Abstract—Bipartite medial cuneiforms are relatively rare but may play a significant role in biomechanical and gait abnormalities. It is believed that a bipartite medial cuneiform may alter the available range of motion due to its larger morphological variant, thus limiting the metatarsal plantarflexion needed to achieve adequate hallux dorsiflexion for normal gait. Radiographic and clinical assessment were performed on two patients who reported with foot pain along the first ray. Both patients had visible bipartite medial cuneiforms on MRI. Using gait plate and Metascan™ analysis, both were noted to have four measurements far beyond the expected range. Medial and lateral heel peak pressure, hallux peak pressure, and 1st metatarsal pressure were all noted to be increased. These measurements are believed to be increased due to the hindrance placed on the available ROM of the first ray by the increased size of the medial cuneiform. A larger patient population would be needed to fully understand this developmental anomaly.

Keywords—Bipartite medial cuneiforms, cuneiform, developmental anomaly, gait abnormality.

I. INTRODUCTION

ACCESSORY and bipartite bones of the foot are common developmental anomalies. By definition, bipartition means the division of one whole element into two separately-formed parts. The most common frequently observed example of a bipartite condition amongst tarsal bones is the medial cuneiform. It is a rare tarsal developmental variant at the Lisfranc joint. The first case of bilateral bipartite medial cuneiform in a cadaver foot was in the 18th century [12]. Barlow cited a 1757 report by Morel describing the first case of a bilateral bipartite medial cuneiforms which he termed os cuneiform I bipartum [12].

In 1942 Barlow gave the classic anatomical description and quoted an incidence of one in 320 [12]; Gruber in 1877 reported an incidence of 0.33% [22], Trolle reported 2.4% in 1948 [25], and Burnette 0.27% in 2001 [15]. A more recent study from 2014 Chang et al. [5] sought to determine the prevalence with a retrospective review of 1,000 consecutive MRI. Using gait plate and Metascan™ analysis, both were noted to have four measurements far beyond the expected range. Medial and lateral heel peak pressure, hallux peak pressure, and 1st metatarsal pressure were all noted to be increased. These measurements are believed to be increased due to the hindrance placed on the available ROM of the first ray by the increased size of the medial cuneiform. A larger patient population would be needed to fully understand this developmental anomaly.

A reported meta-analysis of described case series and indicated that a genetic component [22] likely exists with a male predominance of 84.1% [15]. Anatomists, including Barlow, Gruber, Jones, Smith, and Trolle have described this rare phenomenon in cadaver specimens, with the first symptomatic case reported by Barclay in 1932 [12], [14], [16], [25].

Bipartition is hypothesized to occur when there is a failure of coalescence of two primary centers of ossification. They are generally bilateral and have smooth cortical borders, whilst fractures are generally unilateral with irregularities of the cortical borders. Bilateral occurrence has been shown to be > 60% [19].

Ossification begins laterally with the cuneiform during the first year of life, followed by the medial in the second year, and the middle cuneiform in the third year. The normal medial cuneiform is thought to develop from a single ossification center. When there are two ossification centers that fail to fuse, it results in the bipartite medial cuneiform.

The single ossification center originates from a single mesenchymal primordium that ultimately develops into a single complete bone. The non-chondrified tissue constitutes a closely-packed mesenchymal-looking, cell-forming interzone that eventually disappears and is replaced by other proliferating cells giving rise to a single complete bone. Formation of a divided mesenchymal primordium could result from altered cell-intrinsic patterning at the gene level and/or variation in activator/inhibitor positional signaling mediators responsible for local control of the mesenchymal primordium.

Three possible morphological categories are described in 2010: complete bipartition, incomplete bipartition, and division of the distal articular surface with the former being the most commonly observed [6]. Bipartition of bones does not always necessarily equate to two separate equal parts.

The developmental variant is generally segmented horizontally; dividing the bone into an upper and lower half. The plantar ossicle is typically larger than the dorsal ossicle. Pfitzner in 1896 termed the two halves “os cuneiform I dorsal” and “os cuneiform I plantare” respectively [24]. A pseudo articulation between the segments forms and is bridged with a cartilaginous synchondrosis, fibrous syndesmosis, or combination thereof. Chang et al. in 2014 studied the anatomy of this developmental variant [5]. They found that the tibialis anterior tendon attaches to the proximal superomedial aspect of the dorsal segment with the posterior tibialis tendon attaching to the distal inferolateral portion of the plantar segment. The peroneal longus tendon attaches to the proximal interomedial and distal inferolateral portions of the plantar segment. The Lisfranc ligament proper (interosseous portion)
and the dorsal Lisfranc ligament extend from the dorsal segment of the bipartite medial cuneiform to insert into the base of the second metatarsal. The plantar Lisfranc ligament extends from the plantar segment of the bipartite medial cuneiform.

