The Journal of Foot & Ankle Surgery 58 (2019) 1019–1024

Contents lists available at ScienceDirect

The Journal of Foot & Ankle Surgery

journal homepage: www.jfas.org

Adult Flat Foot With Multiple Accessory Navicular Bones Treated Surgically: A Case Report and Review of the Literature

Toshinori Kurashige, MD, Yoshihiko Tsunoda, MD

Surgeon, Department of Orthopaedic Surgery, Chiba Aiyukai Memorial Hospital, Chiba, Japan

ARTICLE INFO

Clinical Level of Evidence: 4

Keywords:
multi-ossicle configuration
os tibiale externum
surgery
variation

ABSTRACT

Multiple accessory navicular bones is an extremely rare condition. To the best of our knowledge, only 8 cases in 2 imaging studies have been published. We report a case of a patient with flat foot with 2 accessory navicular bones. This patient needed to be treated surgically, and the surgery was successful, with short-term follow-up. We believe this is the first case of multiple accessory navicular bones to be treated surgically in English literature. The incidence of multiple accessory navicular bones might be higher. There is a risk to remaining ossicles without resection or fixation during surgery; therefore, we strongly recommend using not only radiographs, but also 3-dimensional computed tomography scans or magnetic resonance imaging scans to confirm the type of accessory navicular bone, at least before surgery, for both painful accessory navicular bone and flat foot with accessory navicular bone.

The accessory navicular (AN) bone is one of the most common accessory bones of the foot. It is considered a normal anatomic and skeletal variant, accounting for an incidence rate of 2%–14% (1,2). This ossicle is also referred to as os tibiale externum, os naviculare secundarium, os naviculare accessorium, os scaphoidea accessoria, and prehallux. The AN is located in the posteromedial aspect of the foot, proximal to the navicular bone, and continuous with the tibialis posterior tendon (PTT).

Three types of AN have been described (3). Type 1 is the sesamoid bone lying within the PTT insertion. Type 2 results from a secondary ossification center adjacent to the navicular bone; it is the insertion site of the PTT and is connected to the navicular tuberosity by a synchondrosis. Type 3 results from the fusion of the secondary ossification center with the navicular bone and is also called navicular. Sella and Lawson (3) subdivided type 2 into A and B. Type 2A represents a more superior placement of AN, which is more prone to disruption owing to chronic tension forces or acute avulsion. Type 2B represents a more inferior location, which is more prone to chronic shearing forces causing pain. Although AN is usually asymptomatic, it can be the source of painful midfoot syndrome owing to altered biomechanics.

Multiple ANs is an extremely rare condition. To the best of our knowledge, only 8 cases in 2 imaging studies have been published (4,5). We report here a case of a patient with flat foot with 2 ANs. This patient needed to undergo AN resection, lateral column lengthening, and flexor digitorum longus (FDL) tendon transfer. The result of the surgery was successful, with short-term follow-up. We believe that this is the first patient with multiple AN treated surgically in English literature.

Case Report

This is a case of a 41-year-old female patient who was diagnosed with flat foot in her childhood. She had been treated using orthopedic shoes and insoles until 8 years old. She began to experience bilateral foot pain while standing and walking during her teenage years. She was diagnosed with bilateral painful ANs at another clinic 5 years prior. She was treated conservatively; however, pain worsened, and she began to experience pain even while resting and at night. She also had a history of subluxation of her left shoulder. Subsequently, she was referred to our department.

Physical examination showed that she had flexible flat foot bilaterally, with the presence of callosity beneath the right navicular bone (Fig. 1). Both navicular bone and PTT insertions were tender. Pain increased when bearing weight. She could not raise her heels. General joint laxity test (6) was negative; however, bilateral flat feet were flexible enough to be reduced easily by manipulation.

On radiographs and computed tomography (CT) scans, bilateral ANs were seen (Figs. 2 and 3), with 2 ossicle configurations present in the right foot, and type 2 configuration was observed in the left foot. Both navicular bones were enlarged and curved dorsomedially, and bilateral ANs were located at the posterodorsal site of the navicular bones. Partial calcification was observed in the right PTT. On the weightbearing

Financial Disclosure: None reported.
Conflict of Interest: None reported.

Address correspondence to: Toshinori Kurashige, MD, Department of Orthopaedic Surgery, Chiba Aiyukai Memorial Hospital, 1-1, Hiregasaki, Nagareyama city, Chiba, 270-0161, Japan.

