

Stress fracture of tarsal cuboid bone in early childhood

Sun Young Joo · Changhoon Jeong

Received: 24 June 2014 / Accepted: 9 September 2014 / Published online: 24 September 2014
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Abstract

Purpose We present the clinical characteristics, radiographic, and bone scintigraphy finding of the cuboid fracture in early childhood.

Methods From 2008 to 2012, we identified 25 patients (13 boys and 12 girls) with cuboid fracture who were seen in our institution. Medical records and radiographs as well as bone scintigraphy of 25 patients were reviewed. Nutcracker test was performed as a provocation test.

Results The mean age of the patients was 24.7 months (range 15–38 months). The average duration of symptom before visit was 7 days (range 2–14 days). Most of the parents/caregivers (76 %) did not recall a traumatic episode. Patterns of limping were variable. Nutcracker test was positive in 11 patients. In 10 of 25 patients, initial plain radiographs of the foot showed no abnormal finding. The average duration of symptom of these patients was 4.5 days (range 2–7 days). In 15 patients, the radiograph of the foot showed sclerosis along the base of the cuboid. Bone scintigraphy of the patients with normal radiograph showed hot spot in cuboid. Eleven patients visited our institution to seek for second opinion as their child has persistent limping gait.

Conclusions It is not always possible to make an early diagnosis of this fracture since the initial radiographic finding and physical examination are often negative.

Considering the consequences of a missed fracture and avoiding unnecessary treatment, bone scan might be useful in the early diagnosis of the stress fracture of the cuboid in young children.

Level of Evidence Diagnostic study, Level IV.

Keywords Stress fracture · Cuboid · Children

Introduction

Stress fractures in early childhood are not uncommon. Classic toddler's fracture, first described by Dunbar et al. [1], is a non-displaced spiral fracture of the tibia. Although tibia is the most commonly affected bone in toddlers, stress fracture of calcaneus and metatarsals has also been reported [2–4]. Cuboid fracture, on the other hand, has thought to be rare. However, there is an agreement that this fracture has been overlooked due to its benign nature. Recently, Senaran et al. [5] reported on the clinical and radiographic presentation of 28 preschool children with cuboid fracture and concluded that with awareness of this fracture, and a well-focused physical examination can help avoid delay in diagnosis. Additionally, bone scan is not routinely necessary to make the diagnosis. However, it is not always easy to perform a thoughtful physical examination in younger children because the child's irritability may make it difficult to localize the source of pain. Moreover, other conditions such as osteomyelitis or infection cannot be excluded when the pain is accompanied by local heat and swelling.

We present the clinical characteristics and radiographic findings, and the Tc99-m bone scintigraphy findings of the tarsal cuboid fracture in early childhood. This report will serve to reemphasize the usefulness of Tc99-m bone

S. Y. Joo

Department of Orthopedic Surgery, Incheon St. Mary's Hospital,
The Catholic University of Korea, Incheon, Korea

C. Jeong (✉)

Department of Orthopedic Surgery, Bucheon St. Mary's
Hospital, The Catholic University of Korea,
Bucheon 420-717, Korea
e-mail: changhoonj@naver.com

scintigraphy in the early detection of cuboid fractures in young children.

Materials and methods

Approval was obtained from our institutional review board for this retrospective study. From 2008 to 2012, we retrospectively reviewed the medical records and radiographs of 25 children who were diagnosed with stress fracture of cuboid at our institution. Patient demographics, age at presentation, duration of symptom, presence or absence of traumatic episodes, physical examination, and treatment methods were assessed from the medical records. Nutcracker test was applied as a provocation test, and it was performed as described by Senaran et al. [5] with the calcaneus stabilized, while the forefoot was abducted. This maneuver compresses the cuboid between the calcaneus and the base of the fourth and fifth metatarsals, causing pain on the cuboid area. A positive nutcracker test was defined whether the patient describes pain, or expressed irritability or guarding during the test. Standard anteroposterior, lateral, and oblique radiographs of the foot taken at the first, follow-up, and final visit were reviewed. Tc-99m bone scintigraphy was performed in all patients. Laboratory tests were performed in three children who had systemic fever or localized swelling on their affected foot to exclude infectious causes or inflammatory disease. Patients were treated with plaster splint immobilization for a short period. In addition, pain medicine such as nonsteroidal anti-inflammatory drugs was prescribed as needed.

