Case report

Spontaneous osteonecrosis of navicular and talus (SONNT)

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ABSTRACT

We describe a rare case of simultaneous and spontaneous osteonecrosis affecting bilateral navicular and talar bones in a young male. On both sides, there was associated symptomatic talonavicular and naviculocuneiform arthritis. The more symptomatic left foot was managed successfully by talonavicular-cuneiform (TNC) arthrodesis and core decompression of talar body. A probable cause of this rare disorder was most likely due to a developmental anomaly in the preaxial ray of the foot. As no literature is currently available for this rare condition, our case report is intended to provide a referral point for future studies in the early diagnosis and management of this condition.

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1. Introduction

Spontaneous osteonecrosis of navicular and talus simultaneously is an exceedingly rare occurrence, and until now no such cases have been reported in the literature. We report a rare case of osteonecrosis of talus and navicular, bilaterally in a young male. This unusual condition of spontaneous osteonecrosis of navicular and talus has been acronymed as SONNT, by us. It is characterized by idiopathic affection leading to the collapse of talar and navicular tarsal bones associated with osteoarthritic (OA) changes around the talonavicular-cuneiform (TNC) joints. It was managed successfully by arthrodesis of these affected joints.

2. Case report

One 24-year-old male presented with a history of pain in both feet (left > right), for 8 years in left foot and 1-year duration in right foot. This insidious onset of pain was dull aching and was progressive in nature. The pain was aggravated by walking and partially relieved with rest and medications. Over the period, pain progressed to a severity limiting his independent ambulation for less than 30 min. The patient denied any specific traumatic event

or associated constitutional symptoms. The patient received conservative management in the form of analgesics, rest, and ice packs, without any significant improvement in his symptoms.

The patient was nonalcoholic and nonsmoker with no history of chronic disease or any drug, particularly steroid, intake. Patient has no history of diabetes mellitus, hyperlipidemias, inflammatory arthritis, metabolic disorders, connective tissue, or an autoimmune disorder.

General examination was unremarkable, and the local examination revealed bilateral planovalgus deformities (Fig. 1) of the feet. These deformities were mobile and partially correctable after standing tiptoes. There was a restriction of supination and pronation movements of both the feet (L > R), and the ankle movements were preserved. No abnormality was found in blood investigations, with normal vitamin D, Hb electrophoresis, antinuclear antibodies, lipid profile, inflammatory markers, and rheumatoid arthritis factor tests. Plain radiographs of both the feet showed increased bone density and sclerosis in talus and navicular bones, with the partial collapse of involved tarsals with the fragmentation of navicular (Figs. 2 and 3). Arthritic changes were also seen in the TNC joints and were more pronounced in left foot. Magnetic resonance imaging (MRI) revealed bilaterally symmetrical loss of bony architecture involving articular surface of navicular and talar body with subchondral collapse (Figs. 4a, b and 5a, b). Underlying area revealed patchy areas of altered signal intensity showing heterogeneously hypointense signal on T1-weighted and heterogeneously hyperintense signal on T2-weighted, more prominent on the left side. Both radiograph and MRI findings

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Fig. 1. Pes planus deformity of both feet.

were consistent with osteonecrosis of navicular and talar bones, with secondary OA changes. The multiphasic bone scan was performed and revealed focal photopenia surrounding increased uptake in the bilateral talonavicular region representing vascular interruption, with no abnormal uptake elsewhere. Due to severe pain (VAS score: 7/10) in the left midfoot, TNC arthrodesis was performed, through a dorsomedial approach centering over navicular. A careful subperiosteal dissection was done to identify

talonavicular (TN), and naviculocuneiform (NC) joints and the osteophytes around these joints were removed. Articular cartilage from TNC was removed using a curette and both of these tarsal joints were denuded. An arthrodesis of TNC was achieved using one 80 mm long, 7.3 mm cancellous, cannulated screw (16 mm thread) and another 4.0 mm cancellous, cannulated screw to fix the NC joint. Fragments of the removed bone from the surface of the navicular body were placed in TN and NC joints to fill in small voids. The decompression of talar body was done using 4.0 mm cannulated drill over the guide wire, under image intensification. The bone specimen on histopathological examination revealed degenerative and reactive changes with evidence of empty lacuna, consistent with the diagnosis of osteonecrosis.