E-mail address: to-kurashige@chibaaiyu-kai.com (T. Kurashige).

1067-2516/$ - see front matter © 2018 by the American College of Foot and Ankle Surgeons. All rights reserved.
https://doi.org/10.1053/j.jfas.2018.12.032

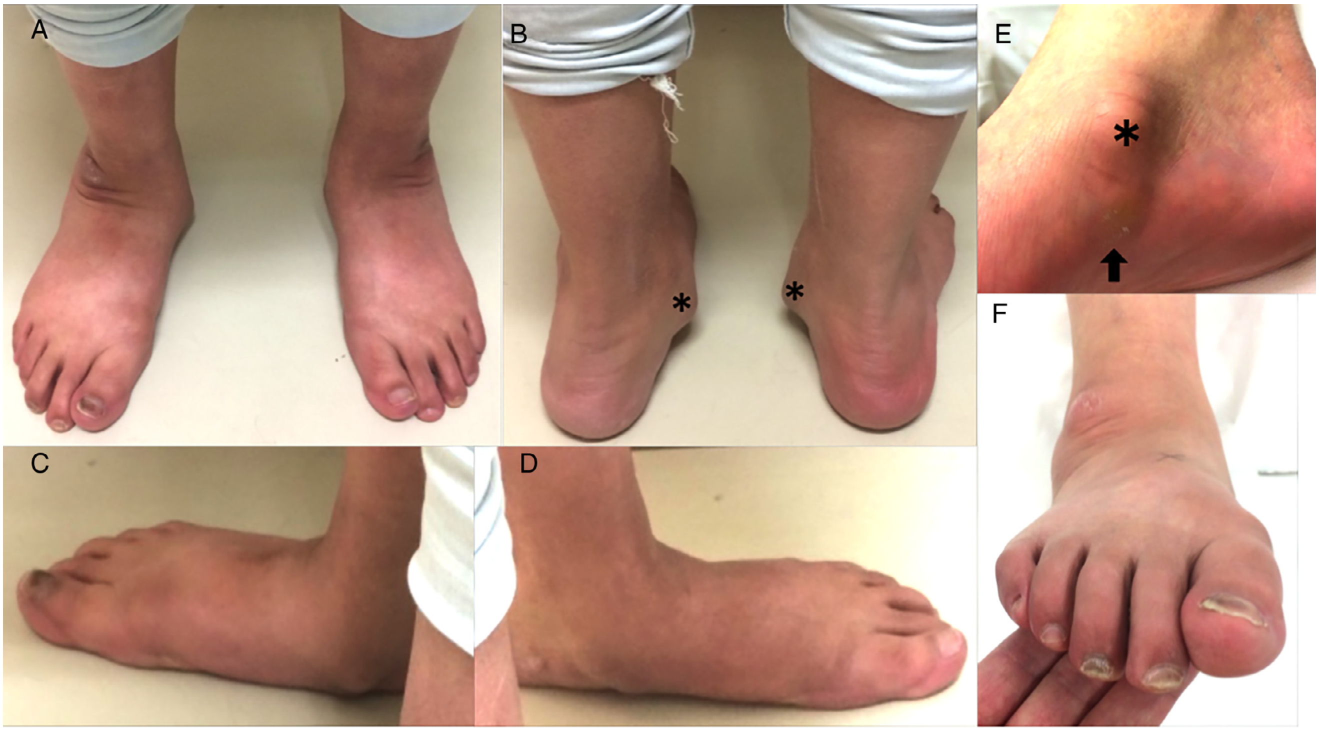

Fig. 1. Preoperative macroscopic findings. (*A*) Image of the anterior aspect of the feet shows pes planovalgus with the patient in a standing position. (*B*) Image of the posterior aspect of the lower legs shows valgus deformity of the bilateral heels with excessive bony protrusions of accessory naviculars (*black asterisks*) with the patient in a standing position. (*C* and *D*) Images of the lateral aspect of the feet show bilateral flat feet with the patient in a standing position. (*E*) Callosity (*black arrow*) is observed beneath the right accessory navicular (*black asterisk*). (*F*) Flat feet were flexible and reduced easily.

lateral radiographs, the lateral talo-first metatarsal angle was 29° and 26° in the right and left feet, respectively. The calcaneal pitch angle was 12° and 11° in the right and left feet, respectively. The talonavicular coverage angles were not measured because of the enlargement and deformities of the navicular bone. On the hindfoot alignment view, the valgus angles were 7° and 6° in the right and left feet, respectively. Magnetic resonance imaging (MRI) using short TI-inversion recovery sequences demonstrated the high signal lesions at the PTT insertions (Fig. 4). No high signal lesion was found at the posterior site of the medial malleolus bilaterally.