Chiodo et al. state the 30-degree external oblique radiograph as being the best for visualization of the segments. Because the two ossicles overlap, it can be difficult to diagnose on plain radiographs [3]. Chang et al., after reviewing 1,000 MR studies, describe the “E-sign” which can be seen on the cross-sectional imaging on MRI or CT which they state is the definitive in diagnosing the anomaly and can also reveal regional anatomy [5]. The “E-sign” is formed by the cleft or joint space between the two segments as seen in the horizontal plane in a sagittal plane section [23]. The partition should demonstrate smooth well corticated margins. It is uncommon to see this type of orientation for a fracture.

The summative mass of these two ossicles is slightly larger than the single normal medial cuneiform. Jashashvili et al. concluded that the “bipartite condition in the medial cuneiform represents developmental variation that does not cause significant overall morphological differences”; however, they do not comment on the biomechanical differences that arise from the structural changes [6].

Fulwadhva and Parker alluded that a relatively minor traumatic event could cause significant long-term symptoms due to altered biomechanics within the midfoot [10]. Chang et al. agree that “it can be a potential source for both nontraumatic and traumatic midfoot pain” [5], [11], [13].

Most patients present once symptomatic, with symptoms typically described after a recallable injury or high impact injury/activity [9], [4]. Physical examination reveals pain with range of motion of the first ray and localized pain to the medial cuneiform when pressure is applied [9]. Panu et al. describe symptomatic patients as those who typically present with chronic midfoot pain exacerbated with ambulation or acute injury and assert it is the inherent instability of the pseudoarticulation resulting in a stress response and/or degeneration that result in pain [7]. Steen et al. also believed the disruption of the fibrocartilaginous articulation results in the chronic pain [9].

Numerous treatment modalities for symptomatic patients have been described in the literature. Bismil in 2001 performed a CT-guided corticosteroid injection [2]. Azurza in 2001 preformed an arthrodesis of the segments with a transcortical screw [1]. Chiodo in 2002 resected the dorsal segment [3]. Surgical indications reserved for when conservative measures fail and consist of either excision of the smaller ossicle when the smaller dorsal segment comprises 30% or less of the combined volume or arthrodesis of the segments [7], [1]. All authors portray successful treatment results with immobilization, injection, surgical therapy or a combination thereof.

Root in 1977 described the function of the metatarsophalangeal joint as a glinglymoarthrodial-type joint and as a rule stated that 65 degrees to 75 degrees of sagittal plane dorsiflexion is required for normal gait [19].

Plantarflexing and gliding of the first metatarsal in a plantar proximal fashion relative to the base of the proximal phalanx allows the transverse axis of the first metatarsophalangeal joint to shift more dorsally and proximally. This permits articulation of the first metatarsal with the base of the proximal phalanx; Buell in 1988 stated that it is this transverse axis movement that allows a large range of motion at the metatarsophalangeal joint during propulsion [26].

The axis of motion of the first ray was described by Hicks in 1953 and was identified to be proximal medial to distal lateral. As the calcaneal inclination angle decreased, the first ray would then dorsiflex on the rearfoot [18].

Normal dorsiflexion of the first metatarsophalangeal joint is dependent on the capability of the first ray to plantarflex. Root et al. noted the range of motion available at the first metatarsophalangeal joint declines as the first ray dorsiflexes; similarly, the range of motion available at the first metatarsophalangeal joint increases with first ray plantarflexion [19].

Little rotary motion exists at the first metatarsocuneiform joint according to Ebisu in 1968, with flexion and extension as the primary motion [17]. “At the end range of motion in the first metatarsocuneiform joint, the joint appeared to have a locking mechanism. When the first metatarsocuneiform joint was locked, and increasing extensory force was applied, rotation occurred at the medial cuneonavicular joint, with the medial cuneiform rotating medially away from the intermediate cuneiform” [8]. The actual range of motion in plantarflexion and dorsiflexion for the medial cuneonavicular joint was determined in 1989 by Ouzounian and Shereff and was found to be between 0.7 and 8.7 degrees with a mean of 3.5 degrees and between 3.5 and 9.9 degrees with a mean of 7.3 degrees in supination-pronation [20]. They also reported between 1.9 degrees and 5.3 degrees, with a mean of 3.5% for the plantarflexion-dorsiflexion range of motion at the first metatarsocuneiform articulation and 0.0 degrees to 2.6 degrees, with a mean of 1.5 degrees in supination-pronation [8].

