

CASE REPORT

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

Garry Shtofmakher,¹ Adam Rozenstrauch,¹ Randy Cohen²

¹New York College of Podiatric Medicine, New York, New York, USA
²Department of Radiology, New York College of Podiatric Medicine, New York, New York, USA

Correspondence to
 Garry Shtofmakher,
 gshtofmakher@nycpm.edu

Accepted 16 April 2014

SUMMARY

A tarsal 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.^{1 2} The most recognised deformity in tarsal coalitions to date is the flatfoot deformity (pes planus).³ 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.⁴ 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.⁵ 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.⁶

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.⁷ Union between the talus and navicular bones is called a TN coalition. Coalitions



CrossMark

To cite: Shtofmakher G, Rozenstrauch A, Cohen R. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2014-204510

Figure 1 (A) (Taken in 2009): Weight bearing, anterior-posterior view of the left foot. There is no evidence of fracture or dislocation, no lytic or sclerotic lesions were noted, and there was no fusion between the talus and navicular. (B) (Taken in 2009): Weight bearing, lateral view of the left foot. The talus is not fused with the navicular.



are classified as either congenital or acquired in origin. Congenital coalitions are more common than the acquired type.⁷ The currently accepted aetiology of congenital tarsal coalitions is a failure of primitive mesenchymal tissue to differentiate with autosomal dominant inheritance,⁸ although the literature suggests an autosomal recessive inheritance pattern is possible.⁹ Tarsal coalitions may be bony (synostosis), cartilaginous (synchondrosis) or fibrous (syndesmosis) in nature.¹⁰

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.¹ The incidence of TN coalition has been estimated to be approximately 1.3%, with fewer than 50 cases described in the literature.^{7 10} 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 imaging¹¹ and 11% using MRI.¹²

In 1879 Anderson described the first TN coalition.¹³ O'Donoghue and Sell¹⁴ were the first to utilise radiographs to identify a case of bilateral symmetrical TN synostosis. Boyd¹⁵ reported a case of bilateral congenital TN synostosis, and stated that the coalition had no clinical significance. Schreiber¹⁶ described a case of familial bilateral TN coalitions with an associated ball and socket ankle joint. Challis¹⁷ 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 *et al*⁹ suggested an autosomal recessive mode of inheritance. Yeates¹⁸ was the first to report a case of TN synostosis in twins as well as their biological mother. Ertel and O'Connell¹⁹ described the first case of an acquired TN coalition following avascular necrosis of the navicular. Kramhoft and Monberg²⁰ described a case of a unilateral TN coalition that was overlooked on radiographic analysis. Lahey *et al*²¹ identified bilateral involvement in more than half of TC and TN coalitions. Bonk and

Figure 2 (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.



Tozzi²² identified an acutely painful TN synostosis in an athlete. Pontious *et al*⁵ performed objective gait analysis in a patient with a bilateral TN coalition, establishing an altered gait pattern. David *et al*²³ were the first to utilise a foot orthosis specifically tailored to a paediatric patient with a TN coalition. Doyle and Kumar¹³ were able to surgically treat paediatric patients who had midfoot pain caused by a TN coalition. Frost and Fagan²⁴ reported a case of simultaneous bilateral TN and CC (calcaneocuboid) coalitions. Migueis *et al*²⁵ reported a case of symptomatic TN coalition with associated pedal anomalies. Bryson *et al*²⁶ described a case of symptomatic bilateral TN coalition with associated co-morbidities spanning two decades. Brennan *et al*²⁷ 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.²⁸ Asymptomatic (pain free) TN coalitions are more common.⁷ A medial hard prominence rather than pain has been shown to be a clinical finding associated with TN coalitions.²³

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.²⁸ AP and lateral are the best views for demonstrating a TN coalition.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.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.²⁹

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.



Treatment options for tarsal coalitions range from conservative to surgical interventions. Conservative therapy is considered first-line,³⁰ while surgical intervention is utilised if conservative therapy fails. Surgical correction of a TN coalition has been demonstrated using either an osteotomy or arthrodesis.³¹

Interestingly, recent research has shown that the incidence of tarsal coalitions may vary according to the population studied.^{32–35} In particular, examination of skeletal remains has suggested variability of tarsal coalitions among different populations.^{34, 35} Burnett and Case demonstrated with statistical significance that naviculocuneiform coalitions were more prevalent among South African Bantu individuals than those with European ancestry.³⁴ 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.³⁵

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.