Injection of 1 mL of lidocaine in the synchondrosis using a 26-G needle resulted in a temporary but almost complete resolution of pain. We diagnosed the patient with adult flexible flat foot and painful ANs.

Because conservative treatment was unsuccessful, surgical treatment for the right foot was performed under general anesthesia. First, lateral column lengthening and calcaneocuboid joint fusion were performed from a lateral incision. We prefer a fusion of the joint to an Evan's lateral column lengthening to avoid the joint pain owing to the increasing of the joint pressure. Iliac crest autograft was fixed with locking plate and screws. Second, an oblique skin incision was made along the PTT in the medial aspect of the foot. A severe adhesion was found around the PTT insertion, and the insertion was widened to the dorsal aspect of the dorsal AN. Extensive degenerative change was observed at the PTT insertion; therefore, repair along with FDL tendon transfer for augmentation was needed. The PTT fibers inserted in the dorsal AN were fewer than those in the plantar AN. The 2 ANs were resected. Because they were small, screw fixations to the primary navicular bone had the risk of fracturing not only the ANs, but also the primary navicular bone after drilling for FDL tendon transfer. The dorsal spur and medial enlarged portion of the navicular were also resected. The FDL was passed from the plantar to the dorsal through the drill hole in the navicular bone. After the talonavicular capsule/spring ligament sutures were secured in a pants-over-vest fashion, the FDL was sutured to the surrounding soft tissue with the foot in inversion. The longitudinal tears of the PTT insertion were repaired and sutured to the navicular bone using a soft suture anchor. The ankle could not return to a neutral position after these procedures; therefore, gastrocnemius recession was performed through a small incision in the medial calf. Pathological examination revealed no osteonecrosis in the 2 resected ANs.

The foot was immobilized postoperatively in a short-leg splint in the varus position. The splint was removed and weightbearing with an arch support was permitted as tolerated 6 weeks postoperatively. The arch support was used for 6 months (Fig. 5). Although she experienced transient pain in the lateral side of the foot postoperatively, no major complications, such as superficial or deep infection, nonunion, painful hardware, or nerve damage occurred. At follow-up of 12 months postoperatively, the medial aspect of the right foot was nontender, and slight pain was experienced after walking for long durations. She could raise her right heel. The callosity beneath the left navicular bone disappeared. The patient was satisfied with the result of the surgery, and she underwent another surgery for the left foot 9 months after the first surgery. The left PTT showed a smaller tear and less degeneration than the right; therefore, lateral column lengthening, resection of AN and spur of the navicular, repair of the PTT insertion, and gastrocnemius recession were performed for the left foot. On weightbearing radiographs of the right foot without arch support 12 months after the first surgery, the lateral talo-first metatarsal and calcaneal pitch angles in the right foot were 14° and 16°, respectively. On the hindfoot alignment view, the valgus angle in the right foot was 2°. The American Orthopaedic Foot and Ankle Society ankle–hindfoot scale score of the right foot improved from 49 preoperatively to 87 postoperatively (7,8).

This work has been approved by the appropriate ethical committees related to the institution where it was performed, and informed consent was obtained from the subject.

Fig. 2. Preoperative radiographs. Anteroposterior radiographs reveal bilateral accessory naviculars (*A* and *B*), type 2 in the left foot (*C*), and 2 ossicles in the right (*D*) (*black asterisks*). A calcification in the right tibialis posterior tendon (*black arrow*) is seen. Both navicular bones are enlarged and curved dorsomedially. (*E* and *F*) Weightbearing lateral radiographs show bilateral flat feet and accessory naviculars located at the posterodorsal site of the navicular bones. (*G* and *H*) Weightbearing posterior-anterior radiographs show bilateral calcaneus in the valgus position. Calcifications in the bilateral tibialis posterior tendons (*black arrows*) are seen.

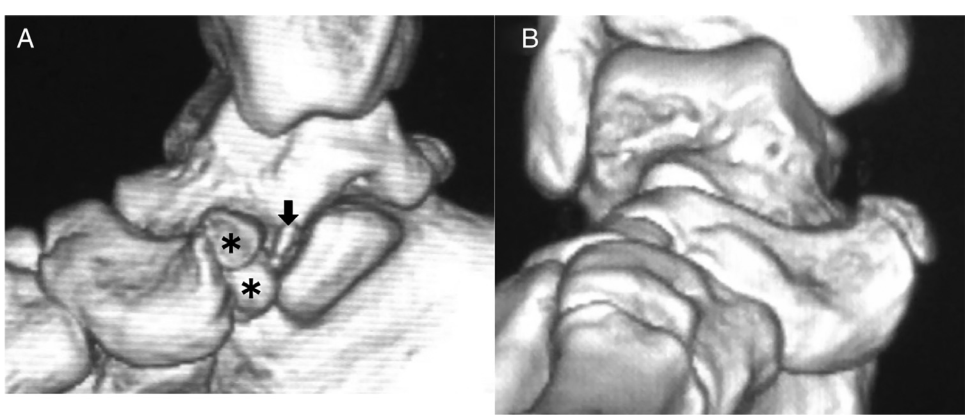

Fig. 3. Preoperative computed tomography scan findings. (*A*) Three-dimensional computed tomography scans reveal the presence of 2 ossicles in the right foot (*black asterisks*). Calcification in the tibialis posterior tendon is identified (*black arrow*). (*B*) The navicular bone is enlarged and curved dorsomedially, and the accessory naviculars are located at the posterodorsal site.

Discussion

The navicular bone is the last bone to ossify at age 2–3 years and 4–5 years in females and males, respectively (5). The navicular bone ossifies endochondrally from a single center. The AN forms when the tuberosity of the navicular bone develops from a secondary center of ossification, and it begins to ossify at ~9 and 12 years of age in females and males, respectively. The bones of the feet mature at an average of

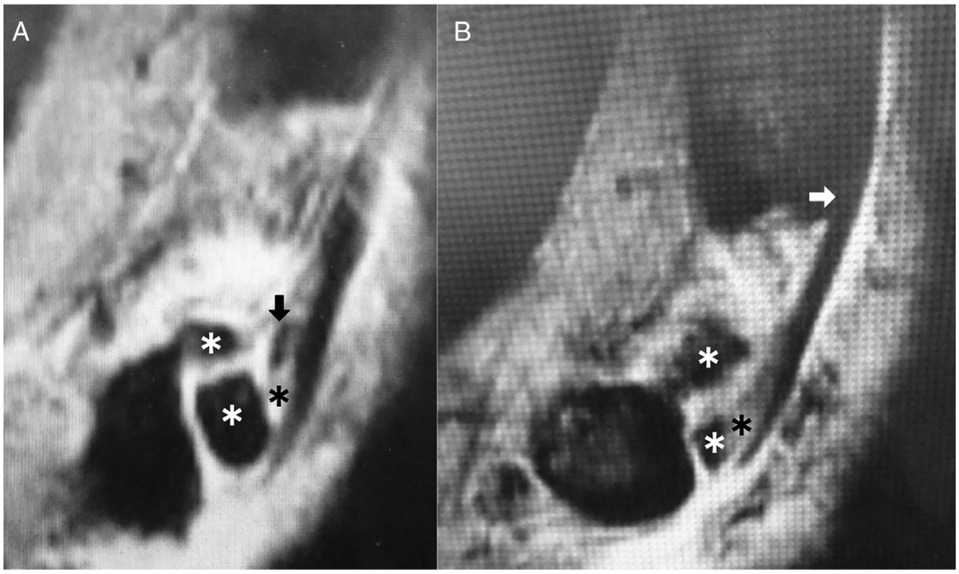

Fig. 4. Preoperative magnetic resonance imaging scan findings. (*A*) Magnetic resonance imaging scans using short TI-inversion recovery sequences demonstrate 2 ossicles proximal to the navicular bone (*white asterisks*) and the high signal lesion at the tibialis posterior tendon insertion (*black asterisk*). Calcification in the tibialis posterior tendon is identified (*black arrow*). (*B*) No high signal lesion of the tibialis posterior tendon at the posterior site of the medial malleolus is observed (*white arrow*).