The first tarsometatarsal joint was further explored in 1989 by Wanivenhaus and Pretterklieber who determined several ranges of motion: abduction of 4.4 degrees, adduction of 5.0 degrees, inversion of 4.1 degrees after dorsal displacement, eversion of 6.2 degrees after dorsal displacement, and a dorsal displacement of 2.6mm [21].

Additionally, in 1996 Phillips et al. revealed that the first cuneiform rotated 12 degrees to 28 degrees further forward than the navicular and the first cuneiform rotated forward 11 degrees to 26 degrees more than the calcaneus; thus there was significantly more forward rotation distal to the cuneonavicular joint than proximal to the cuneonavicular joint [8]. The implication of this finding suggests “the only way that these rotations could happen is for a significant plantarflexion motion of the first cuneiform relative to the navicular and almost no net plantarflexion motion of the navicular relative to the rearfoot or of the metatarsal relative to the cuneiform... almost all of the plantarflexion motion of the first ray occurs at the cuneonavicular joint and failure of the
metatarsophalangeal joint to dorsiflex adequately can produce interphalangeal joint hyperextension which may contribute a role in the heel lifting off the ground during propulsion”[8].

It is our belief that the bipartite medial cuneiform alters the available range of motion of the first ray due to its slightly larger morphological variant, limiting the amount of eventual plantarflexion needed to achieve adequate dorsiflexion for normal gait to occur. The purpose of this investigation was to see if there was indeed a relationship that could be demonstrated upon gait analysis in individuals with a bipartite medial cuneiform.

II. CASE REPORTS

Two patients were discovered to have bipartite medial cuneiforms within the same time frame. Both patients were symptomatic with pain to the corresponding foot. Neither case was noted to have bilateral morphologic variants. Both complained of hallux limitus like symptoms and both had decreased range of motion of the first metatarsophalangeal joint with weight bearing.

A. Case One

A 65 year old female presented to a Virginia-based Foot and Ankle Clinic with the chief complaint of right foot pain of numerous years duration. Plain radiographs (Figs. 4 (a)-(c)) demonstrated the anomaly, followed by Magnetic Resonance Imaging (MRI) which revealed an incomplete separation proximally of the two segments with a pseudo articulation between the two visualized in the sagittal plane (Figs. 3 (a)-(c)).

B. Case Two

A 65 year old female presented to a Virginia-based Foot and Ankle Clinic with the chief complaint of right foot pain of numerous years duration. Plain radiographs (Figs. 4 (a)-(c)) revealed the pedal variant. MRI demonstrated the area with a synchondrosis between the segments (Figs. 6 (a)-(c)), without bone marrow edema and mild degenerative change between the two ossicles. Also noted was moderate first metatarsophalangeal joint degenerative change of the first metatarsal head and adjacent medial cuneiform.

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<th>Table I</th>
<th>SUBJECTIVE PAIN SCORES</th>
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<tr>
<td>Visual analog score (VAS)</td>
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<td>4 out of 5</td>
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<td>AOFAS Ankle-Hindfoot Scale</td>
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<td>AOFAS Hallux Metatarsophalangeal-Interphalangeal Scale</td>
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III. MATERIALS AND METHODS

The clinical and imaging findings were reviewed. This led to journal article review and an observational study that was approved by the Institutional Review Board.

Subjective pain was measured with various scales including: American Orthopedic Foot and Ankle Society (AOFAS) pain scales for the foot, visual analog score scales (VAS), and functional pain scale (Table I). Footmaxx™ located in Roanoke VA allowed utilization of their gait plate system and Metascan™ technology to visualize and quantify dynamic, weight-bearing biomechanical assessment while generating a dynamic gait and pressure analysis report with both 2-D and 3-D images (Figs. 2, 5). The system also tracks dynamic plantar foot pressure during contact, midstance, and propulsion via an eight point pressure vs. time graph (Figs. 7 (a), (b)).