Acknowledgements We would like to thank Dr Khurram Khan, DPM for helping us identify the TN coalition. Dr Khan has been a great educator, clinician and mentor.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- Stormont DM, Peterson HA. The relative incidence of tarsal coalition. *Clin Orthop Relat Res* 1983;181:28–36.
- Harris RI, Beath T. Etiology of peroneal spastic flat foot. *J Bone Joint Surg Br* 1948;30B:624–34.
- Mosier KM, Asher M. Tarsal coalitions and peroneal spastic flat foot. A review. *J Bone Joint Surg Am* 1984;66:976–84.
- American Diabetes Association. Standards of medical care in diabetes—2014. *Diabetes Care* 2014;37(Suppl 1):S14–80.
- Pontious J, Hillstrom HJ, Monahan T, et al. Talonavicular coalition. Objective gait analysis. *J Am Podiatr Med Assoc* 1993;83:379–85.
- DuVries H. *Anomalies and congenital defects. Surgery of the foot*. St. Louis: C.V. Mosby, 1959:431.
- Downey MS, Dewaters AM. Tarsal coalition. In: Southerland JT, ed. *McGlamry's comprehensive textbook of foot and ankle surgery*. 4th edn. New York: Lippincott Williams & Wilkins, 2012;126:598–635.
- Leonard MA. The inheritance of tarsal coalition and its relationship to spastic flat foot. *J Bone Joint Surg Br* 1974;56B:520–6.
- Zeide MS, Wiesel SW, Terry RL. Talonavicular coalition. *Clin Orthop Relat Res* 1977;225–7.
- Zaw H, Calder JD. Tarsal coalitions. *Foot Ankle Clin* 2010;15:349–64.
- Solomon LB, Ruhli FJ, Taylor J, et al. A dissection and computer tomograph study of tarsal coalitions in 100 cadaver feet. *J Orthop Res* 2003;21:352–8.
- Nalaboff KM, Schweitzer ME. MRI of tarsal coalition: frequency, distribution, and innovative signs. *Bull NYU Hosp Jt Dis* 2008;66:14–21.
- Doyle SM, Kumar SJ. Symptomatic talonavicular coalition. *J Pediatr Orthop* 1999;19:508–10.
- O'Donoghue DH, Sell LS. Congenital talonavicular synostosis a case report of a rare anomaly. *J Bone Joint Surg* 1943;25:925–7.
- Boyd HB. Congenital talonavicular synostosis. *J Bone Joint Surg* 1944;26:682–6.
- Schreiber RR. Talonavicular synostosis. *J Bone Joint Surg* 1963;45:170–217.
- Challis J. Hereditary transmission of talonavicular coalition in association with anomaly of the little finger. *J Bone Joint Surg Am* 1974;56:1273–6.
- Yeates HA. Talonavicular synostosis. *Ulster Med J* 1980;49:165–8.
- Ertel AN, O'Connell FD. Talonavicular coalition following avascular necrosis of the tarsal navicular. *J Pediatr Orthop* 1984;4:482–4.
- Kramhoft M, Monberg J. Total talonavicular coalition. A case report. *Acta Orthop Belg* 1988;54:90–1.
- Lahey MD, Zindrick MR, Harris EJ. A comparative study of the clinical presentation of tarsal coalitions. *Clin Podiatr Med Surg* 1988;5:341–57.
- Bonk JH, Tozzi MA. Congenital talonavicular synostosis. A review of the literature and a case report. *J Am Podiatr Med Assoc* 1989;79:186–9.
- David DR, Clark NE, Bier JA. Congenital talonavicular coalition. Review of the literature, case report, and orthotic management. *J Am Podiatr Med Assoc* 1998;88:223–7.
- Frost RA, Fagan JP. Bilateral talonavicular and calcaneocuboid joint coalition. *J Am Podiatr Med Assoc* 1995;85:339–41.
- Migues A, Slullitel GA, Suarez E, et al. Case reports: symptomatic bilateral talonavicular coalition. *Clin Orthop Relat Res* 2009;467:288–92.
- Bryson D, Uzoigwe CE, Bhagat SB, et al. Complete bony coalition of the talus and navicular: decades of discomfort. *BMJ Case Rep* 2011;2011:pil: bcr0320114031.
- Brennan SA, Kiernan C, Maleki F, et al. Talonavicular synostosis with lateral ankle instability—A case report and review of the literature. *Foot Ankle Surg* 2012;18: e34–6.
- Cowell HR, Elener V. Rigid painful flatfoot secondary to tarsal coalition. *Clin Orthop Relat Res* 1983;177:54–60.
- Kernbach KJ. Tarsal coalitions: etiology, diagnosis, imaging, and stigmata. *Clin Podiatr Med Surg* 2010;27:105–17.
- Varner KE, Michelson JD. Tarsal coalition in adults. *Foot Ankle Int* 2000;21: 669–72.
- Ellington JK, Myerson MS. Surgical correction of the ball and socket ankle joint in the adult associated with a talonavicular tarsal coalition. *Foot Ankle Int* 2013;34:1381–8.
- Byun SE, Lee HS, Ahn JY, et al. Treatment of naviculo-first cuneiform coalition of the foot. *Foot Ankle Int* 2014;35:489–95.
- Kumai T, Tanaka Y, Takakura Y, et al. Isolated first naviculocuneiform joint coalition. *Foot Ankle Int* 1996;17:635–40.
- Burnett SE, Case DT. Naviculo-cuneiform I coalition: evidence of significant differences in tarsal coalition frequency. *The Foot* 2005;15:80.
- Burnett SE, Wilczak CA. Tarsal and tarsometatarsal coalitions from Mound C (Ocmulgee Macon Plateau site, Georgia): implications for understanding the patterns, origins, and antiquity of pedal coalitions in Native American populations. *Homo* 2012;63:167–81.

Copyright 2014 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow