

# Prevalence and characteristics of tarsal coalition in East Asians: what are the differences compared to Caucasians?

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## Article

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# Abstract

Previous studies have reported a prevalence of tarsal coalition of 1–13%. Calcaneonavicular coalition is known as main anatomical type, and the bilateral occurrence of tarsal coalition is known to be 50–60%. These are the results of studies on Caucasians, there have been few studies targeting large number of East Asians so far. We hypothesized that the prevalence and characteristics of tarsal coalition in East Asians might differ from those in Caucasians. The medical records of 839 patients who underwent bilateral computed tomography on foot and ankle in our hospital from January 2012 to April 2021 were retrospectively reviewed. The overall prevalence was 6.0%, talocalcaneal coalition was the most common anatomical type. The overall bilateral occurrence was 56.5%, talocalcaneal coalition had the highest bilateral occurrence (76.0%) among anatomical types. Isolated union of the posterior facet was the most common subtype of talocalcaneal coalition (43.2%). Talocalcaneal coalition had a significantly higher proportion of coalition-related symptomatic patients than calcaneonavicular coalition ( $p = 0.019$ ). Our study showed a similar trend to other East Asian studies, confirming the existence of racial differences. The possibility of tarsal coalition in foot and ankle patients in East Asians should always be considered, and bilateral examination is essential for diagnosis.

## Introduction

Tarsal coalition is a state in which two or more different tarsal bones are united, and maybe fibrous, cartilaginous, or osseous<sup>1–3</sup>. Failure of mesenchymal separation has been suggested to be the primary cause of tarsal coalition, which is presumed to be due to an autosomal dominant inheritance pattern with high penetrance<sup>1–5</sup>. Tarsal coalition is associated with diminished motion of affected joints and often leads to pathologic conditions including rigid flat foot, peroneal spasm, and secondary degenerative change on adjacent joint<sup>3,6–10</sup>. Therefore, understanding the prevalence and characteristics of tarsal coalition has clinically important value.

Previous studies have reported a prevalence of tarsal coalition of 1–2%<sup>11–14</sup>. Harris et al. evaluated the prevalence of peroneal spastic flatfoot using physical examination<sup>11</sup>. Other authors assessed tarsal coalition by plain radiography in patients with painful feet or primary foot diseases. Considering that most of the tarsal coalition patients are asymptomatic, and plain radiography is not suitable for diagnosing non-osseous coalitions, the prevalence of tarsal coalitions is expected to be much higher. Recent studies using cadavers or magnetic resonance imaging (MRI) have reported prevalence of 11–13% in Caucasians<sup>15,16</sup>.

The most common anatomical type of tarsal coalition is calcaneonavicular (CN) coalition<sup>5,15,17,18</sup>. Bilateral occurrence have been reported in 50–68% of cases<sup>2,5,17,19–21</sup>, and multiple tarsal coalition are very rare<sup>22–25</sup>. Talocalcaneal (TC) coalition is further subdivided according to the location of union in the subtalar joint, and middle facet is known as the most common coalition site<sup>20,26–29</sup>. But, most of these results were obtained in Caucasian studies. According to some East Asian studies which used computed

tomography (CT) or MRI for diagnosis, TC coalition has a higher proportion than CN coalition, and TC coalition most commonly involves the posterior subtalar facet<sup>30-34</sup>. It appears that the anatomical distributions of tarsal coalition might differ between races. However, East Asian studies on tarsal coalition are still insufficient to support the possibility of racial difference, and there is no study assessing bilaterality in a large sample to the best of our knowledge.

This study was performed using both-side CT in a Korean cohort. We evaluated the prevalence, anatomical distributions, and bilateral occurrence of tarsal coalition. In addition, we assessed the proportion of symptomatic patients associated with tarsal coalition. Racial differences and clinical significance will be discussed based on the results of this study and the trends of previous studies. We hypothesized that the characteristics of tarsal coalition in East Asians might differ from those previously reported in Caucasians.

