IJAE
Vol. 122, n. 2: 110-120, 2017
ITALIAN JOURNAL OF ANATOMY AND EMBRYOLOGY

Research article - Basic and applied anatomy

Prevalence of pedal symphalangism in Turkish population. A radiographic survey

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Abstract

Objective: The purpose of this study is to investigate the prevalence of biphalengeal toes in Turkish population and to analyze the differences between genders and symmetry patterns. *Material and methods:* Bilateral foot radiographs of 279 subjects with a mean age of 40.7±16.3 (range, 18-78) years were reviewed to detect presence of pedal biphalangism. There were 146 (52.3%) female and 133 (47.7%) male subjects. *Results:* The overall prevalence of biphalangeal 3rd toe was 0.9%, 4th toe was 2.5% and 5th toe was 45.5%. The frequency of biphalangeal 3rd and 4th toe was statistically similar between genders (p=0.456 and p=0.163 respectively); however the biphalengeal 5th toe was more frequent in female subjects (p=0.004). In 138 (49.5%) subjects 5th toe was normal bilaterally. 113 (40.5%) subjects had bilateral 5th biphalangeal toe, and 28 (10.5%) subjects had biphalangeal 5th toe on one side (asymmetric pattern). 270 (96.8%) subjects had bilateral normal 4th toe, 5 (1.8%) subjects had bilateral biphalangeal 4th toe, and 4 subjects (1.4%) had asymmetric pattern. 276 (98.9%) subjects had bilateral normal 3rd toe, 2 (0.7%) had bilateral bipahangeal 3rd toe, and 1 subject (0.4%) had asymmetric pattern. The symmetric occurrence of biphalangeal toe in 3rd, 4th and 5th toes were equally distributed in both genders (p=0.061, p=0.227 and p=0.477 respectively). *Conclusion:* The present study represents the first report on the prevalence and distribution of biphalengeal toes in Turkish society. The prevalence of biphalengeal toes in Turkish population is considerably different from the Asian and Japanese population, but rather resembles European and North American populations.

Key words

Biphalangeal, fifth toe, epidemiology, Turkish population, symphalangism.

Introduction

The size of the lateral human toes presents a fairly regular mediolateral decrescent trend. This specific morphology of the human toes and decrease in size from medial to lateral has been interpreted as being related to the loss of plantar grip and bipedalism (Lessertisseur and Jouffroy, 1973).

The human foot has a big toe and four lateral toes. Anatomically it is known that the first toe is composed of two phalangeal bones (proximal and distal phalanxes) and the second to fifth toes (lesser toes) are composed of three phalangeal

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http://www.fupress.com/ijae
DOI: 10.13128/IJAE-21315

Figure 1 – (a) A triphangeal toe which has three phalanxes. (b) A biphalangeal toe in which the distal and middle phalanges were fused.

bones (proximal, middle and distal phalanxes). Pedal symphalangism also called as ''biphalangeal toes or two phalanged toes'', refers to the fusion of two contiguous phalanges within the same digit, which is seen as end-to-end synarthrosis of the bones on radiographic examination (Figure 1). In other words, these toes lack distal interphalangeal joints. The pedal symphalangism was first described in 1492 by Leonardo da Vinci (O'Malley CD, 1952). Since the first description several other authors studied this anatomic variant. Although pedal symphalangism can be observed in the second through fifth toes, it is a relatively common anatomic variant that involves the fifth toe.

The incidence of pedal 5th toe in different ethnic populations has been reported between 9.8 and 80.4%, with lower values being seen in Europeans and the highest rates in the Japanese population. However, to the best of our knowledge, there is no study investigating the prevalence of pedal symphalangism in the Turkish population up to date. The purpose of this study was to examine the plain anteroposterior radiographs of the feet in Turkish subjects in order to determine the prevalence of pedal symphalangism and their distribution. Furthermore, we analyzed the differences between genders and symmetry patterns.

