EC ORTHOPAEDICS
Case Report

Vitamin D in Refractory Accessory Navicular Syndrome - A Case Report

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Abstract

Accessory navicular syndrome refers to a condition when the accessory navicular bone and/or the attachment of the posterior tibialis tendon are irritated and inflamed. Treatment generally consists of non-steroidal anti-inflammatory drugs, physiotherapy, custom foot orthoses, shoe modification and steroid injection. Surgery is only indicated when all the conservative modalities fail to provide adequate symptomatic relief.

A case of accessory navicular syndrome which was refractory to conservative treatment but responded to vitamin D supplementation was reported. Prescription foot orthoses and repeated focused shockwave treatments did not resolve the pain in over one year. In view of her being a vegan for over 40 years, she was checked for vitamin D status. Results showed that she was moderately deficient in 25-hydroxyvitamin-D. Supplementation of 2,000 IU of vitamin D3 daily resulted in a reduction of VAS pain score from 4 to 0.5 in two weeks. The deficiency of vitamin D should be considered in patients with refractory accessory navicular syndrome.

Keywords: *Accessory Navicular; Vitamin D; Foot Pronation*

Introduction

Accessory navicular syndrome (ANS) refers to a condition when the accessory navicular bone and/or the attachment of the posterior tibialis tendon on the bone are irritated and inflamed.

It is occasionally a source of pain and local tenderness [1,2].

The accessory navicular (AN) is an accessory ossicle anatomically located on the medial side of the foot, proximal to the navicular and is continuous with the tibialis posterior tendon. It affects 4 - 20% of the population [3-5] and affects both feet in 50 - 90% of the cases [1]. Kalboureh., *et al.* (2017) reviewed the foot radiographs of 1240 patients with chronic foot pain and reported that the incidence of AN varies with the three subtypes (Figure 1), which include:

a. **Type 1:** Type 1 AN is basically a small sesamoid bone (≈ 3 mm) of the posterior tibialis tendon. The ossicle lies proximal to navicular bone and is 5 - 7 mm distant to the navicular tuberosity. The incidence of this type of AN varies with studies, varying from 25.4% to 55.8% [5,6]. It is almost equally divided between males and females [5].

b. **Type 2:** The accessory bone is larger in size and can be up to 1.2 cm in diameter. It is separated from the navicular bone by a radiolucent zone which generally measures 1 - 3 mm [7]. The zone possibly represents the area of synchondrosis [8]. Based on the location of the AN in relation to the navicular bone, two subtypes have been reported [9]. In the 2a subtype, the accessory bone is located superior to the navicular bone. In the 2b subtype, the accessory bone is more inferiorly placed. The 2a subtype of AN is more prone to disruption by chronic tensile forces and avulsion stress, whereas the 2b subtype AN is more prone to shearing stress [5]. The reported incidence of type 2 AN in chronic foot pain patients was 42.4%, with 20% of the patients having 2a and 22.4% having 2b subtypes of AN [5]. In both subtypes, the incidence in females was higher than that of the males [5].

c. **Type 3:** Type 3 AN fuses with the navicular, resulting in a large cornuate, gorilliform or horn shaped navicular bone [10]. The incidence of this type of AN was reported to be 32% of all the AN. This type of AN is more commonly seen in males [5].

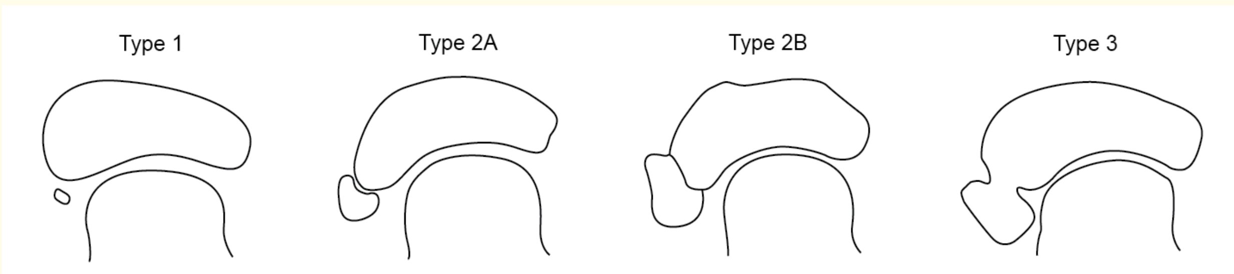

Figure 1: *The three subtypes of accessory bones. (a) Type 1 appears as a small ossicle located behind the navicular bone; (b) The accessory navicular bone of Type 2 is larger and is separated from the navicular bone by a radiolucent zone measuring 1-3 mm. In the 2a subtype, the accessory bone is located more superiorly in relation to the navicular bone. In the 2b subtype, the accessory bone is more inferiorly placed. (c) In type 3, the accessory bone fuses with the navicular bone, to form a cornuate shaped navicular bone.*