Results

There were 13 boys and 12 girls with a mean age of 24.7 months (range 15–38 months). Right side was affected in 10 patients, and the left side was affected in 15. The average duration of symptom before visit was 7 days (range 2–14 days). In 6 of 25 patients (24 %), the parents or caregivers reported a traumatic episode (4 fall from a height and 2 stumbled), while the parents or caregivers of 19 patients (76 %) did not recall such an episode. Patterns of limping to refuse bear weight on the lateral border of the foot were variable. Toe walking was observed in 7 patients (28 %), heel gait was observed in 5 (20 %), and toeing out was noticed in 8 patients (32 %). Five patients (20 %) were able to bear weight on the lateral border of their affected foot, and their limping gait was characterized by a decreased stance phase of the affected limb. Nutcracker test was positive in 11 patients (44 %), while the test was negative in 4 patients (16 %). The test was unable to perform in 10 patients (40 %) due to poor compliance. In 15

patients, the radiographs of the affected foot showed sclerosis along the base of the cuboid suggesting fracture of the cuboid. The average duration of symptoms before visit in patients with radiographs showing a fracture of the cuboid was 9 days (range 7–14 days). In 10 of 25 patients (40 %), the initial plain radiographs of the foot showed no abnormal finding. The average duration of symptom of these patients was 4.5 days (range 2–7 days). However, bone scintigraphy of the patients with normal radiograph showed hot spot in cuboid (Fig. 1). Eleven patients (44 %) visited our institution to seek for second opinion as the child had a persistent limping gait. Radiographs of the foot had been taken in 8 patients at outside hospital and had a presumptive diagnosis of transient synovitis or growing pain. Limping gait returned to normal in all patients within 2 weeks after first visit without any adverse events. Symptom was relieved by short-term resting and pain medication. Radiographic change returns to normal within 6 months in all patients.