13 and 15 years in females and males, respectively. The AN fuses shortly after ossicle fusion to the main navicular bone occurs. The factors determining whether AN unites with the main navicular bone or persists into adulthood are still unexplored.

The study by Perdikakis et al (4) in 2011 was the first report regarding the prevalence of the multi-ossicle configuration of AN (Table). They performed CT or MRI of the foot and ankle applying standard protocols, including all 3 imaging planes. The AN bone was identified in 34 cases (20%) of the 170 examinations performed, which accounted for 11.15% (19 cases), 4.11% (7 cases), and 4.74% (8 cases) for types 1, 2, and 3 classifications, respectively. Of 5 patients with AN, multi-ossicle appearance was observed, which accounted for 14.7% of the configurations (8.8%, 2 ossicle configurations; 5.9%, 3 ossicle configurations). The overall incidence rate of multiple ossicles among all patients was 2.9%. Kalbouneh et al (5) retrospectively reviewed anteroposterior, lateral, and or oblique radiographs of the feet (Table). The AN was found in 259 feet (20.9%) of 1240 patients. Of 259 feet with AN, types 1, 2, and 3 configurations were identified in 25.4% (66 feet), 42.4% (110 feet), and 32.0% (83 feet), respectively. Of 3 patients with AN, multi-ossicle appearance was observed, which accounted for 1.2% of the configurations (2 ossicle configurations in all cases). The overall incidence rate of multiple

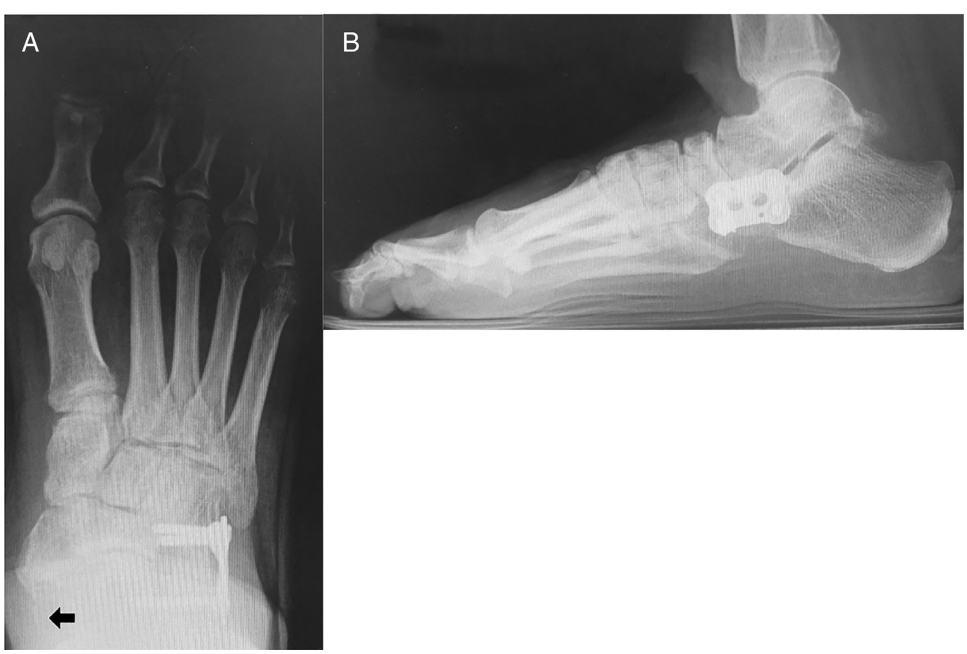

Fig. 5. Postoperative radiographs. (*A* and *B*) On postoperative weightbearing radiographs, flat foot was corrected. Calcification in the tibialis posterior tendon partially remained (*black arrow*).

Table
Review of the literature

Feature	Perdikakis et al (4)	Kalbouneh et al (5)
Nation	Greece	Jordan
Modality	CT or MRI	Radiographs
Numbers of feet	170	1240
Incidence of overall AN, %	20%	20.9%
Incidences of each type, %	Type 1: 55.9, Type 2: 20.6, Type 3: 23.5%	Type 1: 25.4, Type 2: 42.4, Type 3: 32.0
Incidence of multiple ANs, %	2.9 among all patients; 14.7 among patients with AN	0.24 among all patients; 1.2 among patients with AN
Configuration	2~3 ossicles	2 ossicles

Abbreviations: AN, accessory navicular; CT, computed tomography; MRI, magnetic resonance imaging.
The incidence of overall accessory navicular are similar in both studies; however, the incidence of each type, multiple accessory naviculars, and its configurations were different between the 2 studies.

ossicles among all patients was 0.24% in their series. In the present case, not only type 2A, but also type 2B ANs were present in the same foot. In addition, the deformity of the navicular bone existed.