## Materials And Methods

The medical records of all patients who underwent bilateral CT at our hospital from January 2012 to April 2021 were retrospectively reviewed, and 1028 patients were identified. Patients with abnormal anatomical structures or with the possibility of secondary tarsal coalition were excluded from the study (Table 1). Finally, this study was conducted on 839 patients (492 men and 347 women) of average age 44 years (range, 13–75 years).

Table 1  
Exclusion list

Exclusion criteria	Excluded patients (n = 189)
Fracture around foot and ankle	135
Severe osteoarthritic change on subtalar joint	31
Tumor	10
Infection	10
Congenital anomalies of lower extremity	3

All patients were evaluated for the presence and location of the tarsal coalition using coronal, sagittal, transverse, and three-dimensional reconstruction images of bilateral CTs. CT was used as a main imaging technique for identification of tarsal coalition because it is advantageous in understanding the complex anatomical structure of tarsal bone<sup>35,36</sup>. All CTs were taken in the same facility using the same protocol. Images were obtained using a SOMATOM definition AS + unit (Siemens, Germany) using a bone algorithm. Typical scan parameters were as follows; field of view, 25 cm; peak voltage, 120 kVp; quality reference, 82 mAs; scan time per slice, 1 s; and slice thickness, 2 mm. All radiographic evaluations were performed using a picture archiving and communication system (Maroview1, version 5.4; Marotech, Seoul, Korea) in Digital Imaging and Communicating in Medicine (DICOM) format.

The diagnostic criteria of tarsal coalition used in this study were as follows: the presence of a bone bridge, narrowing of the joint surface, irregular cortical bone surface, subchondral bone sclerosis, and cyst formation<sup>37</sup>. The overall prevalence of tarsal coalitions was evaluated as the proportion of patients who displayed these features on CT images among the patients included in this study. Proportions of tarsal coalition patients by groups according to age and gender were also evaluated and ages were classified using 20-year intervals.

Anatomical types of tarsal coalition were classified according to the united tarsal bone as TC, CN, naviculocuneiform (NC), talonavicular (TN), and calcaneocuboid (CC) coalition. Anatomical subtypes were further classified according to location of tarsal bone union for TC and NC coalitions. For TC coalition, union was assessed at the anterior, middle, and posterior facet, and for NC coalition, union was assessed at the medial, intermediate, and lateral cuneiform. The middle and posterior facets are apposed and difficult to differentiate. The canalis-tarsi is anatomically located between the middle and posterior facets, and therefore, we carefully differentiated union of the middle and posterior facets (Fig. 1)<sup>32</sup>. In addition, non-osseous TC coalitions may present CT findings similar to degenerative osteoarthritis with abnormal joint space narrowing and minimal marginal reactive osseous changes (Fig. 2)<sup>36</sup>. Although we excluded patients with severe subtalar osteoarthritis before the study, it was important to distinguish between non-osseous TC coalition and mild subtalar osteoarthritis. When there was a subtalar lesion showing the above findings on CT in relatively young patients under 40 years old, it was diagnosed as TC coalition because of the low possibility of degenerative changes. In patients over 40 years old, talar beak sign and C sign were additionally assessed by simple radiography to diagnose TC coalition (Fig. 3)<sup>36,38</sup>. Also, a drunken waiter sign was evaluated on the CT coronal view (Fig. 4)<sup>38</sup>.

Bilateral coalition was defined as coalition in both feet of the same anatomical type. Bilateral occurrence was evaluated for each anatomical type, and its prevalence are presented as percentages of bilateral patients among patients of each anatomical type.

All patients were assessed retrospectively for symptoms related to tarsal coalition. Clinical features were evaluated based on the pathologic condition patients complained of at the time of CT scan and were obtained by chart review. Symptoms associated with tarsal coalition were as follows: repeated ankle sprains and pain, pain at the coalition site, adjacent joint pain due to secondary degeneration, sinus tarsi syndrome, peroneal spasm, and tarsal tunnel syndrome<sup>3,39-41</sup>. If there was no other disease causing these symptoms, it was concluded as a symptom caused by tarsal coalition. In addition, pain at the coalition site was judged as a symptom of coalition when there was ill-defined fluid signal intensity in the bone marrow of the adjacent bone. Proportions of symptomatic patients associated with tarsal coalition were identified in main anatomical type of TC and CN coalitions.