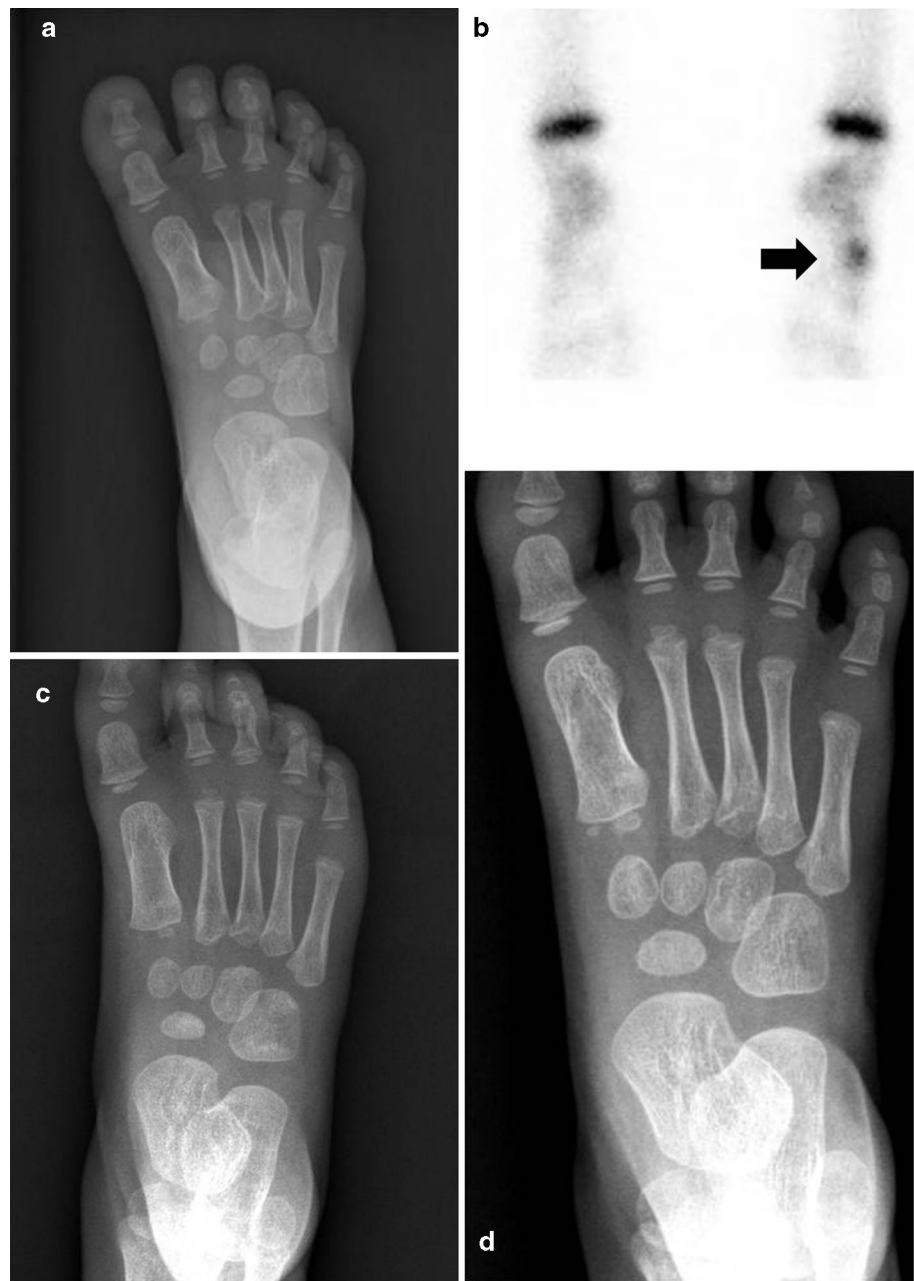
Discussion

Stress fractures are one of the causes of limping in young children. Unfortunately, it is not always possible to locate the cause of limping and pain in these patients. Young children are poor historians; they are frequently uncooperative when undergoing physical examinations and irritability of the child make it difficult to localize the source of pain. Furthermore, initial radiographs are usually normal.

Cuboid fracture in toddlers was once thought to be rare [2, 6–8]. However, recent studies showed that this fracture has been overlooked. According to Oestreich and Bhojwani [9], stress fracture of the cuboid was the second most common fracture of tarsal bone after calcaneus in children. We experienced 25 patients with cuboid fracture over 5-year period, which is similar to the study of Senaran et al [5] who reported 28 patients over 6-year period, suggesting that this fracture is common than we once thought. In our series, only 3 patients were noticed in the first year. However, after increased awareness of this fracture, there were an increasing number of patients with newly diagnosed fractures, and in the last year, we found 9 patients with cuboid fracture.

Most of the parents or caregivers (76 %) in our present study did not recall a traumatic episode. This can be explained in several ways. First, there are increasing numbers of children sent to a day care center in developed country. Therefore, in a chaotic environment, the injury may have gone unnoticed by the caregivers. Hermel and Gershon-Cohen [10] suggested a nutcracker mechanism of injury in cuboid fracture that is the cuboid is compressed

Fig. 1 A 2-year-old girl presented with limping gait for 7 days. Initial radiograph of the right foot showed no abnormal findings (**a**). Bone scintigraphy demonstrated an increased uptake in the cuboid (**b**). Focal sclerosis along the base of the cuboid appeared at 2 weeks on plain radiograph (**c**). Radiographic lesion return to normal at 6-month follow-up (**d**)



between the bases of the fourth and fifth metatarsal. Toddlers frequently walk in plantar-flexed foot, jump from a height and trip and stumble frequently. Therefore, it is not surprising that this injury mechanism can be commonly observed in toddlers while they perform normal activity. In the same context, as the younger children frequently jump from a height and frequently stumble, parents or caregivers may not regard these occurrences as traumatic episodes. Finally, repeated stress on cuboid rather than a single traumatic episode could be another explanation. Nicastro and Haupt [11] described a case of an 18-month-old girl with stress fracture of the cuboid. Since young children

step on plantar-flexed and everted foot while learning to walk, fracture can be caused by the chronic stress of an abnormal gait as the child learns to walk.

