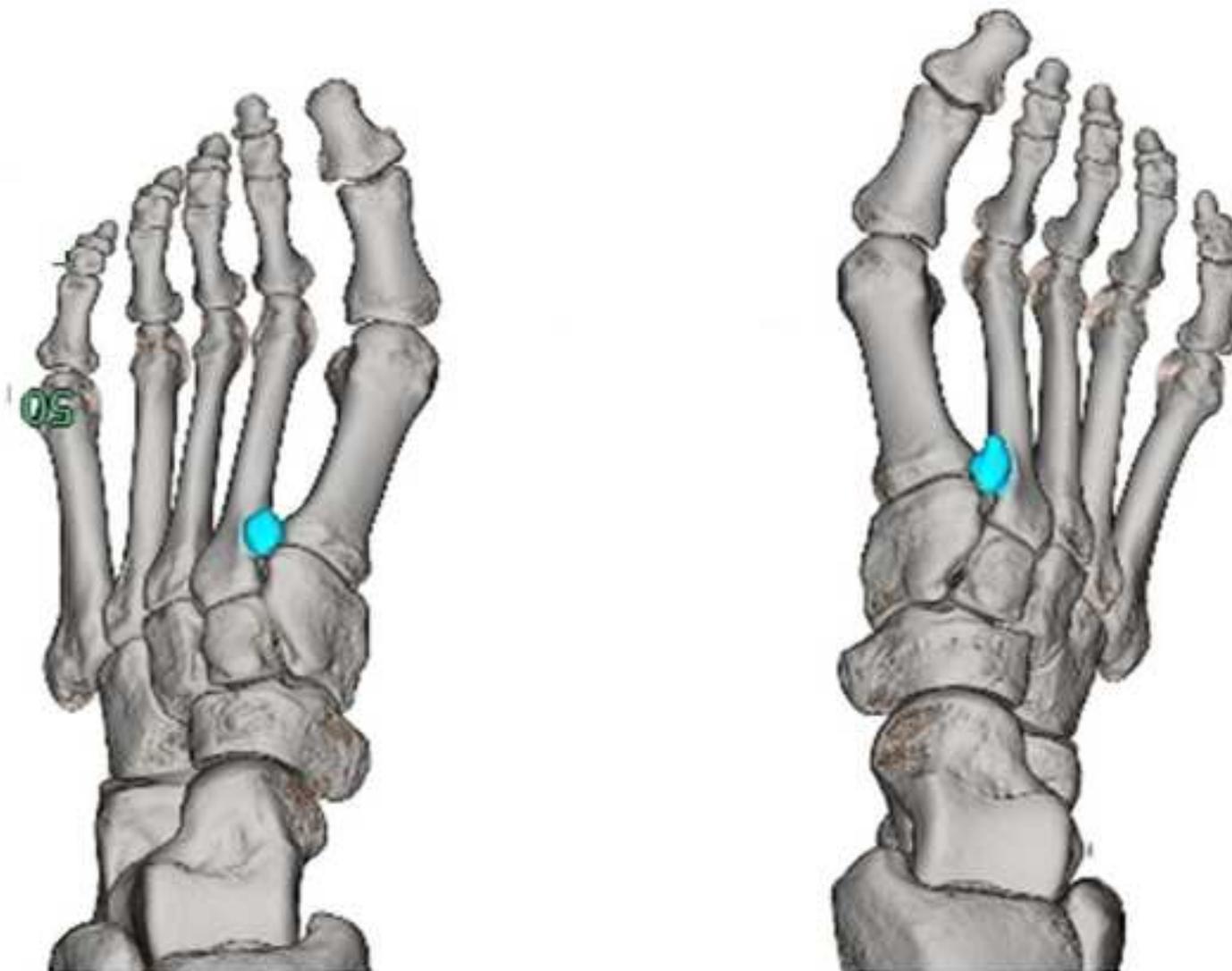
