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Talocalcaneal coalition in a 15 year old female basketball player

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This case reports an adolescent athlete with activity related chronic bilateral dorsal foot pain and stiffness. A 15 year old competitive female basketball player presented to a chiropractor subsequent to an unsuccessful course of conservative treatment for posterior tibial dysfunction. The patient's plain films were incorrectly read as normal and a CT scan obtained by the radiologist called the findings bilateral osteoarthritis. The patient was awaiting a referral to a rheumatologist at the time of initial consultation with the chiropractor. Examination revealed limited subtalar mobility and review of the images revealed bilateral non-osseous talocalcaneal coalition. The patient was subsequently directed to a pediatric orthopedic surgeon and is scheduled for a resection of the coalition. Primary care practitioners should be aware of this uncommon, but not rare, variable clinical presentation as misdiagnosis and mismanagement could lead to suboptimal patient outcomes. To our knowledge this is the first case report of a patient with tarsal coalition published in chiropractic literature. In addition, this case is the first to report radiographic evidence of chronic mechanical stress to the second metatarsal associated with tarsal coalition.
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KEY WORDS: adolescent, tarsal coalition, talocalcaneal

Ce cas concerne une athlète adolescente souffrant de douleurs et de raideur chroniques à la région dorsale du pied bilatéral. Une joueuse de basketball de 15 ans se présenta chez un chiropraticien suite à une série de traitements conservateurs n'ayant pas réussi à guérir un dysfonctionnement tibial postérieur. Les radiographies de la patiente furent jugées normales à tort, et un tomodensitogramme obtenu par le radiologue ont démontré de l'arthrose bilatérale. La patiente attendait d'être recommandée à un rhumatologue au moment de sa première consultation avec le chiropraticien. Un examen a révélé une mobilité dans l'articulation sous-astragalienne, et un examen des radiographies révéla une synostose astragalocalcanéenne non osseuse bilatérale. La patiente fut envoyée à un chirurgien orthopédique pédiatrique, et doit subir une résection de la synostose. Les médecins de premier recours doivent savoir que cette présentation clinique variable peu courante, mais pas rare, constitue un mauvais diagnostic et peut entraîner des résultats sous-optimaux pour les patients. Selon nos connaissances, c'est la première fois où l'on fait état dans une publication sur la chiropratique d'un patient souffrant d'une synostose du tarse. En outre, c'est la première fois que radiographies démontrent un stress mécanique chronique au second métatarsien associé à la synostose du tarse.
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MOTS CLÉS : adolescente, synostose du tarse, astragalocalcanéenne

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Introduction

Congenital tarsal coalition is a diagnosis that is often overlooked in young patients who first present with foot and ankle pain.[1] Tarsal coalition is an abnormal fibrous, cartilaginous or osseous fusion of two or more tarsal bones.[2,3] The prevailing etiologic theory for congenital coalitions, is failure of complete segmentation of mesenchyme with the absence of normal joint formation.[3–5]

Approximately 50%–80% of cases of tarsal coalition are bilateral,[4,6] and more than 90% of coalitions are located between the calcaneus and the navicular or between the talus and calcaneus.[3,6] Calcaneonavicular and talocalcaneal coalitions have been reported to occur in approximately equal frequency,[6,7] however, recent investigations suggest that calcaneonavicular coaltions occur 3 times more frequently.[2,8]

The true prevalence of tarsal coalition in the general population is unknown. However, conservative estimates from x-ray diagnostic series' report a prevalence of approximately 1%.[1] A recent prospective analysis of 674 consecutive patients referred for ankle MRI's at the NYU Hospital for Joint Diseases demonstrated a 12% prevalence of tarsal coalition.[2] Thus the prevalence of tarsal coalition in clinical populations may in fact be much higher than is commonly reported.

According to the practice analysis of Chiropractic conducted in 2003 in the United States, lower extremity conditions contribute to 8.8% of the chief clinical complaints for which a patient seeks care from a chiropractor.[9] To our knowledge there have not been any cases of tarsal coalition published in chiropractic literature. The purpose of this diagnostic case report is to create awareness of an uncommon, but not rare, clinical condition that may present to chiropractors. This report details the complex diagnosis of bilateral non-osseous talocalcaneal coaltion in a 15 year old female with chronic bilateral activity-related dorsal foot pain and stiffness who was previously misdiagnosed and mismanaged.