A below knee fiber cast was applied for 2 months. Radiograph of left foot at 6 months follow-up confirmed the sound fusion of TNC joints ([Fig. 6](#)). There was significant improvement postsurgically in the ankle-hindfoot score (from a preoperative score of 36 to postoperative score of 98), and the patient returned to his occupation as a businessperson satisfactorily.

3. Discussion

Although osteonecrosis of the isolated tarsal bones has been previously described in the talus and navicular bones ([Table 1](#)), the spontaneous osteonecrosis of navicular and talus (SONNT)

Fig. 2. Anteroposterior and lateral radiographs of left foot showing increased bone density and sclerosis in talus and navicular bones with deformation of navicular bone and osteoarthritic changes in the talonavicular-cuneiform joints.

Table 1

Characteristics of osteonecrosis of bone of preaxial ray of the foot.

Bone	Eponym	Authors	Year	Age	Etiology	Remark
(A) Navicular	Kohler	Kohler	1908	Adolescent	Vascular accidents	Indolent course
	Müller–Weiss syndrome	Müller	1927	Adult	Idiopathic	Progressive disease
		Konrad Weiss	1929			
(B) Talus	Diaz disease Mouchet's disease	Diaz Gracia F Mouchet	1928	Adult	Idiopathic	Self-limiting and nonprogressive
(C) Navicular and talus	SONNT	Vaishya et al.	2016	Adult	Developmental	Could be bilateral and associated with OA

Fig. 3. Anteroposterior and lateral radiographs of right foot showing increased bone density and sclerosis in talus and navicular bones and osteoarthritic changes in the talonavicular-cuneiform joints.

simultaneously has not been reported in the literature, as yet. Our case with bilateral involvement is a unique one, and we have coined a new terminology of SONNT to describe it. This idiopathic entity is characterized by symptomatic osteonecrosis in both the navicular and talar bones simultaneously in an adult, with associated OA of the involved joints. Various hypotheses have been put forward to explain the etiology of osteonecrosis in talus and navicular bones. The possible reasons for its occurrence in atraumatic variety include the following: (a) an insufficient blood supply, affecting aerobic metabolism leading to ischemic necrosis of bones and (b) developmental reasons.¹ The other possible confounding and risk factors include alcoholism, chronic steroids use, connective tissue disorders (such as systemic lupus erythematosus), metabolic disorders (such as hyperlipidemia), sickle cell disease, and radiation therapy.^{2–4}

Müller, in 1927, first described an isolated osteonecrosis of the navicular bone.^{5,6} Initially, he thought that the chronic deformation led to dynamic compression of tarsus. It was manifested by medial and dorsal protrusion of navicular bone and a comma-shaped deformity due to the collapse of a lateral portion of the bone. However, 1 year later, he proposed that the affection was congenital and not due to compression injury, as described by him earlier. Konrad Weiss (1929) later described osteonecrosis of navicular to be a primary pathology resulting from disruption of blood supply to the navicular.⁷ Müller–Weiss syndrome in adults needs to be differentiated from a well-recognized Kohler's disease where spontaneous osteonecrosis occurs in navicular bone, in children. Clinically, Kohler's disease is marked by an indolent course, whereas Müller–Weiss syndrome is a chronic slowly progressive disease resulting in disabling pain and deformity.⁸ We suggest that the SONNT should be differentiated from bone changes occurring secondary to stress fracture, Charcot's neuroarthropathy, Kohler's disease, osteomyelitis, Paget's disease, and metabolic disorders, such as systemic lupus erythematosus.