IV. DATA AND RESULTS

Data from the metascans were collected. For each patient and each foot, the average distance out of range was calculated for each measurement. Values within the normal range were assigned a zero. Values outside of the normal range were assigned a positive or negative value for above or below the range, respectively. There were four measurements that are further beyond the expected range: medial and lateral heel peak pressures, hallux peak pressure, and 1st metatarsal peak pressure (Fig. 8). The right foot (foot with the anatomical variant) had differences in those four measurements (Fig. 9). Heel peak pressures of the right foot were less than the left. The right foot value was greater than the left for 1st metatarsal peak pressure. Hallux peak pressure showed more pressure in the left foot than the right. Specific to the right foot of both patients was a decrease in the 1st and 5th metatarsal peak time and heel peak time difference. Less time is being spent in the propulsive and contact phases of the gait cycle with an increase in midstance duration. Both midfoot and Lesser metatarsophalangeal-interphalangeal AOFAS scores were low and similar between patients (Table I).

V. DISCUSSION

Although this data set is small and only between two patients, the deviations are still worth noting. The anatomical variant is rare, and a large patient population would be required to state statistical significance. From the data given, one can extrapolate that there are differences in heel strike and the medial column of the foot. The first metatarsal is spending less time but more pressure during gait with less force to the hallux during toe off; and thus dorsiflexion of the hallux and push off capability of the right foot is lessened when compared to the left. The available range of motion of the first ray is limited. The amount of first ray plantarflexion needed to achieve adequate dorsiflexion of the hallux for normal gait must be affected, and is more so in the right foot than the left.

Heel strike, represented by heel pressure and peak time, is also noticeably different. Heel pressure is greater in both feet, and lesser to the right foot than the left. Overall, the right foot is spending less time in the contact phase and more time in midstance phase of gait with most of the gait cycle being spent...
in midstance. This may contribute to the increase in heel pressure observed. This would also go along with decreased push off capability of the hallux. The first metatarsophalangeal joint is not adequately dorsiflexing and thus restricted, leaving the interphalangeal joint to hyperextend to contribute a role in the heel lifting off the ground during propulsion. The inability to achieve 1st metatarsophalangeal joint range of motion contributes to the prolonged time in the midstance phase longer.

To a degree, both feet have similarities in the deviated measurements. It can be postulated that the overall gait was altered bilaterally to accommodate the foot variant.

![Fig. 2 Dynamic gait and pressure analysis of case study #1](image1)

(a) Oblique view
(b) AP view
(c) Lateral view

![Fig. 1 Plain radiographs of case study #1](image2)

(a) Solid arrow points to the slightly larger volume of the combined bipartite medial cuneiform; (b) Solid arrow points to the dorsal overlap of the smaller ossicle; (c) Double arrows point to the bipartition of the medial cuneiform

![Fig. 3 MRI of case study #1](image3)

(a) Sagittal view of the bipartite medial cuneiform; (b) E-sign visible only on the sagittal view marked by the solid red lines; (c) Coronal view of the medial cuneiform bipartition
VI. CONCLUSION

Since the metatarsophalangeal joint failed to dorsiflex adequately, the plantarflexion motion of the first ray must have been altered and since this motion occurs at the cuneonavicular joint the medial cuneiform with the bipartition appears to have limited its plantarflexion capabilities resulting in the decreased motion observed which led to its decreased
propulsion forces for toe off. The bipartite medial cuneiform alters the available range of motion of the first ray due to its slightly larger morphological variant. This limits the amount of eventual plantarflexion needed to achieve adequate dorsiflexion for normal gait to occur. To fully understand this developmental anomaly a larger patient population would have to be identified and analyzed.

Fig. 7 (a) Footmaxx™ Metascan© report: Functional rigidity of the foot is apparent with the first metatarsal peaking early and at a high pressure while contacting the ground for a prolonged duration. A functionally rigid plantarflexed first ray is suggested by the first metatarsal peaking early and at a high pressure.

Fig. 7 (b) Footmaxx™ Metascan© report: A functionally rigid plantarflexed first ray is suggested by the first metatarsal peaking early and at a high pressure. A tight posterior group, a weak anterior group, or a plantarflexed first ray is suggested by a severely reduced contact subphase duration.

Fig. 8 Mean out of range both feet, both patients. Notice heel peak pressures are greatly elevated along with an increase in 1st metatarsal and hallux peak pressures.
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REFERENCES