The presence of AN may alter the biomechanics of the longitudinal plantar arch. A substantial amount of posterior tibialis tendon fibres insert on the accessory navicular instead of on the tarsal and metatarsal bones. The reduction of the moment arm places undue stress on the posterior tibialis tendon [6,11]. The presence of type 2 or type 3 AN has been regarded as a risk factor for posterior tibialis tendinopathy and possible posterior tibialis tendon tear [12].

Majority of people with AN do not have symptoms. Symptoms are only reported in 1% [13] to 10% of the cases [5]. Symptoms generally follow irritation from shoes and/or follow minor or mild trauma, yet the mechanisms causing the production of symptoms have not been clearly elucidated. The reduction in leverage of the posterior tibialis tendon around the medial malleolus as result of increased proximity of the insertion has been postulated to be one of the mechanisms [10,14]. Other theories include irritation of the navicular prominence and its overlying bursa [1,15]. The latter postulate is supported by a recent study which showed no association between the degree of flat feet and the development and severity of symptoms in patients with AN [16].

Symptoms when present consist of pain and swelling on the AN over the insertion of the posterior tibialis tendon [1,2]. They are aggravated by shoe pressure on the accessory bone, weight bearing, walking or exercise [2]. Treatment is generally by non-steroidal anti-inflammatory drugs (NSAID), physiotherapy, custom foot orthoses and shoe modification. When the above treatments fail to provide adequate symptomatic relief, steroid injection may be indicated. The response of non-athletes to the conservative treatment was reported to be better than that of athletes. Thirty four percent of the non-athletes responded to the treatment as opposed to only 6.9% of the athletes [17]. When all the conservative treatment fails to provide relief, surgical treatment can be considered [1,10,18]. Surgery generally involves fusion of the accessory navicular to the navicular bone, or excision of the former with re-attachment of the posterior tibialis tendon [1,10,18].

We report a case of type 1 AN presented with pain which was refractory to conservative treatment, but responded to vitamin D supplementation.

Case Presentation

A lady aged 45 complained of medial foot pain located over the right navicular bone. The onset of pain was insidious and the patient could not recall any history of trauma or injuries. She also denied any irritation of the bony prominence by footwear. The foot pain was worse with walking and standing. Sitting and lying relieved the pain.

Physical examination revealed prominence of bilateral navicular bones. Knee valgum was more prominent in the right. Both forefeet were in varus (Table 1), with marked heel valgus on weight bearing (Figure 2). Tenderness was elicited by pressure on the right navicular bone, but not on the left.

	Parameters	Left foot (°)	Right foot (°)
Weight bearing	RCSP	-11	-13
	NCSP	0	-4
Non-Weight bearing	Forefoot varus	+12	+20
	First ray dorsiflexion (D)/plantar flexion (P)	D > P	D > P

Table 1: *Measurement of the foot angles in non-weight bearing and weight bearing conditions. RCSP refers for resting calcaneal stance phase; NCSP refers to neutral calcaneal stance phase. Forefoot varus refers to the extent of forefoot varus in relation to the hind foot. + refers to varus, whereas - refers to valgus.*