Material and methods

We retrospectively reviewed consecutive adult patients (>18 years of age, skeletally mature) to whom bilateral foot radiographs were taken between January 2015 and January 2016 (12 months) for all possible indications, from picture archiving and

communication systems (PACS) and institutional clinical database of two hospitals. All subjects were patients admitted to either the emergency departments or orthopedic outpatient clinics. We excluded 54 patients, in whom the toes were not clearly depicted due to incorrect patient positioning making it impossible to judge about the presence of a biphalangeal toe, or all phalangeal bones of the foot were not demonstrated. Finally, bilateral foot radiographs of 279 patients (558 foot radiographs) were eligible and included in this study. This study was carried out in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments. Since this study was retrospective on radiographs taken for diagnostic purposes it was approved by default by Institutional board.

The presence of biphalangeal toes and their distribution from second through fifth toe was evaluated and recorded by two observers independently at separate times and any discrepancy was subsequently resolved by consensus. Data on patient age and sex were recorded from hospital records. Continuous variables were stated as mean and standard deviation and categorical variables as percentage and frequency distribution. The differences between the biphalangism at a particular location and the side and sex were analyzed using Pearson Chi-Square Test. A P-value < 0.05 was considered as statistically significant.

Results

A total of 279 subjects with a mean age of 40.7±16.3 (range, 18-78) years were analyzed. There were 146 (52.3%) female and 133 (47.7%) male subjects. No subjects had biphalangeal 2nd toe on either side. The overall prevalence of biphalangeal

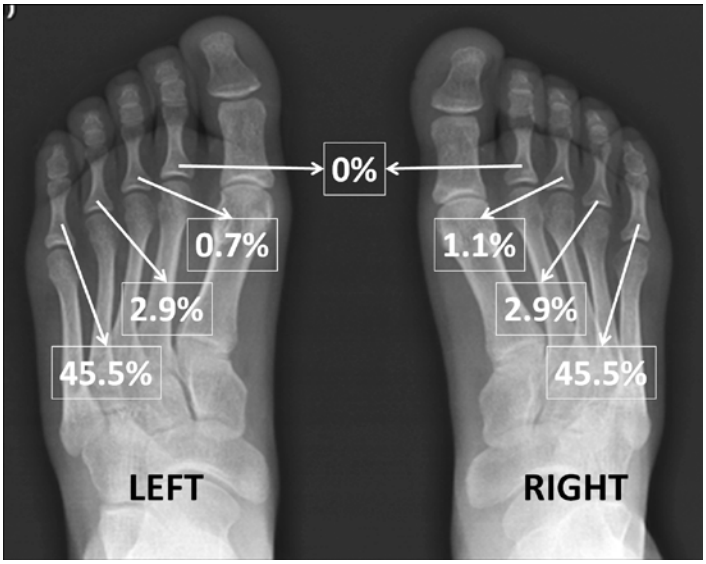

Figure 2 – The distribution of biphalangeal toes and their prevalence in each foot.

3rd toe was 0.9%, 4th toe was 2.5%, and 5th toe was 45.5%. The overall distribution of biphalangism and their percentage incidence on each side is shown in Figure 2. The frequency of biphalangeal 3rd and 4th toe was statistically similar between genders (p=0.456 and p=0.163 respectively); however the biphalangeal 5th toe was more frequent in female subjects (p=0.004) (Table 1).

In 138 (49.5%) subjects the 5th toe was normal bilaterally. 113 (40.5%) subjects had bilateral 5th biphalangeal toe (symmetric pattern), and 28 subjects (10.5%) had biphalangeal 5th toe on one side (asymmetric pattern). 270 (96.8%) subjects had bilateral normal 4th toe, 5 (1.8%) subjects had bilateral 4th biphalangeal toe (symmetric pattern), and 4 subjects (1.4%) had asymmetric pattern (Figure 3). 276 (98.9%) subjects had bilateral normal 3rd toe, 2 (0.7%) subjects had bilateral biphalangeal 3rd toe (symmetric pattern), and 1 subject (0.4%) had asymmetric pattern (Table 2). The symmetric occurrence of biphalangeal toe in 3rd, 4th and 5th toes were equally distributed in both gender (p=0.061, p=0.227 and p=0.477 respectively) (Table 3).