We randomly selected 51 CT images of the 51 patients based on the calculation of sample size according to Bonnett's approximation to assess intra- and inter-observer reliabilities for the diagnosis of tarsal coalition<sup>42</sup>. Diagnoses were performed independently without personal information by two

orthopaedic surgeons and repeated two weeks later. Reliabilities for diagnosing tarsal coalition were analyzed using Kappa statistics<sup>43</sup>.

The statistical analysis was performed using IBM SPSS version 23 (IBM Corp., Armonk, NY, USA). The chi-square test was used to compare prevalence of tarsal coalition according to gender and age, and to compare proportions of symptomatic patients associated with tarsal coalition between anatomical types, including CN and TC coalition. Statistical significance was accepted for  $p$ -value < 0.05.

This study was reviewed and approved beforehand by the Institutional Review Board (IRB) of Yeungnam University Hospital, which waived the requirement for informed consent because of the retrospective design of the study. All methods were carried out in accordance with the IRB's guidelines and regulations.

## Results

A total of 839 patients were included in this study. Fifty patients had tarsal coalition, an overall prevalence of 6.0%. Overall prevalence and proportions of tarsal coalition patients by groups according to age and gender are described in Table 2. The proportion of tarsal coalition patients was the highest in the patient group under 20 years old (11%), and the proportion was significantly lower in the older groups ( $p = 0.024$ ) (Table 2).

Table 2

Overall prevalence and proportions of coalition patients by group according to age and gender

Variable	Patients with tarsal coalition (%)	Patients without tarsal coalition (%)	$p$ -value
Total (n = 839)	50 (6)	789 (94)	
Gender			0.492
Male (n = 492)	27 (5.5)	465 (94.5)	
Female (n = 347)	23 (6.6)	324 (93.4)	
Age (yr)			0.024
≤ 20 (n = 91)	10 (11)	81 (89)	
21–40 (n = 157)	14 (8.9)	143 (91.1)	
41–60 (n = 308)	12 (3.9)	296 (96.1)	
≥ 61 (n = 283)	14 (4.9)	269 (95.1)	
Values are presented as number (%).			

Proportions of anatomical types of tarsal coalition are described in Table 3. Isolated TC coalition was the most common anatomical types with a prevalence of 50.0% (Table 3). Bilateral occurrence of tarsal

coalition is described in Table 4. Overall, the bilateral occurrence was 56.5% and TC coalition had the highest bilateral occurrence among anatomical types at 76.0% (Table 4).

Table 3  
Distribution of anatomical type

Subtype	Patients with Tarsal Coalition (n = 50)
Isolated TC coalition	25 (50)
Isolated CN coalition	15 (30)
Isolated NC coalition	6 (12)
CN coalition + TC coalition	2 (4)
TC coalition + TN coalition	1 (2)
TC coalition + CC coalition	1 (2)
Values are presented as number (%).	
TC = talocalcaneal; CN = calcaneonavicular; NC = naviculocuneiform; TN = talonavicular; CC = calcaneocuboid.	

Table 4  
Bilateral occurrence

Type	Bilateral occurrence (%)
Total (n = 46) <sup>a)</sup>	26 (56.5)
TC coalition (n = 25)	19 (76)
CN coalition (n = 15)	4 (26.7)
NC coalition (n = 6)	3 (50)
Values are presented as number (%).	
TC = talocalcaneal; CN = calcaneonavicular; NC = naviculocuneiform.	
<sup>a)</sup> Multiple tarsal coalition patients are excluded from this list.	

Proportions of anatomical subtypes according to union sites of TC and NC coalitions are provided in Table 5. Isolated union of the posterior facet was most common at 43.2% in TC coalitions, and medial cuneiform was most common coalition site in NC coalitions accounting for 77.8% (Table 5). Coalition of multiple cuneiform bones was not observed.