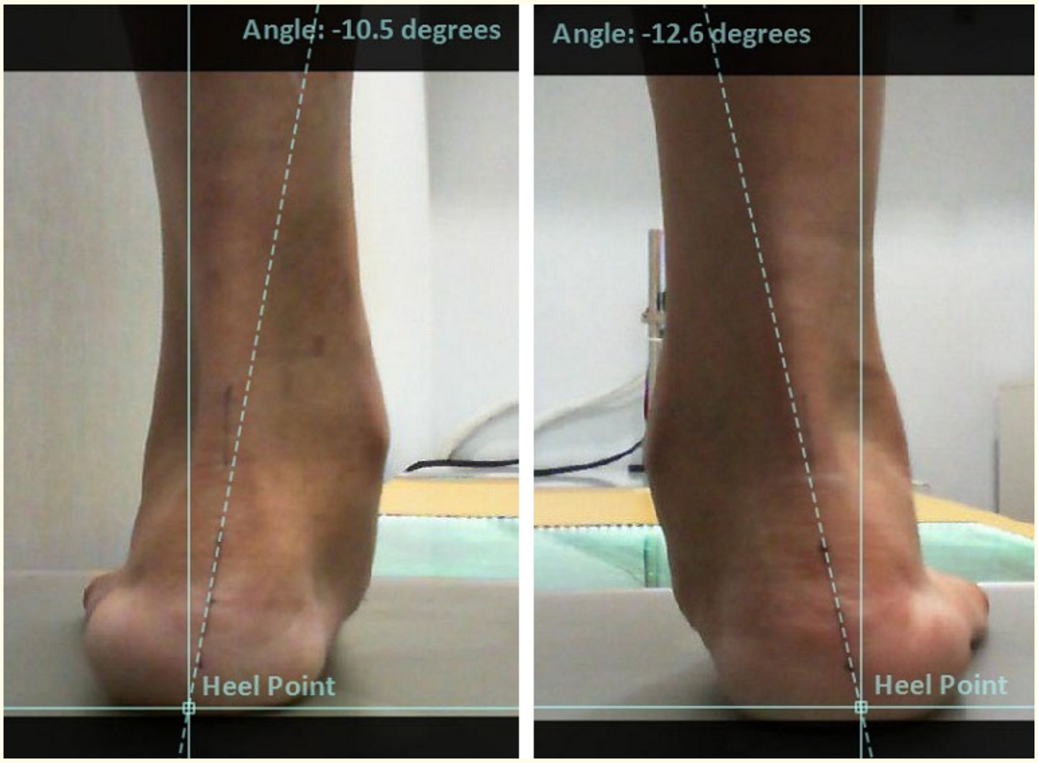

Figure 2: *The posterior view of both heels in weight bearing positions. It can be seen that both heels are in marked valgus.*

Radiograph of the right foot showed a cornuate navicular with a small ossicle situated behind the cornu, which corresponded to a type 1 accessory navicular bone (Figure 3). No serological or other imaging studies were performed as the patient had had a general body checkup which showed nothing of significance four months prior. Also, the symptoms and signs were indicative of a diagnosis of ANS.

Figure 3: *Postero-anterior view of the right foot shows the presence of a small ossicle behind the navicular bone, suggesting that the patient has a type 1 accessory navicular.*

She was treated by extracorporeal shockwave therapy (ESWT) and was referred for a pair of custom foot orthoses and custom sandals to address the abnormal foot biomechanics. Also, she was instructed to perform exercises to strengthen the external hip rotators and the posterior tibialis muscle. The custom foot orthoses were made of 3 mm polypropylene, with a high heel cup of 18 mm, high medial flanges and medial heel posting. The orthoses were topped by a 2 mm Airmed and 2 mm poron. They were made from neutral cast in non-weight bearing position with Kirby skive. The custom sandals were made according to the same prescription with EVA. The patient was advised to wear the foot orthoses at all times when standing and walking.

At the same time, she was treated by focused shockwave. After five shockwave treatments in two months, the pain reduced from a VAS of 7 to 5. The intensity of pain, however, did not reduce despite an additional five shockwave treatments, which were given monthly. Low intensity pulsed ultrasound (LIPUS) was additionally applied onto the right navicular bone 9 months after the onset of the symptoms. This also did not significantly impact the intensity of the pain; four monthly treatments only minimally reduced the intensity of the pain from a VAS of 5 to 4.5. At the time, the patient started to complain of right humeral epicondylitis.

In view of the fact that the patient has been a vegan since childhood, the possibility that she has a low serum vitamin D level is high. Baig., *et al*. (2013) in a study in Pakistan showed that vegetarians regardless of whether they lived in urban or rural areas have low serum vitamin D level [19]. As vitamin D has been found to influence tendon-to-bone healing [20], she was referred for serum 25 hydroxyvitamin-D measurement. Results showed that the patient was moderately deficient in vitamin D level. Her serum level of 25-hydroxyvitamin-D was 23.4 nmol/L, when the normal range was 50 - 200 nmol/L.

In spite of her being a vegan, she was advised to supplement her diet with 2,000 IU of vitamin D3 daily. In two weeks, the pain in the right navicular bone reduced from VAS of 4 to 0.5 and weight bearing no longer aggravated the pain.

Discussion

The case presentation suggested that the patient was suffering from ANS. The custom foot orthoses and ESWT provided partial relief. Custom foot orthoses were prescribed to address the marked forefoot varus and the concomitant mid-foot pronation, to relieve the tensile stress exerted by the posterior tibialis tendon on the navicular bone, as patients with AN have significantly greater extent of abduction and pronation of the forefoot and midfoot than the control patients [16]. Also, she was advised to wear the custom sandals to reduce possible irritation to the navicular bone by footwear, as feet with AN have wider navicular bones than those without an AN [21].