Halsey et al. [12] retrospectively reviewed the characteristics (i.e., history, gait, and physical examination) of 39 patients who were presumptively diagnosed as toddler's fracture. Sixteen of 39 patients (41 %) were confirmed on follow-up radiographs as sustaining a toddler's fracture. In their study, they were not able to find a particular characteristic to predict the outcome. In the present study, the pattern of limping and the avoidance of bearing weight on the lateral border of the affected foot were variable, and we could not

find a characteristic gait pattern. Nutcracker test was positive in 11 patients (44 %), and we were unable to perform it in 10 patients (40 %). We think that the rate was high because the mean age of the patients in our study was 24.7 months, which is relative younger than those reported in the literature [5–8, 10, 11]. Even in Senaran et al.'s [5] study, which emphasized careful physical examination in the diagnosis of cuboid fractures, only 13 of 28 patients showed a positive nutcracker test. Furthermore, they did not mention the number of patients in whom the test was unable to be performed on. Therefore, the test may not be useful in the diagnosis of stress fracture in younger children. However, since the nutcracker test was positive in 11 of 15 patients who were able to perform, we think that the test is useful in the diagnosis of cuboid fracture in children with good compliance.

Plain radiograph had limitation in the early diagnosis of cuboid fracture in our study. Radiographic finding was negative in patients who presented earlier than 7 days after the symptoms began. The radiograph in children who present later than 1 week showed sclerosis along the base of the cuboid. There are several other methods, such as ultrasonography and magnetic resonance imaging (MRI) used for the diagnosis of stress fracture. Studies demonstrated the usefulness of ultrasonography in the diagnosis of occult fractures [13, 14]. Simanovsky et al. [13] prospectively evaluated 58 children who sustained an acute ankle and wrist injury and were suspected as having a fracture despite normal radiographs. Fifteen patients with positive ultrasonographic findings were diagnosed as fracture on follow-up radiograph. The advantages of ultrasound imaging include bedside availability and the relative ease of performing repeated examinations. Imaging is a real time and free of harmful radiation. However, it is highly operator dependent. MRI is another excellent tool for identifying occult fractures. However, younger children are unable to lie still and sedation, and sometimes, general anesthesia may be necessary to obtain an adequate MRI examination. Therefore, many clinicians prefer a bone scintigraphy over MRI for the evaluation of an occult fracture, because it is less expensive and rarely requires sedation.

Although the usefulness of bone scan in the early diagnosis of occult fracture and as a diagnostic tool in patient with unclear etiology has been widely accepted, there is still disagreement on performing bone scintigraphy in the early diagnosis of this self-limiting condition [15–18]. In limping toddlers, Blumberg and Patterson suggested that when there is no clinical suspicion for infection or tumor, or when trauma to the foot was witnessed or otherwise well documented, it may be sufficient to obtain delayed radiographs after 10–14 days to avoid the additional risk and cost of scintigraphy [8]. Conversely, De

Boeck et al. [16] recommended performing bone scintigraphy to differentiate between fracture and bone infection since it is not always possible to differentiate these two conditions clinically or on plain radiograph. In our series, the initial radiograph of 10 patients showed no abnormalities, and the fracture was confirmed by bone scintigraphy and by the radiograph that was taken at the follow-up visit. We preferred to make a definitive diagnosis using bone scan for several reasons. First, limping gait resolved in all patients with short-term restriction of motion and pain medication without any adverse event, ascertained the benign nature of this fracture. One can argue that such mild injuries usually need only 2–3 weeks of casting or splinting. However, for children that do not have evidence of the fracture, this treatment could be shortened or abandoned. Moreover, cast-related problems such as skin irritation or heel sore can be avoided. In case of a fracture, casting or splinting may prevent unnecessary suffer and possible additional injury. Legal and insurance issues may also arise, and the possibility of hospital shopping cannot be ignored. Eleven of 25 patients visited our institution seeking for second opinion as the child had persistent limping. Eight of the 11 patients had radiographs taken at an outside hospital, which showed no abnormality, and they were given a presumptive diagnosis of transient synovitis or growing pain. We think that bone scintigraphy is indicated in children who presented with limping gait without obvious trauma history, in children who were unable to perform the nutcracker test, normal radiographic finding, and in children when other conditions such as inflammatory disorder and infection cannot be excluded.

In summary, stress fracture of the cuboid is not uncommon. Early diagnosis of this fracture is not always possible since the initial radiographic findings and physical examinations are often negative. Although this fracture is a self-limiting condition, bone scintigraphy might be useful for early diagnosis of the stress fracture to avoid unnecessary treatment and to avoid legal and insurance issues that may arise.

Conflict of interest The authors have no conflict of interest to disclose.

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