Perdikakis et al (4) suggested that the multi-ossicle configuration of AN can be explained as a complication of repetitive microtrauma and fatigue fracture of the secondary ossification center. They also suggested that it could result from an avulsion injury to AN, in which the fractured bone fragment eventually became a secondary ossicle, resulting in a multi-ossicle configuration. Kalbouneh et al (5) suggested that AN can have 1, 2, or more nuclei of ossification that fail to fuse and remain separate from the navicular mass. Some nuclei of ossification could also represent a traumatic division of AN center of ossification during development. In the present case, general joint laxity test was negative; however, the patient had a history of subluxation of the left shoulder, and bilateral flat feet were flexible enough to be easily reduced by manipulation despite the long history. The cause of navicular bone deformity could also be explained by the traction force of the PTT.

From a biomechanical point of view, the AN generally alters the functional and anatomic balance of the longitudinal plantar arch (9). Several of the PTT fibers insert on the accessory ossicle instead of its insertion on the tarsal and metatarsal bones, resulting in unbalanced stress forces on the tendon. The presence of a type 2 or 3 AN is a risk factor for PTT tendinopathy and possible PTT tear (4).

The goal of surgery for flat foot with AN is pain relief and restoration of the dynamic support of the longitudinal plantar arch. For painful AN in adults, current surgical techniques recommend either AN removal and PTT reinsertion or primary navicular and AN fusion (10,11). Screw osteosynthesis of the AN, which is large enough to accept small screws, achieved good results; however, for screw