Table 1 – Prevalence of biphalangeal toe with respect to gender and side. P values are the results of Pearson Chi-Square test. (Asterisk means significant p values).

	Male		Female		Significance (*p* value)	
	Right (n,%)	**Left(n,%)**	**Right(n,%)**	**Left(n,%)**	**Right**	**Left**
3rd toe	2 (0.7%)	1(0.4%)	1(0.4%)	1(0.4%)	0,465	0,727
4th toe	4(1.4%)	5(1.8%)	2(0.7%)	3(1.1%)	0,299	0,311
5th toe	53(19.0%)	52(18.6%)	74(26.5%)	75(26.9%)	0,045*	0,026*

Table 2 – Symmetry patterns of biphalangeal toes.

	3rd toe	**4th toe**	**5th toe**
Bilateral normal	276 (98.9%)	270 (96.8%)	138 (49.5%)
Bilateral biphalangeal	2 (0.7%)	5 (1.8%)	113 (40.5%)
Asymmetric	1 (0.4%)	4 (1.4%)	28 (10%)

Table 3 – Differences between male and female subjects with respect to symmetry patterns. *p* values are the results of Pearson Chi-Square test.

	Male		Female		Significance (*p* value)
	Symmetric (n,%)	**Asymmetric (n,%)**	**Symmetric (n,%)**	**Asymmetric (n,%)**	
3rd toe	132 (47.3%)	1 (0.4%)	146 (52.3%)	0 (0%)	0.477
4th toe	130 (46.6%)	3 (1.1%)	145 (52.0%)	1 (0.4%)	0.277
5th toe	124 (44.4%)	9 (3.2%)	127 (45.5%)	19 (6.8%)	0.061

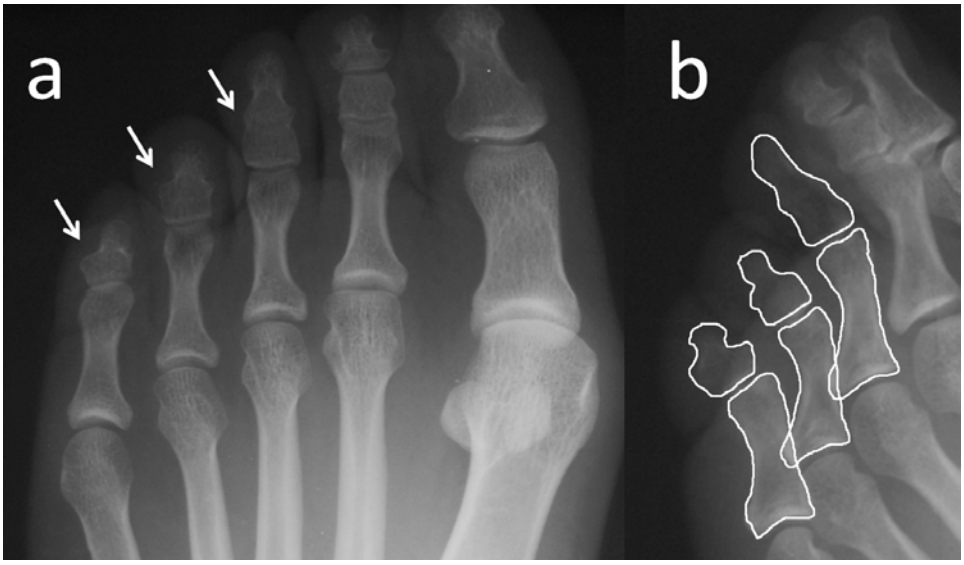

Figure 3 – (a) A 34 year-old female patient with 3rd, 4th and 5th biphalangeal toes. (b) Lateral oblique radiograph of the same subject.