These conditions can be differentiated based on history, clinical examination, and blood investigations, which in our case did not show any abnormality. However, SONNT cannot be differentiated

from secondary causes of osteonecrosis only on the radiological findings. In this instance, the bilateral affection of talar and navicular bones simultaneously can be explained by an abnormal embryological development. Morphologically, the foot is divided into two cranial (preaxial) and caudad (postaxial) regions linked with the tibia and fibula,⁹ because the interosseous groove of the leg extends onto the ventral aspect of the foot. The second ray, talus, navicular, and cuneiform constitute the preaxial region, whereas the third ray, cuboid, and calcaneus form the postaxial region. Here, the involvement of two adjoining preaxial ray bones (i.e., talus and navicular) together strongly supports the possibility of a developmental cause.

There are no clear guidelines available for the treatment of osteonecrosis of tarsus bones. We believe that an early presentation of SONNT can be managed with protected weight bearing with supportive orthotics,¹⁰ anti-inflammatory medications, and calcium and vitamin D supplementation. Those who are presenting late with established arthritic changes need surgical intervention in the form of arthrodesis of arthritic joints and restoration of the medial longitudinal arch, with or without core decompression of the talus.

Various surgical procedures available for management of SONNT could be an arthrodesis of TNC joint, core decompression of talus, triple arthrodesis, pantalar arthrodesis, excision of fragmented navicular bone, and medial longitudinal arch reconstruction. Triple arthrodesis does not address the pathology at NC joint and restricts the subtalar joint mobility, and hence it is not a good option for TNC involvement in SONNT.¹¹ Pantalar arthrodesis is a more radical surgery and has additional drawback of limitation of ankle movements.¹²

The TNC arthritis can be managed surgically by different methods. An arthrodesis is less destructive and has more favorable outcomes. This procedure has the additional advantage of the restoration of the medial longitudinal arch. It can be done with the help of cannulated screws or with low profile plates. de Retana et al. advocated the use of bone graft (from iliac crest) to achieve fusion at TNC joint using a dorsal approach between the tibialis anterior and extensor hallucis longus tendons.¹⁰ He also

Fig. 4. (a, b) Sagittal T1-weighted, (a) coronal T2-weighted, and (b) magnetic resonance imaging sequences of the left foot. Patchy areas of altered signal intensity showing heterogeneously hypointense signal on T1-weighted images and heterogeneously hyperintense signal on T2-weighted images, suggestive of edema with the subchondral collapse of talar dome and loss of bony architecture and fragmentation of navicular.

Fig. 5. (a, b) Sagittal T1-weighted, (a) coronal T2-weighted, and (b) magnetic resonance imaging sequences of the right foot revealed focal serpiginous altered signal intensity showing heterogeneously hypointense signal on T1-weighted images and heterogeneously hyperintense signal on T2-weighted images, suggestive of edema with the involvement of articular surface of talar dome with slight depression and fragmentation of navicular.

discouraged routine internal fixation of the graft, as it was thought of having inherent self-locking properties. In 2011, Tan et al. advocated complete excision of fragmented navicular and reconstruction of the medial longitudinal arch with the help of allograft between the talus and medial cuneiform.¹³ Stabilization of the construct was achieved using a low profile plate extending from talus to medial cuneiform. In 2012, Cao et al. described TNC arthrodesis with a dorsal-based closing wedge osteotomy through

talonavicular joint. This process has the advantage of regaining medial longitudinal arch height.¹⁴ We preferred using partially threaded cannulated screws for TNC arthrodesis because of their less invasive nature and good stability.

Tibiotalar fusion is not encouraged because of longer operative time and more chances of nonunion pseudoarthrosis.^{15–17} Urquhart et al. reported unfavorable outcome and a longer period of immobilization.¹⁸ On the other hand, core decompression provides satisfactory results for symptomatic patients and is associated with less morbidity, and hence it was used to treat the talar lesion in this case. Marulanda and co-workers studied the

Fig. 6. Follow-up radiographs (anteroposterior and oblique views) showing TNC fusion.

results of core decompression treating osteonecrosis in 44 patients and reported 91% successful clinical outcome.¹⁹ Delanois et al. also reported favorable outcomes with core decompression in avascular necrosis of talus.²⁰

Conflicts of interest

The authors have none to declare.

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