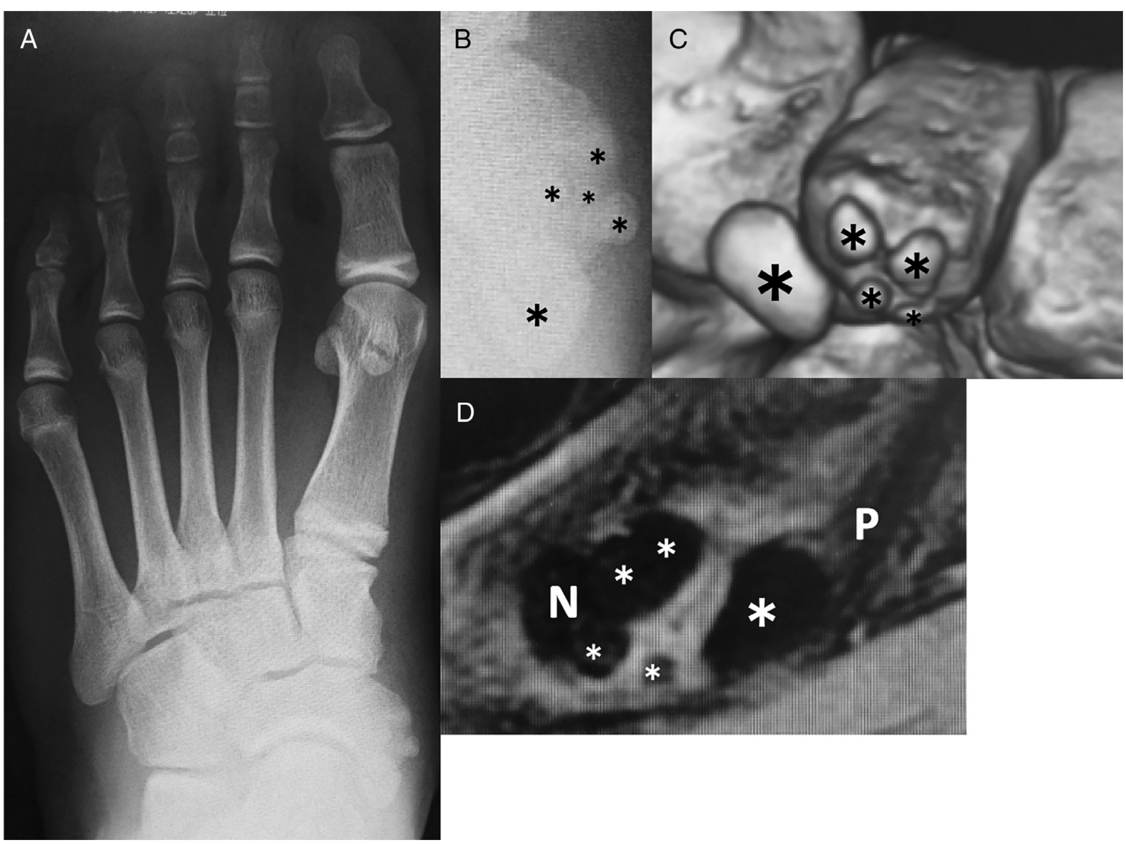

Fig. 6. Image findings of case 2. A 14-year-old male with tibial sesamoid disorder. The medial side of his foot was painless; however, the radiographs (*A* and *B*) and computed tomography scans (*C*) reveal 5 ossicles (*black asterisks*). (*D*) Magnetic resonance imaging scan also shows 5 ossicles (*white asterisks*) near the navicular bone (N, navicular; P, tibialis posterior tendon).

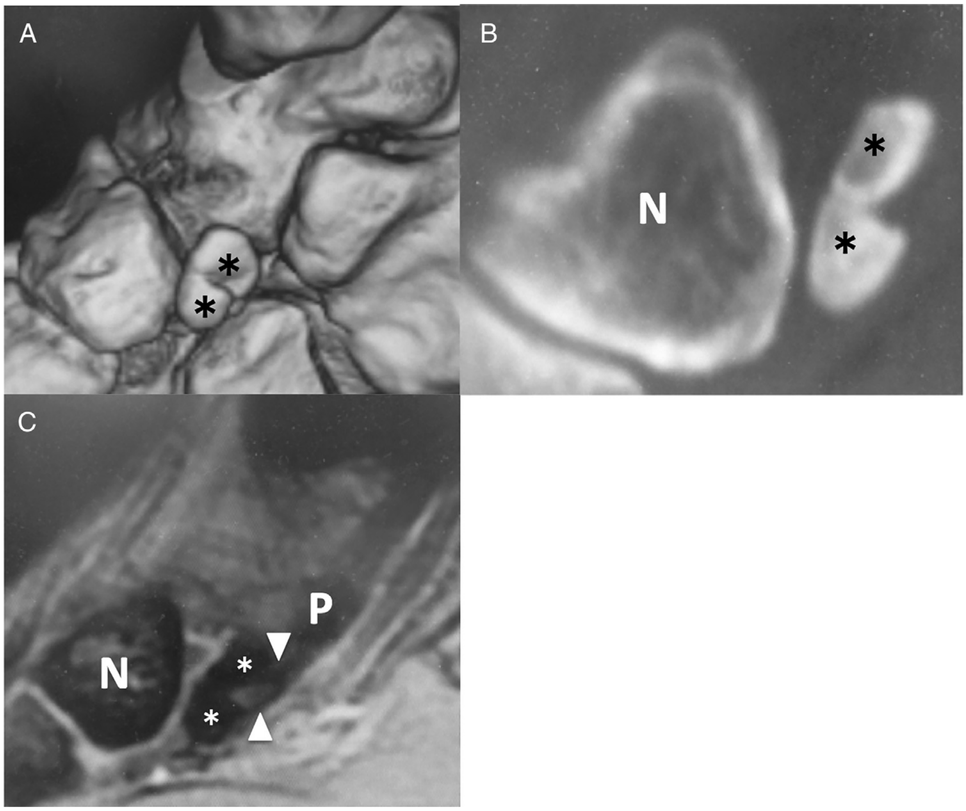

Fig. 7. Image findings of case 3. A 46-year-old male experiencing pain at the lateral side of his right foot after running. He was diagnosed with arthritis of the calcaneocuboid joint. The medial side of his foot was painless. Plain radiographs show type 2 accessory navicular. (*A* and *B*) However, computed tomography scans reveal 2 ossicles (*black asterisks*). (*C*) Magnetic resonance imaging scans also shows 2 ossicles (*white asterisks*) and 2 tibialis posterior tendon insertions (*white arrowheads*) (N, navicular; P, tibialis posterior tendon).

fixation, the diameter of the AN should be at least 2 to 3 times wider than the screw or drill bit necessary for the procedure, and these procedures have a risk for AN fracture. In the present case, the 2 ANs were resected because of the reasons described previously. Some authors have suggested that the combination of tendon transfer and osteotomy is most efficient in correcting flexible flat foot in children and adults. The procedures for flat foot reconstruction should consider the situation of each flat foot (12,13).

