CASE REPORT

An incidental talonavicular coalition in a diabetic patient: a podiatric perspective

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SUMMARY
A talocalcaneal coalition is a pathological union of two or more tarsal bones. The authors present an incidental finding of a unilateral talonavicular (TN) coalition that was overlooked in a 57-year-old diabetic female with signs and symptoms of peripheral neuropathy. This case highlights the clinical implications and important teaching points in recognising a TN coalition. This is particularly relevant for new, upcoming clinicians who may have never been exposed to this diagnostic rarity during clinical training.

BACKGROUND
The incidence of tarsal coalitions is believed to be less than 2% in the general population. The most recognised deformity in tarsal coalitions to date is the flatfoot deformity (pes planus). Our clinical finding of flat foot is in agreement with previously published research. To our knowledge, no case report has previously described a talonavicular (TN) coalition in the setting of diabetes. One of the earliest manifestations of diabetes is peripheral neuropathy, usually in the lower extremities. Thus, if the coalition were to be symptomatic (painful), this would make the diagnosis more difficult because of the patient’s neuropathic symptoms.

Gait analysis has revealed that TN coalition leads to increased pressure under the first metatarsophalangeal joint, with hyperkeratosis under the first metatarsal head. As podiatric physicians, we believe that it is particularly important to treat hyperkeratotic lesions in patients with diabetic peripheral neuropathy because they can progress into limb-threatening ulcerations in the future.

The abnormal union of tarsal bones may lead to excessive strain on other joints proximally in an effort to regain lost motion distally. In this case the subtalar joint (STJ) would be the primary compensatory joint. This increased strain is expected to lead to arthritic changes, but, interestingly, this patient did not develop such changes in the STJ.

We present a unique case of an incidentally discovered unilateral TN coalition in a diabetic patient with systemic co-morbidities. In addition, this case highlights the important teaching point that a coalition can be missed in radiographs due to operator error. As was the case in this patient, the coalition was mistaken for an enlarged talar head in 2009 and 2012. Although previously reported in the literature, a TN coalition is still rarely encountered and must be kept in mind by new, upcoming clinicians who may have never seen one during training.

CASE PRESENTATION
A 57-year-old female presented to a local outpatient podiatry clinic with a chief complaint of bilateral foot pain. She described more than a year of pain on the dorsum of both feet that radiated to her toes. The patient described the pain as a ‘burning’ sensation. She had been managing the pain with acetaminophen and naproxen. Past medical history indicated type II diabetes for over 20 years, with her most recent haemoglobin A1c being 8.9%. The patient had been undergoing physical therapy for previous heel pain that was relieved with a steroid injection. The orthopaedic component of the physical examination revealed hallux limitus of the right foot, severe flatfoot, and forefoot varus of the left foot. Pain was elicited with palpation of the dorsum of the left foot.

INVESTIGATIONS
Radiographs were taken of both feet in 2009. The left was negative for a TN coalition (figure 1A, B). However, on the anterior-posterior (AP) view, the right rearfoot was poorly visualised because of low milliamperage while the radiograph was being taken (figure 2A). Thus, the TN coalition was not visible. Retrospectively, the lateral view taken of the right foot in 2009 (figure 2B) was positive for a TN synostosis when compared with the lateral view (figure 3C) taken in 2012. However, the diagnosis of a TN coalition was not made in 2009. The authors believe that the coalition was most likely perceived as an enlarged talar head articulating with the cuneiforms, and thus was missed (figure 3B). On retrospective review of the written interpretation of the radiographs taken in 2012 (figure 3A–C), there was no mention of a TN coalition. However, when the radiographs taken in 2012 (figure 3A–C) were personally reviewed, a TN synostosis was evident. The coalition was once again most likely perceived as an enlarged talar head articulating with the cuneiforms.

OUTCOME AND FOLLOW-UP
The patient was subsequently lost to follow-up, and no further imaging or treatment modalities could be employed.

DISCUSSION
A tarsal coalition is defined as an abnormal union between two or more tarsal bones that may result in restricted or absent range of motion at the affected joint. Union between the talus and navicular bones is called a TN coalition. Coalitions

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are classified as either congenital or acquired in origin. Congenital coalitions are more common than the acquired type. The currently accepted aetiology of congenital tarsal coalitions is a failure of primitive mesenchymal tissue to differentiate with autosomal dominant inheritance, although the literature suggests an autosomal recessive inheritance pattern is possible. Tarsal coalitions may be bony (synostosis), cartilaginous (synchondrosis) or fibrous (syndesmosis) in nature.

Talocalcaneal (TC) and calcaneonavicular (CN) coalitions are generally the most common types of rearfoot coalitions, with reported incidences of 48.1% and 43.6% among individuals with coalitions, respectively. The incidence of TN coalition has been estimated to be approximately 1.3%, with fewer than 50 cases described in the literature. However, with the use of advanced imaging modalities, authors have concluded that tarsal coalitions are likely underdiagnosed, especially when radiographs are employed. The calculated incidence is 13% using CT imaging and 11% using MRI.