Table 5  
Detailed subtypes of talocalcaneal and naviculocuneiform coalitions

Subtype	No. of tarsal coalition
TC coalition (n = 44) <sup>a)</sup>	
Isolated anterior facet	0
Isolated middle facet	7 (15.9)
Isolated posterior facet	19 (43.2)
Anterior facet + middle facet	0
Middle facet + posterior facet	11 (25)
Anterior facet + posterior facet	0
Anterior facet + middle facet + posterior facet	7 (15.9)
NC coalition (n = 9) <sup>a)</sup>	
Medial cuneiform	7 (77.8)
Intermediate cuneiform	2 (22.2)
Lateral cuneiform	0
Values are presented as number (%).	
TC = talocalcaneal; NC = naviculocuneiform.	
<sup>a)</sup> Bilateral lesions are counted individually.	

The proportion of patients with symptoms related to tarsal coalition in anatomical types including TC and CN coalitions are described in Table 6. TC coalition was significantly more associated with symptoms than CN coalition ( $p = 0.019$ ) (Table 6).

Table 6  
Proportion of patients with symptoms in main anatomical types

Subtype	Patient with coalition symptom	Patient without coalition symptom	p-value
Total (n = 50)	35 (70)	15 (30)	0.019 <sup>a)</sup>
Isolated TC coalition (n = 25)	22 (88)	3 (12)	
Isolated CN coalition (n = 15)	7 (46.7)	8 (53.3)	
Values are presented as number (%).			
TC = talocalcaneal; CN = calcaneonavicular.			
<sup>a)</sup> The Chi-square test was used to compare proportions of symptomatic patients associated with tarsal coalition between CN and TC coalition.			

Intra- and interobserver reliabilities for diagnosing tarsal coalition were 0.938 and 0.916, respectively.

## Discussion

In this study, the overall prevalence of tarsal coalition was 6.0%. As we expected, this result was appreciably higher than the prevalence of 1–2% from previous studies which assessed tarsal coalition using simple radiography<sup>12–14,44</sup>. However, the prevalence was lower than that of recent cadaver and MRI studies<sup>35,45,46</sup>. We believed two factors may have influenced these results. First, considering that the proportion of symptomatic patients among the 50 tarsal coalition patients was high at 70%, asymptomatic patients were not sufficiently included. Second, CT is more vulnerable to the misdiagnosis of non-osseous coalition than MRI. Solomon et al. reported nine non-osseous coalitions were diagnosed among 100 dissected feet, and only 55% of them were correctly diagnosed by prior CT<sup>47</sup>. Guignand et al. found four coalitions (2 cartilaginous and 2 fibrous forms) among 11 CN coalitions diagnosed during surgery were not diagnosed by CT, though diagnosis was possible by MRI<sup>48</sup>. Therefore, the actual prevalence is expected to be higher. We found the proportion of tarsal coalition patients was significantly higher at 11% in the patient group under 20 years of age. Since tarsal coalition mostly causes symptoms in adolescence and early adulthood<sup>14,49</sup>, a relatively large number of symptomatic young patients may have visited the hospital. Therefore, it is expected that the probability of receiving a diagnosis by conducting an examination was relatively higher than that of the older group.

In terms of anatomical types, TC coalition was the most common followed by CN coalition. These two types accounted for most of the total tarsal coalition, which concurs with previous studies<sup>15,17</sup>. In many Caucasian studies, either CN coalition dominated or the prevalence of both types were similar<sup>5,15–18</sup>, whereas in the present study, TC coalition was more common, which is in line with the results of recent East Asian studies<sup>30,31,33,50</sup>. These results indicate the anatomical distribution of tarsal coalition is

racially dependent. In addition, differences between the clinical presentation of TC and CN coalitions may explain reported TC coalition predominance. According to Solomon et al., CN coalition appears to be unrelated to arthritic change, which means that limitations of subtalar joint motion are less likely for CN coalition<sup>47</sup>. Since motion restriction causes symptoms of tarsal coalition and secondary arthritis, TC coalition likely accounts for a higher proportion of symptomatic patients than CN coalition. In fact, in this study, the proportion of symptomatic patients was significantly higher for TC coalition. Due to these differences in clinical presentation, TC coalition patients are more likely to visit the hospital and be diagnosed with tarsal coalition. Furthermore, coalition type may have influenced the results of this CT study. According to Nalaboff et al., TC coalition involves osseous coalition in 33.3% of patients, whereas most of the CN coalition is composed of non-osseous coalition (56% cartilaginous and 44% fibrous union)<sup>16</sup>. In the present study, CT was the only imaging modality used, and thus, non-osseous forms of coalition may have been under diagnosed.