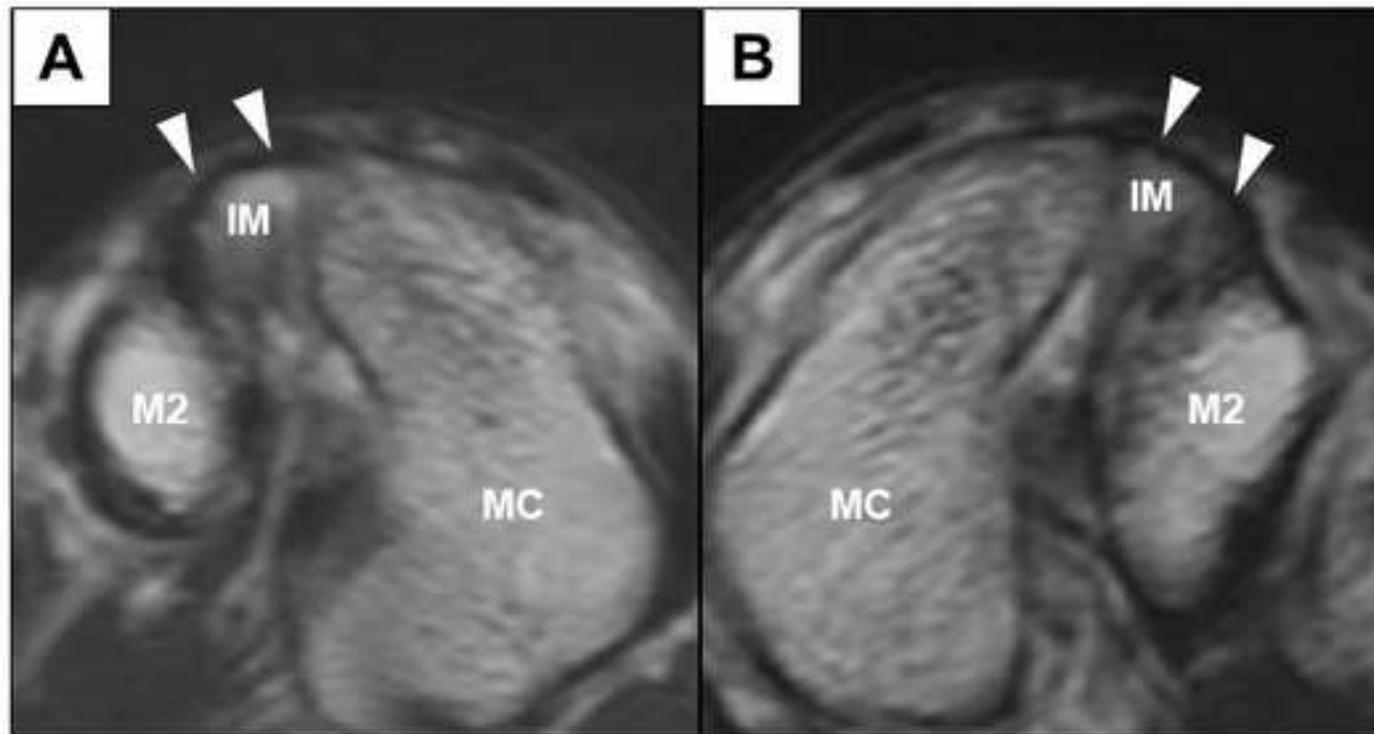