ESWT was applied on the insertion of the posterior tibialis tendon on the navicular bone, as many studies have shown that ESWT is effective in the management of tendinopathies involving the upper and lower extremities [22,23]. Dedes., *et al.* (2018) reported that ESWT reduced pain, improved functionality and quality of life in patients with tendinopathies by at least two points on the five-point Likert scale both post-treatment and at 4-week follow up [23]. The number of treatment sessions was mostly three and sometimes four [23]. It has, however, to be noted that the authors are not aware of any studies targeting ESWT specifically on ANS. In retrospect, the ESWT treatments should be stopped after the first five sessions, as subsequent therapy sessions did not improve the outcome.

Of interest is the prompt resolution of symptoms after the supplementation of vitamin D3. The patient is ectomorphic with a BMI of 18.7 kg/m^2. She has been a vegan since childhood, when she was adopted by a Buddhist nun. A few studies have shown that vitamin D deficiency reduces the tendon to bone healing [24-27]. Twenty eight male Sprague-Dawley rats were fed a vitamin D deficient diet and were restricted on ultraviolet exposure [24]. The right supraspinatus tendon of the rats was then sectioned from the greater tuberosity of the humerus. The lesions were then immediately sutured [24]. The quality of the healed tendons was compared to that of the control rats which were vitamin D sufficient. Histological analysis of the tendon showed that in the vitamin D deficient group, there was less bone formation and collagen fibre organization at four weeks when compared with controls, suggesting that vitamin D deficiency is associated with reduced tendon to bone healing [24]. Similarly, Rai., *et al.* (2016) investigated the effects of vitamin D on inflammation, fatty infiltration and cartilage loss in the knees of hyperlipidemic microswines [25]. They fed 13 microswines with high cholesterol diet and divided them into three groups, viz. the vitamin-D deficient group, the vitamin D-sufficient group and the vitamin D-supplementation group. At the end of one year, they found that the microswines in both the vitamin D-deficient and vitamin D-sufficient groups had increased inflammation in knee joint tissues, fatty infiltration in quadriceps, patellar tendon and collateral ligaments [25]. Histological studies showed that the inflammation of the vitamin D deficiency group was greatest, followed by the vitamin D sufficient group and the vitamin D supplementation group [25]. This was possibly related to the differences in the level of vitamin D in the three groups which were 8.1 ng/ml, 24.91ng/ml and 52,6 ng/ml respectively [25]. The vitamin D supplementation group had the highest serum vitamin D level [25]. The relationship between the degree of inflammation and vitamin D level has not been clearly elucidated. It is possible that the level of vitamin D in the vitamin D sufficiency group fell short of the optimal vitamin D level.

Vitamin D deficiency possibly influences inflammation through its effects on the matrix metalloproteinases (MPs) levels. MPs are calcium dependent zinc-containing endopeptidases capable of degrading all kinds of extracellular matrix proteins. Up-regulation of MMP-1 and MMP-9 is highly correlated with failed healing of rotator cuff after a repair [27]. Vitamin D is an important regulator of MMP-9, which varies inversely with the inflammatory factors [26,27]. Also, it down regulates the cellular response to tumour necrosis factor α (TNF-α) [28]. Patients who had sufficient levels of serum vitamin D generally have lower levels of interleukin-6 (IL-6) and C-reactive protein (CRP), both of which are indicators of inflammation.

It is possible that the reduction in tendon to bone healing is related to the increased circulating matrix metalloproteinases (MMP)-2 and MMP-9 in vitamin D deficiency [26]. Vitamin D increases bone mineral density [29], which correlates to a greater degree of healing in patients [30]. Also, it positively correlates with muscle growth and metabolism [31]. More importantly, it mediates osteoclastogenesis and regulates coordinated bone remodeling [32], which is important for bone-to-tendon healing as ingrowth of bone forming cells is required for secure healing [33]. This is supported by the findings that vitamin D positively influences rotator cuff tendons healing through its effects on MMPs [34].

We speculate that the vitamin D supplementation in the present case reduces the inflammation and improves the tendon to bone healing, with resultant resolution of symptoms. Thus vitamin D supplementation should be considered in vitamin D deficient patients with ANS, particularly when the symptoms are refractory to conservative treatments.

The case presentation has some weaknesses. Firstly, we did not take any MRI of the foot to re-confirm the diagnosis nor did we record the power and frequency of the ESWT used. Also, we did not measure the post-intervention vitamin D level to ensure that the serum level of vitamin D was within normal range.

Conclusion

In addition to standard conservative treatment, vitamin D supplementation should be considered in patients with ANS who are deficient in vitamin D before referral for surgical intervention.

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Volume 10 Issue 2 February 2019