In current study, the bilateral occurrence of tarsal coalition was 56.5% among the 46 patients, which is in line with previous studies<sup>19,51,52</sup>. Regarding anatomical types, TC and CN coalition had bilateral prevalence of 76.0% and 26.7%, respectively. However, Caucasian studies have reported the bilateral occurrence of CN coalition is higher or similar to that of TC coalition<sup>5,17,20,26</sup>. In addition, single posterior facet lesions accounted for the largest proportions (43.2%) of TC coalition subtypes, which concurs with other East Asian studies<sup>30,31,33</sup>, whereas in Caucasian studies middle facet lesions accounted for more lesions than posterior facet lesions<sup>20,26-29</sup>. Racial differences may explain these differences, but additional larger-scale Asian studies are needed to clarify this topic.

The proportions of patients with coalition-related symptoms were compared in TC and CN coalitions. TC coalition (88.0%) accounted for a significantly higher proportion of symptomatic patients than CN coalition (46.7%), which may be the result of less subtalar motion limitation in CN coalition<sup>35</sup>.

Summarizing the characteristics of tarsal coalition in East Asians, TC coalition is the most common anatomical type and has a very high bilateral occurrence. In addition, TC coalition most often invades the posterior facet, and accompanies coalition-related symptoms at a high rate. This suggests that there are relatively more symptomatic tarsal coalition patients than Caucasians in East Asians. Therefore, it is necessary to consider the possibility of tarsal coalition in not only patients with rigid flat foot deformity but also all East Asian patients who visit the hospital for foot and ankle symptoms. If tarsal coalition is suspected, it is better to perform CT or MRI together than simple radiograph alone. Also, since patients with coalition-related symptoms are more likely to have TC coalition, bilateral examination is very important for diagnosis.

Our study has several strengths. First, it included a large number (839 subjects) of variously aged patients, and second, bilateral CT was performed on all patients. However, there is a possibility that the diagnosis of non-osseous coalition was insufficient due to non-availability of MRI findings. In addition, we depended on the medial records of the hospital to confirm the clinical features at the time of the

examination. Therefore, the possibility of insufficient evaluation on other pathological conditions that may cause symptoms cannot be excluded.

## Conclusion

The prevalence of tarsal coalition in our study was found to be 6%, which is higher than previously reported. TC coalition was the most common coalition type and usually involved the posterior facet, which contrasts with that reported for Caucasians. In addition, talocalcaneal coalition was associated with a high prevalence of bilateral involvement and a high rate of coalition-related symptoms. The possibility of tarsal coalition in foot and ankle patients in East Asians should always be considered. When tarsal coalition is suspected, bilateral examination is essential for diagnosis.

## Declarations

### Data availability statement

Data and materials used and analysed during the current study are available from the corresponding author on reasonable request.

### Competing interests

The authors declare that they have no competing interests

### Authors' contributions

CHP takes responsibility for the integrity of the work as a whole, from inception to published article. JJP designed the research study. JJP, HGS and IHW collected and analysed the data. CHP, JJP and HGS interpreted the results and wrote manuscript, with all authors editing and approving the final manuscript version.

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## Figures

### Figure 1

The canalis-tarsi between the middle and posterior subtalar facets.

## Figure 2

Computed tomography findings of non-osseous talocalcaneal coalition which is similar to degenerative osteoarthritis changes.

## Figure 3

Talar beak sign and C sign on a simple radiograph.



## Figure 4

Drunken waiter sign of talocalcaneal coalition