The incidences of AN are similar in both studies by Perdikakis et al (4) and Kalbouneh et al (5) (Table); however, differences were found between the 2 studies in terms of the incidences of each type of AN and the incidence of multiple ANs and its configurations. The differences may cause from nationality and or modality. Plain radiographs may be insufficient in diagnosing the pathology. These additional ossicles can be mistaken for calcific tendonitis of the PTT, a condition rarely reported in the literature (14). CT and MRI can verify the presence of small ossicles that otherwise might be overlooked. In fact, we encountered 2 other cases of patients with multiple ANs who were asymptomatic. Five ossicles could be identified by plain radiographs in case 2 (Fig. 6). However, they could not be identified until CT images were obtained in case 3 (Fig. 7). The incidence of multiple ANs might actually be higher. If so, there is a risk to remaining ossicles without resection or fixation during surgery.

In conclusion, we strongly recommend obtaining not only radiographs, but also 3-dimensional CT or MRI scans to confirm the type of AN preoperatively for both painful AN and flat foot with AN.

References

1. Coughlin MJ. Accessory navicular. In: Coughlin MJ, Saltzman CL, Anderson RB, eds. *Mann's Surgery of the Foot and Ankle*, 9th ed, Philadelphia: Elsevier, 2013:544–553.
2. Sarrafian SK, Kelikian AS. Accessory bones. In: Kelikian AS, ed. *Sarrafian's Anatomy of the Foot and Ankle: Descriptive, Topographic, Functional*, 3rd ed, Philadelphia: Lippincott Williams & Wilkins, 2011:100–102.
3. Sella E, Lawson J. Biomechanics of the accessory navicular synchondrosis. Foot Ankle 1987;8:156–163.
4. Perdikakis E, Grigoraki E, Karantanas A. Os naviculare: the multi-ossicle configuration of a normal variant. Skeletal Radiol 2011;40:85–88.
5. Kalbouneh H, Alajoulin O, Alsalem M, Humoud N, Shawaqfeh J, Alkhoujah M, Abu-Hassan H, Mahafza W, Badran D. Incidence and anatomical variations of accessory navicular bone in patients with foot pain. Clin Anat 2017;30:436–444.
6. Sueyoshi T, Emoto G, Yuasa T. Generalized joint laxity and ligament injuries in high school-aged female volleyball players in Japan. Orthop J Sports Med 2017;4:1–5.
7. Kitaoka HB, Alexander IJ, Adelaar RS, Nunley JA, Myerson MS, Sanders M. Clinical rating systems for the ankle-hindfoot, midfoot, hallux, and lesser toes. Foot Ankle Int 1994;15:349–353.
8. Ibrahim T, Beiri A, Azzabi M, Best AJ, Taylor GJ, Menon DK. Reliability and validity of the subjective component of the American Orthopaedic Foot and Ankle Society clinical rating scales. J Foot Ankle Surg 2007;46:65–74.
9. Dyal CM, Feder J, Deland JT, Thompson FM. Pes planus in patients with posterior tibial tendon insufficiency: asymptomatic versus symptomatic foot. Foot Ankle 1997;18:85–88.
10. Kopp FJ, Marcus RE. Clinical outcome of surgical treatment of the symptomatic accessory navicular. Foot Ankle Int 2004;25:27–30.
11. Malicky ES, Levine DS, Sangeorzan BJ. Modification of the Kidner Procedure with fusion of the primary and accessory navicular bones. Foot Ankle Int 1999;20:53–54.
12. Irvin O, Williams BR, Ellis SJ, Kwon DJ, Deland JT. Reconstruction of the symptomatic idiopathic flatfoot in adolescents and young adults. Foot Ankle Int 2011;32:225–232.
13. Niki H, Hirano T, Okada H, Beppu M. Outcome of medial displacement calcaneal osteotomy for correction of adult-acquired flatfoot. Foot Ankle Int 2012;33:940–946.
14. Delmi M, Kurt AM, Meyer JM, Hoffmeyer P. Calcification of the tibialis posterior tendon: a case report and literature review. Foot Ankle Int 1995;16:792–795.