Case Report

A 15 year-old female, competitive basketball player, consulted a chiropractor with a 2 year history of bilateral foot pain, worse on the left. Her chief complaint was a dull and achy pain across the dorsal aspect of the foot, in the region of the talus, but was poorly localized. An occasional sharp pain in the medial hindfoot with activity was also noted by the patient. A secondary complaint was bilateral stiffness with global ankle movement. The patient attributed her activity-related foot pain over the past few years to the "Active Ankle" braces she was required to wear by her team to prevent ankle sprains. The patient did not have a history of acute trauma.

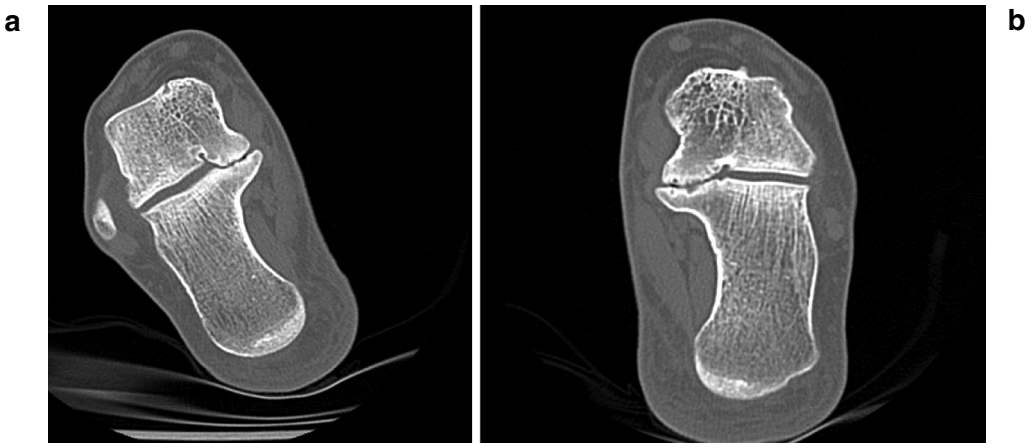
When she was 12 years of age she was prescribed custom fit orthotics to help manage bilateral activity related knee pain. At 13 years of age she experienced occasional bilateral dorsal foot pain after a lot of walking, which was alleviated initially by regularly wearing her orthotics.

The intensity of her symptoms rapidly increased in the previous five months as she was training 5 days per week, 2 times per day in preparation for a regional development basketball camp. She was also training for track and field during this period. The increase in dorsal foot pain forced her to leave the basketball camp and she has been unable to return to play.

At the time of her chiropractic consultation her medical care to date had included a recent visit to her family doctor. Plain films were taken of the left ankle, which were repeated two weeks later. Both sets of plain films were read as normal. A bone scan was subsequently ordered to investigate the possibility of metatarsal stress fractures. The bone scan was unremarkable in the region of the metatarsals, but showed abnormal focal uptake at the posterior and medial aspects of the talus bilaterally. A diagnosis of osteochondritis dessicans was suspected by the radiologist who subsequently recommended a CT scan of both ankles. The CT scan showed irregular articular contours, narrowed joint space and subchondral cysts with sclerosis in the middle subtalar joints in both ankles, worse on the left (Figure 1). The CT report impression was bilateral osteoarthritis and subsequently the family doctor referred the patient to a rheumatologist. The patient was awaiting consultation with a rheumatologist at the time of the initial chiropractic consultation.

Her medical care also included regular physiotherapy since being forced to stop playing basketball. The physiotherapist treated her for posterior tibial dysfunction. Treatment included soft tissue friction, ultrasound, wobble board exercises and strengthening exercises. The patient found the visits painful and they aggravated her condition, but she was encouraged to continue with the plan of management. This continued for approximately three months with no improvement in the foot and ankle

Figure 1 *Non enhanced coronal CT images of the right (image a) and left (image b) medial subtalar joint demonstrating small joint spaces with irregular articular contours, subchondral cyts and sclerosis, which appears more advanced on the left (image b). These findings were interpreted by the initial radiologist as bilateral osteoarthritis, but were eventually interpreted by a 2nd chiropractic radiologist as incomplete osseous talocalcaneal coalition.*



pain. She also consulted the MD who had made her last pair of orthotics, who prescribed and fitted the patient for a new pair.