Discussion

This study investigated the prevalence of biphalangism in Turkish subjects. According to our results, the overall prevalence of pedal biphalangism in Turkish population was found to be 45.5% for the fifth toe, 2.5% for the fourth toe, and 0.9% for the third toe respectively. In other words, almost half of the population had biphalangeal 5^{th} toe. The prevalence of biphalangeal 5^{th} toe was statistically higher in female subjects. The symmetric distribution of biphalangeal digits was more frequent than asymmetric pattern.

Several radiographic and cadaveric studies have documented the prevalence of pedal biphalangism in different ethnic groups (Table 4). Although isolated cases of the absence of phalangeal secondary ossification centers have been published, the first quantitative data concerning the occurrence of the phalangeal secondary centers in the lateral four toes was published by Venning (1956). Later on, Billman and Le Minor (2007) stated that the absence of one or more secondary ossification centers could be a derived pattern specific to the human species, like an autapomorphic pattern.

The overall prevalence of pedal biphalangism in Caucasian populations (European and North American) has been reported between 35 and 45%; on the other hand, the overall prevalence of pedal biphalangism in Asian populations (Japanese, Indian, Korean) has been reported between 70 and 80% (Thompson and Chang, 1995). In a study of 2,550 European individuals, lateral toes presenting two rather than three phalanges (biphalangeal variants) were observed in 43.53% of the fifth toes, 2.51% of the fourth, 0.20% of the third, and 0.12% of the second toes (Le Minor, 1995). In the

Table 4 – Previously reported studies fifth toe symphalangism (biphalangeal fifth toe).

Author ,Year	Sample	Feet (n)	Counting method	Study type	2nd toe	3rd toe	4th toe	5th toe
Pfitzner , 1896	German adults	838	Feet	Cadaver	0,4	0,5	1,6	37
Hasselwander, 1903	European children/fetuses	172	Feet	Cadaver	_	_	1,1	47,1
Adachi and Adachi, 1905	Japanese adults	97	Feet	Cadaver	_	_	3	80,4
Hasselwander , 1910	European children/adults	256	Feet	Cadaver	0,8	1,2	4,3	41
Hasebe, 1912	Japanese adults	260	Feet	Cadaver	_	_	7,7	73,5
Nakanishi, 1942	Japanese adults	500	Feet	Radiograph	_	0,4	5,4	72,2
Trolle, 1948	Danish fetuses	370	Feet	Cadaver				
Venning, 1956	European children and adults	4632	Feet	Radiograph	0,04	0,45	2,16	42,53
Asin, 1966	American adults	417	Individual feet	Radiograph	_	_	_	47,5
Ellis et al. 1968	American adults	390	Individual feet	Radiograph	_	_	0,8	48,2
Sandstrom and Hedman, 1971	Swedish children/adults	496	Feet	Radiograph	_	_	1,2	34,5
Winlecki, 1978	American adults	974	Feet	Radiograph	_	_	_	42,1
Carroll et al., 1978	American adults	1324	Individual Feet	Radiograph	_	_	_	33,8
Le Minor, 1995	French adults	2550	Individual Feet	Radiograph	0,1	0,2	2,5	41,02
Nakashima, 1995	Japanese children/adults	488	Feet	Radiograph	0	0,8	11,9	72,5
Park and Sohn, 1998	Korean adults	1187	Feet	Radiograph	_	_	_	74
George , 2001	English old and young adults	204	Feet	Radiograph	_	_	_	37,7
Chae et al., 2002	Korean adults	1290	Feet	Radiograph	0,08	0,54	12,48	72,4
Case and Heilman, 2005	African-American Euro-American Japanese skeletons	550	Feet	Cadaver				

(continued)

Table 4 – Previously reported studies fifth toe symphalangism (biphalangeal fifth toe).