Figure 2



MC: medial cuneiform, M2: 2nd metatarsal, IM: os intermetatarsaleum

Figure 3

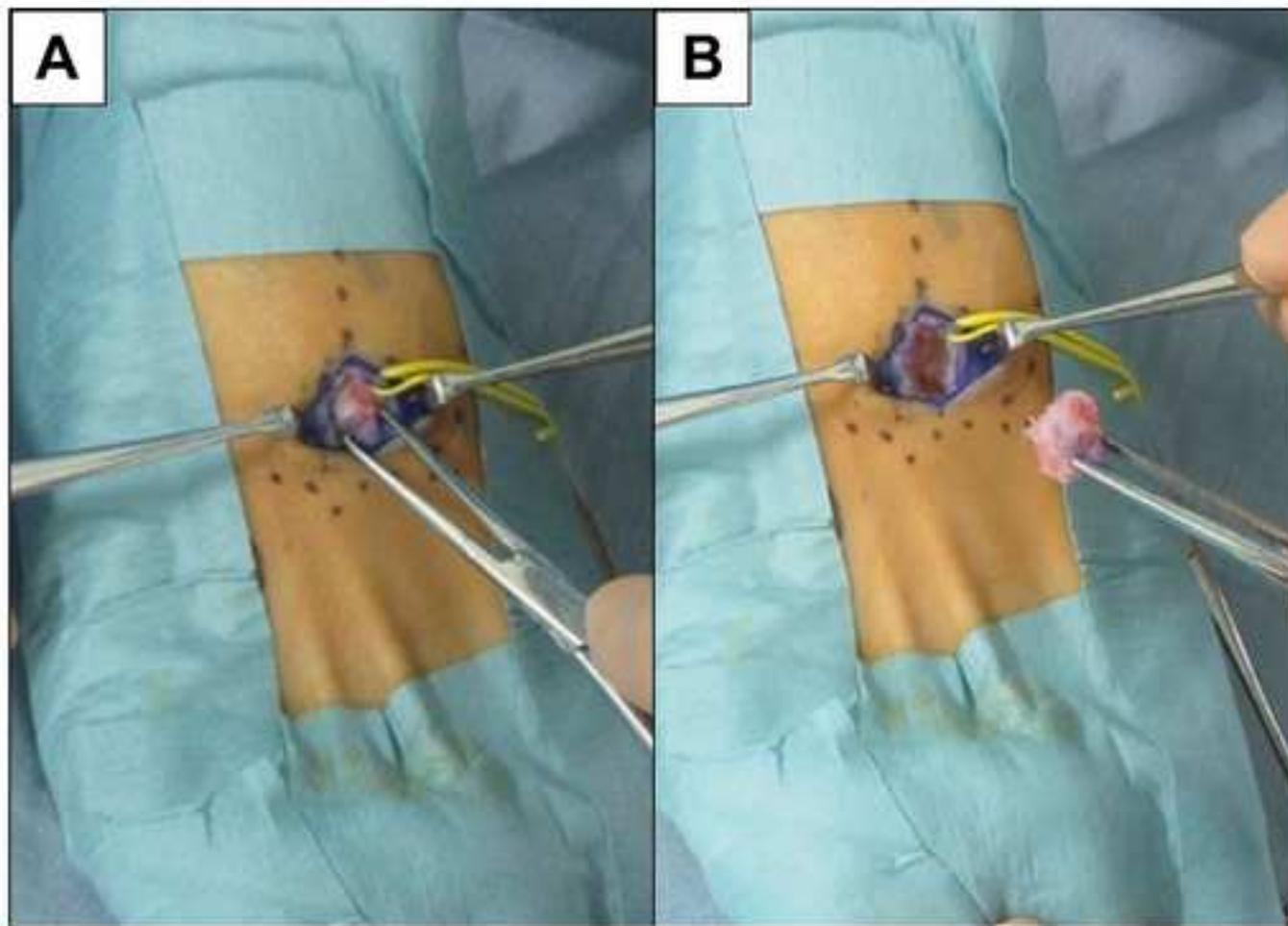