Upon examination the patient had moderate pes planus and moderate rear foot valgus on static observation. She walked with mild bilateral forefoot abduction, but gait was otherwise unremarkable. Range of motion testing revealed bilaterally restricted dorsiflexion, inversion and eversion worse on the left side. Plantar flexion was full and pain free bilaterally. Passive inversion recreated her sharp pain posterior to the medial malleolus on the left. Motion palpation of the subtalar joint was significantly limited bilaterally, particularly in eversion. Resisted range of motion was full and pain free. Pain on palpation was non-specific, in the region of the talus bilaterally. Functional testing, including hopping on one foot, aggravated the patients' chief complaint of dorsal foot pain.

All existing imaging was requested from the patient. Upon reviewing the lateral view of the left foot plain films (Figure 2) an abnormal cortical confluence extending from the talus to the sustentaculum tali was observed by the chiropractor. This radiographic finding has been termed the "C" sign and is suggestive of talocalcaneal coalition. The bone scan and CT findings, previously reported, supported this differential as well.

The chiropractor sent these images to a chiropractic radiologist, who confirmed the suspicion of a C-sign along with non-visualization of the middle subtalar facet joint on the plain films. His interpretation of the plain films in combination with the bone scan and CT results confirmed the diagnosis of bilateral incomplete talocalcaneal coalition. Diffuse cortical thickening along the shaft of the 2nd metatarsal in the plain film AP view of the left foot was also noted at that time.

The chiropractor directed the patient to send the chiropractic radiologist's report and imaging directly to a hospital specializing in pediatric orthopedics. The orthopedic department agreed with the image findings and interpretation and subsequently scheduled a consultation with a pediatric orthopedic surgeon. The patient was advised of a 30% chance of satisfactory resolution of her symptoms with a below knee walking cast by the orthopedic surgeon. The cast was placed on the left leg for 6 weeks, then the right for 6 weeks. This treatment did not change the patient's symptoms and surgery was subsequently scheduled for the excision of the left talocalcaneal coalition. Surgery on the right foot is expected to follow 6 weeks later.

While waiting for the orthopedic consultation the patient received a cortisone injection in the left ankle. This treatment was self-directed. The treatment was not successful and was not repeated on the right ankle.

A second chiropractic consultation after the casting

Figure 2 *Plain film lateral view (image b) of the left foot demonstrates non-visualization of the middle subtalar joint and is associated with a "C" sign of the confluence of medial talar process and sustantaculum tali suggestive of talocalcaneal coalition. Diffuse cortical thickening is also detected in the AP view along the shaft of the 2nd metatarsal (image a).*

treatment for the left leg, revealed that as a child she always had "funny" gait, walking and running with toes out (which was described as a windmill effect). She would also refuse to walk long distances as child. Since the initial chiropractic consultation and left foot casting she was now starting to experience hindfoot pain localized to the region of the posterior calcaneus. Currently the patient is awaiting surgery.

Discussion

Coalitions are usually fibrous or cartilaginous at birth, but many begin to ossify in late childhood.[10] Therefore, the pathogenesis of tarsal coaltions is best viewed as a spectrum of pathology with progressive ossification representing increasing severity of the condition. Many individuals with tarsal coalition remain asymptomatic throughout their life.[4,11] It is suspected that biomechanical stress through physical activity may explain the progression of the spectrum of pathology. With repetitive microfracturing and remodelling the coaltion progressively becomes more osseous, resulting in increasing rigidity and evolving histological changes at the coalition site. This progressive ossification contributes to increasing pain sensitive structures at the coalition site, and also contributes to pain in joints surounding the coaltion due to biomechanical compensations. For example, increasing rigidity at the subtalar joint, results in dysfunctional shock absorption during gait and subsequently the transverse tarsal joint (calcaneocuboid and talonavicular joints) is forced to help dissipate these additional forces.[12] The patient in this case indicated that her left leg was her power generating take off foot for basketball. Her left leg was more symptomatic, had higher intensity of dorsal foot pain and more stiffness, and also demonstrated greater progression of ossification on CT. Future studies might consider investigating the role of leg dominance in the progression of tarsal coalition in athletes.