In 1879 Anderson described the first TN coalition. O’Donoghue and Sell were the first to utilise radiographs to identify a case of bilateral symmetrical TN synostosis. Boyd reported a case of bilateral congenital TN synostosis, and stated that the coalition had no clinical significance. Schreiber described a case of familial bilateral TN coalitions with an associated ball and socket ankle joint. Challis presented a case of a TN coalition and its relationship to abnormalities in the hand, and concluded that this coalition is inherited in an autosomal dominant pattern. Zeide suggested an autosomal recessive mode of inheritance. Yeates was the first to report a case of TN synostosis in twins as well as their biological mother. Ertel and O’Connell described the first case of an acquired TN coalition following avascular necrosis of the navicular. Kramhoft and Monberg described a case of a unilateral TN coalition that was overlooked on radiographic analysis. Lahaye identified bilateral involvement in more than half of TC and TN coalitions. Bonk and
Tozzi\textsuperscript{22} identified an acutely painful TN synostosis in an athlete. Pontious \textit{et al.}\textsuperscript{5} performed objective gait analysis in a patient with a bilateral TN coalition, establishing an altered gait pattern. David \textit{et al.}\textsuperscript{23} were the first to utilise a foot orthosis specifically tailored to a paediatric patient with a TN coalition. Doyle and Kumar\textsuperscript{13} were able to surgically treat paediatric patients who had midfoot pain caused by a TN coalition. Frost and Fagan\textsuperscript{24} reported a case of simultaneous bilateral TN and CC (calcaneocuboid) coalitions. Migues \textit{et al.}\textsuperscript{25} reported a case of symptomatic TN coalition with associated pedal anomalies. Bryson \textit{et al.}\textsuperscript{26} described the first case of a TN and TC coalition with associated ankle instability. This coalition is either symptomatic (painful) or asymptomatic (painless). The symptomatic type has been found to correlate with ossification of the coalition, specifically at 3–5 years of age for the TN coalition, 8–12 years of age for the CN coalition, and 12–16 years of age for the TC coalition.\textsuperscript{28} Asymptomatic (pain free) TN coalitions are more common.\textsuperscript{7} A medial hard prominence rather than pain has been shown to be a clinical finding associated with TN coalitions.\textsuperscript{21}

Radiographic examination of tarsal coalitions begins with X-ray imaging. The most common views taken for rearfoot tarsal coalitions are AP, lateral and medial oblique.\textsuperscript{28} AP and lateral are the best views for demonstrating a TN coalition.\textsuperscript{7} A well-circumscribed navicular or a decrease in TN joint space, rounding of the naviculocuneiform joint, and beaking of the navicular are findings suggestive of a TN coalition on radiographic analysis.\textsuperscript{23} It is important to note that clinicians may often miss a TN coalition on X-ray; a common error is mistaking an enlarged talar head for a TN coalition.\textsuperscript{29}

\begin{figure}[h]
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\caption{(A) (Taken in 2009): Weight bearing, anterior-posterior view of the right foot. Note the radiograph was taken at low millamperage and so the talonavicular (TN) synostosis could not be visualised. (B) (Taken in 2009): Weight bearing, lateral view of the right foot indicating a TN synostosis. A plantar calcaneal spur is also evident.}
\end{figure}
Treatment options for tarsal coalitions range from conservative to surgical interventions. Conservative therapy is considered first-line, while surgical intervention is utilised if conservative therapy fails. Surgical correction of a TN coalition has been demonstrated using either an osteotomy or arthrodesis.

Interestingly, recent research has shown that the incidence of tarsal coalitions may vary according to the population studied. In particular, examination of skeletal remains has suggested variability of tarsal coalitions among different populations. Burnett and Case demonstrated with statistical significance that naviculocuneiform coalitions were more prevalent among South African Bantu individuals than those with European ancestry. Moreover, Burnett and Wilczak reported that the patterns of hindfoot coalitions (TN, TC, CN and CC) vary across populations, with higher frequencies in Asian populations.

**Figure 3** (A) (Taken in 2012): Non-weight bearing, anterior-posterior view of the right foot indicating a talonavicular (TN) synostosis. There is evidence of mild degenerative changes in the midfoot. There is no evident fracture or dislocation. (B) (Taken in 2012): Non-weight bearing, medial oblique view of the right foot indicating a TN synostosis. (C) (Taken in 2012): Non-weight bearing, lateral view of the right foot revealing a TN synostosis. A plantar calcaneal spur was also noted.

**Learning points**

- Diabetic peripheral neuropathy can mask a symptomatic coalition.
- Compensatory mechanisms in the form of excessive strain (motion) in proximal joint(s) may be present leading to symptoms in those joints.
- Many clinicians assume that the talar head is enlarged when assessing a talonavicular (TN) coalition on plain radiographs, leading to a misdiagnosis.
- In this case operator error masked evidence of a coalition on radiograph.
- The TN coalition is the most rarely reported tarsal coalition in the foot.
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Competing interests None.

Patient consent Obtained.

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