Figure 4

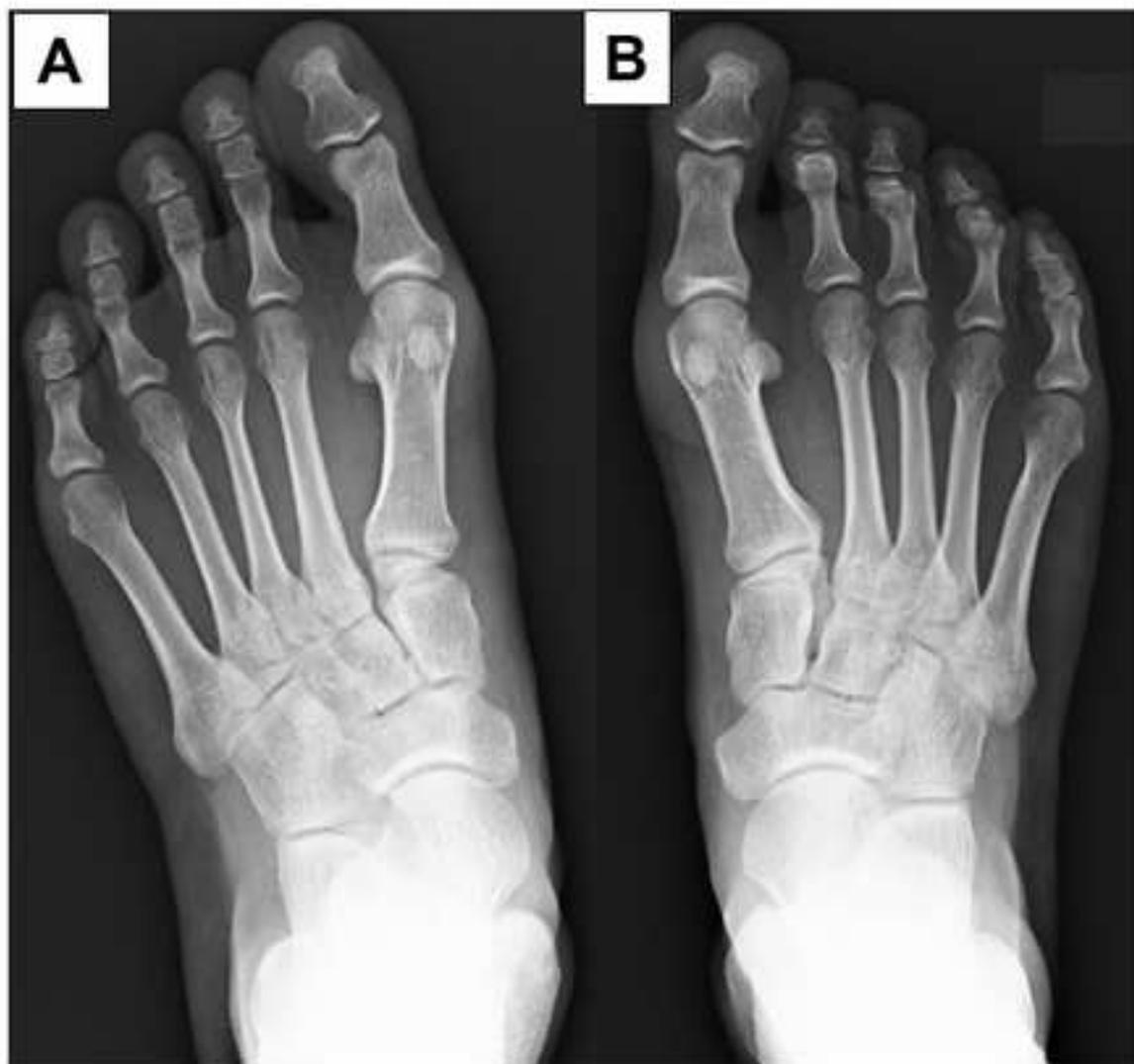


Figure 5

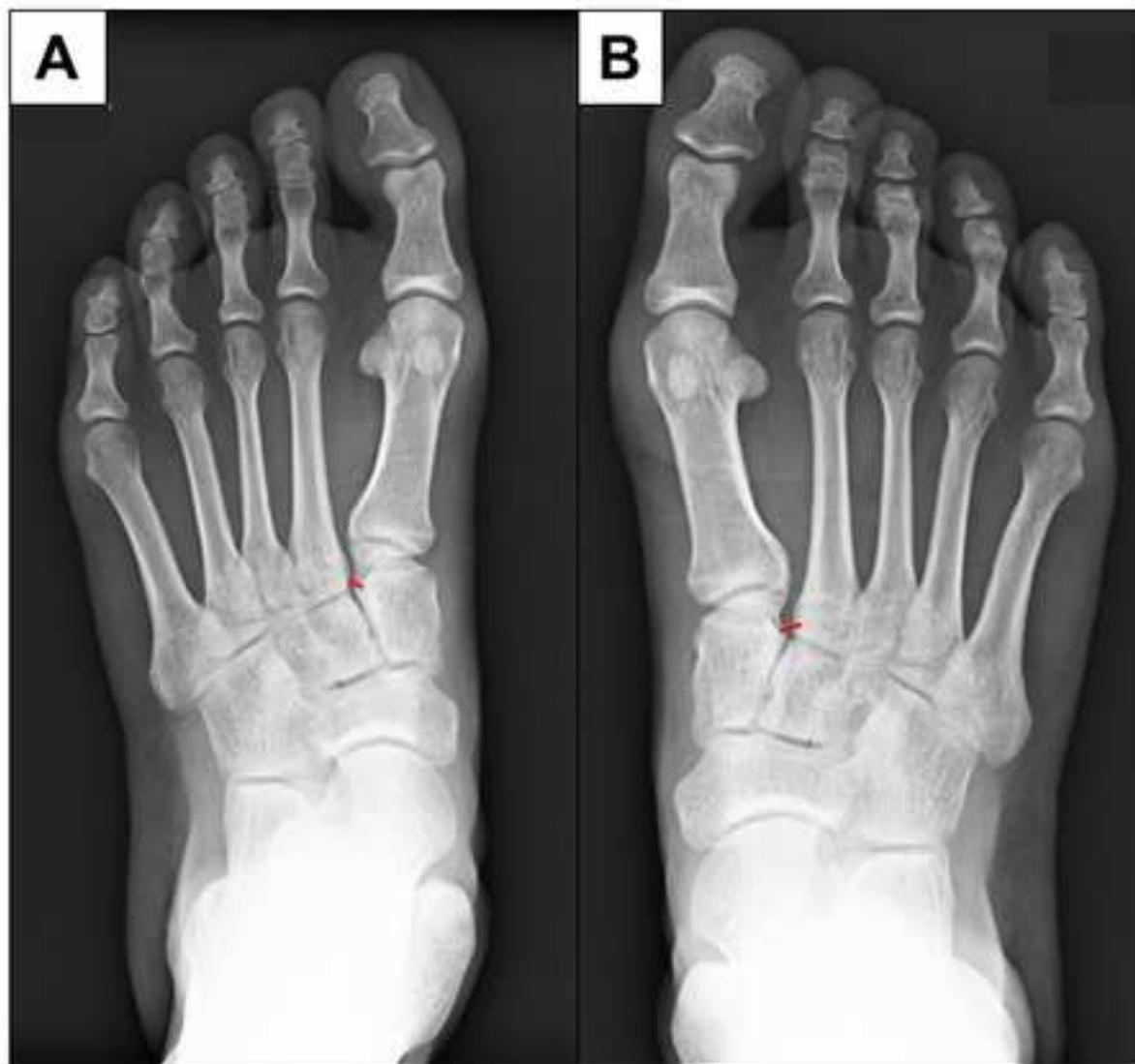


Figure 6