Patients with tarsal coalition most commonly present with hindfoot pain and stiffness.[6,13] The pain is generally reported in the region of the subtalar or talonavicular joint.[10] The patient in this case first presented with bilateral dorsal foot pain that was worse after activity. The

pain was located in the region of the talus, but not specifically confined to the talonavicular joint. She also noted occasional sharp pain posterior to the medial malleolus that was recreated with passive inversion in the physical examination. She eventually started experiencing progressing hindfoot pain two years after the onset of dorsal foot pain on the more symptomatic left foot. The vague symptomatolgy in this case may have contributed the confusion and delay in an accurate diagnosis.

Limited hindfoot mobility, specifically subtalar inversion and eversion, is the most common physical examination finding in patients with talocalcaneal coalition.[13] The patient in this case reported subjective stiffness confirmed with observable limitation with these movements. Subtalar motion palpation also demonstrated limited, although not completely absent, mobility.

Patients may also present with a rigid planovalgus foot, also called peroneal spastic flatfoot, however this occurs in a minority of patients and not the majority as often cited in early publications on tarsal coalition.[3] Our patient did present with moderate bilateral pes planus and moderate rear foot valgus. A large case series by Takakura et al demonstrated that a bony prominence inferior and posterior to the medial malleolus may be palpated in cases of talocalcaneal coalition. These patients may also exhibit symptoms of tarsal tunnel syndrome, such as sensory disturbance in the sole of the foot and a positive tinel's test.[13] A bony prominence was not palpated in this case and the patient did not display symptoms of tarsal tunnel, however the sharp pain with inversion posterior to the medial malleolus may represent pain at the site of medial subtalar coalition. Our case emphasizes the importance of assessing the mobility of the subtalar joint in the physical examination.

The majority of symptomatic talocalcaneal coalition patients present clinically between 12 and 16 years of age.[3,6] Our patient first reported activity related bilateral knee pain at 12 years of age and a year later reported dorsal bilateral foot pain. By 15 years of age the intensity of dorsal foot pain forced her to minimize her activity level. Around this time she also started to note the progression of hindfoot pain.

Patients with tarsal coalition often present clinically after traumatic injury.[3,14] Ankle sprains have been reported by many authors as a common presenting complaint.[14,15] Mubarek et al also added fractures of the fifth metatarsal head and distal tibia as associated injuries in their series of 69 patients with tarsal coalition.[14] The patient in this case did not have a history of an acute traumatic injury but did have a recent history of left forefoot symptoms. Radiographs of the left foot demonstrated periosteal thickening in the second metatarsal suggestive of chronic mechanical stress. This radiographic finding has not been previously reported, to the authors' knowledge, in cases of tarsal coalition in athletes. The radiographic evidence of chronic bony adaptation is suspected by the authors to be related to the progressive biomechanical compensations secondary to the ossifying coalition. Studies that have measured plantar pressure in patients with tarsal coalition have noted excessive medial mid foot peak contact pressures with diminished peak contact pressures in the lateral forefoot and lateral rear foot compared to normal.[16,17] Excessive loading of the medial side of the mid foot and inadequate load distribution of the forefoot may have contributed to chronic stresses of the second metatarsal in this patient.

X-rays of the foot should be the first imaging modality selected for patients with suspected tarsal coalition.[10] Crim et al concluded that routine anterior-posterior and lateral x-rays are a sensitive screening test for both talocalcaneal and calcaneonavicular coalitions.[18] The review by Newman and Newberg and the study by Crim et al provide an excellent evaluation of the key radiographic signs practitioners should be looking for in cases of suspected tarsal coalition.[1,18] Crim et al determined that the most accurate radiographic signs for diagnosing talocalcaneal coalition are the C-sign and talar beaking visualized on lateral views of the foot.[18] Absent visibility of the subtalar facets (usually middle subtalar facet) and a dysmorphic sustentaculum tali are also helpful in diagnosing talocalcaneal coalition on a lateral x-ray of the foot, however the accuracy of these signs can be confounded by the direction of the x-ray beam.[18] The 15 year old patient in this case demonstrated a positive C-sign and absence of the middle subtalar facet on the lateral radiograph. This radiographic series was initially read as normal by the hospital radiologist, and highlights the importance of practitioners reading their patients films with awareness of these signs.