Author ,Year	Sample	Feet (n)	Counting method	Study type	2nd toe	3rd toe	4th toe	5th toe
Rabi et al., 2005	South Indian	24(fetus) 112(children) 263(adult)	Feet	Radiograph Cadaver	–	–	–	87.5 (fetus) 9.8 (children) 11.8 (adults)
Rozen et al., 2005	Australian adults	102	Feet	Cadaver	_	_	_	26
Sohn et al., 2006	Korean children	50	Feet	Radiograph				
Sohn et al., 2006	Korean adults	175	Feet	Radiograph	_	_	_	74,29
Moulton et al, 2012	English adults	606	Feet	Radiograph	_	_	_	44,4
Current study, 2015	Turkish adults	558	Feet	Radiograph	_	0,9	2,5	45,5

present study, our findings resemble the findings in Caucasian population rather than Asian populations.

In one study, no difference in the incidence between men and women was found (Le Minor, 1995), but others concluded that this variant is found more often in women (Venning, 1960). Symptomatic problems of the forefoot predominate in women (Frey, 2000). In our study we found this deformity more frequent among women.

While some studies proposed biphalangism to be symmetric, conflicts may present. George (2001) stated that no difference exists in the percentage of biphalanged 5th toes between left and right foot. Ellis et al. (1968) showed that the patients in whom both feet had been radiographically assesed were either biphalanged or triphalanged on both sides. Moulton et al. (2012) showed a small number of subjects (4.8%) who had different numbers of phalanges in either foot.

Currently there are two different theories about the occurrence of biphalangism. It may be the result of incomplete segmentation, with failure of development of the distal interphalangeal joint during gestational life (Billmann and Le Minor, 2007). Most commonly it is believed that the biphalangeal fifth toe is caused by a pathologic fusion of the middle and distal phalanxes during postnatal life, attributable to trauma or disuse of this joint (Le Minor, 1995).

Dereymaeker and van der Broek (2006) searched for the prevalence of a biphalangeal fifth toe in patients with hammer or claw toe, bunionettes, and overriding fifth toe. They only found statistically difference in hammer or claw toes, in which the prevalence was significantly higher than the control group. So they concluded that the stiffness and rigidity of the biphalangeal fifth toe may predispose it for symptomatic hammer or claw toe. A biphalangeal fifth toe is stiffer and less able to accom-

Figure4 – A 22 year-old male patient with fracture biphalangeal left fifth toe, which was misdiagnosed as normal.

modate the pressure of conventional shoe wear. Likely, Thompson and Chang (1995) have suggested that surgery on the fifth toe is more common in toes with two rather than three phalanges.

With loss of flexibility created by this synostosis, it is often more difficult for the digit to accommodate irritation from shoes and this therefore predisposes the fifth toe to the development of painful pressure keratoses or nail dystrophy (Banks et al., 2001). Kiatisevi et al. (2011) described a giant cell tumor at the distal phalanx of the biphalangeal fifth toe, where it is very rare to find. But they couldn't conclude that biphalangism may have been a reason. Furthermore in context of trauma and fracture biphalangism may be misdiagnosed. But trauma can be easily distinguished from pedal symphalangism because traumatic fusion nearly always results in flexion, mediolateral deviation, or rotation of one phalanx relative to the other, while in pedal symphalangism the combined phalanges will be quite straight (Case and Heilman, 2005; Figure 4).

The present study has some specific strengths. This is the largest series of foot radiographs examined in the current literature in adults. All the radiographs were digital, thus observers could subjectively adjust the display parameters such as contrast, opacity and brightness on the computer screen. This provided a correct identification of all biphalangeal toes. Digital radiography examination is a useful method for determining skeletal ossification and is also simple, objective, fast and relatively inexpensive (Burdan et al., 2002; Rabi et al., 2005). Furthermore, two independent observers reviewed all cases to decrease the errors. Additionally, we could be able to examine the symmetry patterns, as bilateral radiographs were taken.

As a conclusion, the present study represents the first report on the prevalence and distribution of biphalangeal toes in Turkish subjects. The prevalence of pedal biphalangism in Turkish population is considerably high, and it resembles the European population.

Conflict of interest

The authors have no conflict of interest to declare.

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