Axial and coronal CT images are considered the current imaging gold standard for diagnosing tarsal coalition. Many authors have speculated that MRI is more accurate at visualizing non-osseous coalitions. However, in a

blinded prospective investigation Emery et al concluded although MRI has a high rate of agreement with CT, it is not superior in detecting non-osseous coalitions.[19] CT findings for complete tarsal coalition are the presence of an osseous bar between tarsal bones. The more common incomplete coalitions are demonstrated by articular narrowing with subchondral sclerosis or cystic changes[1,19], all of which were apparent in the middle subtalar facet of the case. Although these CT findings were all discussed in the initial report, the radiologist's interpretation of these findings resulted in delayed diagnosis.

Many authors believe symptom presentation correlates with the degree of progressive ossification.[3,8, 14] Our case supports this contention as more advanced CT findings were evident on the symptomatic foot. Clinical investigations quantifying imaging findings and degree of pain and disability would be helpful to determine this degree of correlation.

There is a lack of evidence for outcomes in conservatively managed cases of tarsal coalition. Expert opinion suggests that conservative measures may be adequate in less progressed cases of coalition, where symptoms are tolerable and non-disabling.[3,13,14] Conservative treatment options that have been proposed for symptom management are modifying activity level, cold compresses, NSAIDs and therapeutic modalities or a trial of care with supportive devices such as hard soled shoes, foot orthoses, ankle stabilizing orthoses or a short-leg cast.[3,13] The patient in this case successfully managed her initial dorsal foot symptoms with custom orthotics, but over time the orthotics aggravated her symptoms. Rigid boot casting was unsuccessful for symptom resolution in this case.

To our knowledge, there has been no discussion in the literature regarding the value or contraindications of specific manual therapies in cases of tarsal coalition. Limited hind foot mobility, commonly associated with tarsal coalition, may influence manual therapists to consider joint manipulation or other mobility enhancing techniques. It is our opinion that extremity manipulation of a joint directly affected by the coalition should be contraindicated. Although the biomechanical forces subjected to an extremity joint during a manual manipulation are unlikely to structurally damage a coalition, the intent of the manipulation should be questioned considering the joint is pathologically, and not functionally restricted. Manual therapy in joints surrounding the coalition may be of benefit considering the increased compensatory biomechanical stresses encountered. Exercise based treatments, such as those utilized in this case prior to accurate diagnosis, should also be contraindicated in symptomatic tarsal coalition as this may contribute to the progressive ossification of the coalition.

Surgical resection of the coalition has received the most attention in the literature, with orthopedic case series' reporting successful short term (mean post-surgical follow up of 1 to 5 years) outcomes ranging from 50–94% of cases.[14,20,21,22] Positive prognostic factors for surgical resection of a coalition include minimal joint space involvement (less than 50%), higher fibrous or cartilaginous content, non-skeletally mature patients, lack of secondary osteoarthritis changes and minimal rear foot valgus (less than 16 degrees).[20]

For young athletes who meet the criteria for positive surgical outcomes and wishing to continue to compete at a high level, surgical resection should be strongly considered. Patients who are candidates for surgical resection, but do not elect surgery are less likely to return to desired activity levels.[23,24] Mean duration to return to activity is approximately 10 weeks in those who elect surgical resection.[23]

The long term benefits and repercussions of surgical resection are relatively unknown. The longest available follow-up period post resection surgery in any case series is 10 years.[25] Those authors reported satisfactory results in eight patients with no deterioration of symptom relief, range of motion or progressive degenerative joint changes. Larger, prospective analyses are required to determine the long term effect of surgical resection.

Conclusion

Tarsal coalition is an often overlooked cause of pain in adolescents that present with foot pain. We report a diagnostic case of a competitive 15 year old basketball player who was eventually diagnosed with talocalcaneal coalition. Although tarsal coalition is an imaging based diagnosis that often results in orthopedic management, our case highlights the necessity for all primary care practitioners responsible for diagnosing and managing foot pain to be cognizant of the key diagnostic features of talocalcaneal coalition. In adolescent athletes, delayed diagnosis and inappropriate management may lead to decreased chance of return to competitive activity.

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