

Literature Review on Brachymetatarsia

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Brachymetatarsia is a malformation characterized by an abnormal reduction in the length of 1 or more metatarsal bones. It occurs because of early closure of the growth plate of the affected metatarsal. Generally, it is caused by a congenital disorder and it usually occurs bilaterally. With a greater prevalence in females, it most often affects the fourth metatarsal, followed by the first metatarsal. Surgical treatments proposed include using external mini-fixators or bone grafts in a single step to lengthen the metatarsal. In this review, 62 scientific articles about brachymetatarsia were analyzed with key demographic and epidemiological aspects of this pathology. The prevalence of bilateral brachymetatarsia was 47%, and the female to male ratio was 10.53:1. Both these findings appear to contradict the usual data reported for brachymetatarsia. A better understanding of this disorder will enable an appropriate therapeutic approach according to the psychological and social profile of affected individuals.

Introduction

Brachymetatarsia, or hypoplastic metatarsal, is a rare condition characterized by an abnormally short metatarsal due to premature closure of the growth plate. According to the authors consulted, existing epidemiological data on the pathology are variable and its incidence is low, ranging between 0.02% and 0.05% (Jones, Pinegard, & Rincker, 2015; Urano & Kobayashi, 1978). Most cases of brachymetatarsia published in the literature correspond to cases of idiopathic congenital primary causes, although congenital cases associated with various diseases or syndromes have also been reported (Kim, Lee, Yoo, Kanq, & Suh, 2003). To a lesser extent, cases of brachymetatarsia, derived from trauma, tumors, infections, or iatrogenic conditions, have been reported (Kashuk, Hanft, Schabler, & Kopelman, 1991; Steedman & Peterson, 1992; Shim & Park, 2006). Existing studies coincide in highlighting a greater prevalence in females, although with a very varied relationship (Giannini, Faldini, Pagkrati, Miscione, & Luciani, 2010; Lee, Yang, Chung, Moon, & Jung, 2009; Trujillo Pérez, Reina Bueno, Lafuente, & Munuera Martínez, 2007). Regarding the prevalence of bilaterality, there are also significant discrepancies, ranging between 36% and 72% (Barbier, Neretin, Journeau, & Popkov, 2015; Urano & Kobayashi, 1978). The fourth metatarsal was in all series the most affected (see Figures 1 and 2), followed by the first metatarsal.

The specific trigger that causes premature closure of the growth plate is unknown, although, currently, the most supported etiology appears to be related to the presence of genetic markers (Kawashima, Yamada, Ueda, & Harii, 1994). This hypothesis is supported by the fact that embryonic separation of the membrane between the second and third toes takes place last and that the fourth and the fifth metatarsals are also the last to begin development during the fetal stage. In general, deformity begins during the embryonic period and continues during development of the individual, causing the growth of the metatarsal to slow down during its entire morphogenesis. The pathology is often detected several years after birth and usually becomes evident from childhood until the age of fourteen, when the growth plate of metatarsal closes definitively (Hinrichsen et al., 1994). Incomplete syndactyly has been often associated with brachymetatarsia. The fact that the membrane between the second and third toes is usually the last to appear during fetal development, and the fourth and fifth metatarsals are the last to begin development, suggests that the mechanisms producing the two anomalies could be related (Munuera Martínez, Lafuente Sotillos, Domínguez Maldonado, Salcini Macías, & Martínez Camuña, 2004).

Clinical symptoms of brachymetatarsia may be varied and are conditioned by factors such as age, sex, and number of metatarsals affected in addition to whether there is a bilateral presentation. The common denominator of the deformity is that it turns out to be aesthetically unacceptable for most subjects who suffer from this. During puberty, and especially during adolescence, it is common for this to cause a distorted body image that at times may have a psychological impact. Adults often consult for pain (transfer metatarsalgia), skin irritation of the toe above the corresponding commissure with shoe wear, or difficulty walking (Barbier et al., 2015; Giannini et al., 2010).

Management of the pathology may be conservative or surgical. The latter has been more commonly used

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The authors and planners have disclosed no potential conflicts of interest, financial or otherwise.

DOI: 10.1097/NOR.0000000000000487



FIGURE 1. Unilateral brachymetatarsia of the fourth metatarsal in a 28-year-old woman.

and consists of recovering the metatarsal parabola with the purpose of achieving an aesthetically acceptable foot while relieving metatarsalgia by transfer, as appropriate. The most commonly used surgical techniques were elongation with interposition of bone graft and sequential lengthening by means of callotasis using external fixators (Jones et al., 2015).

Authors increasingly highlight the importance of individually evaluating the emotional and psychological tolerance of patients for the different therapeutic options. Although brachymetatarsia is a rare clinical condition, this may affect the individual not only from a functional point of view but also from a psychological and social point of view. A personalized study of the individual from a general perspective may allow selecting the most suitable therapeutic option and the most appropriate surgical alternative according to available evidence and the subject's characteristics (Jones et al., 2015).

The present study is a critical review of the published data to record and evaluate the demographic data and incidence of brachymetatarsia to compare these data with often reported values. We believe that a descriptive analysis of the demographic, etiological, and morpho-functional aspects related to the pathology may allow identifying key aspects when planning correct treatment management according to the demographic pattern of the individuals affected.



FIGURE 2. Dorsoplantar radiograph of the patient's left foot. An abnormally short fourth metatarsal can be observed.

Search Strategies

The literature related to brachymetatarsia was identified using three common databases: PubMed, CINAHL, and Scopus, as well as other health science database. A search of the published biomedical data was performed using "brachymetatarsia" as the key word. The inclusion criteria were studies in any language reporting on patients treated conservatively or surgically. The exclusion criteria were studies that described only cases of brachymetatarsia derived from disease processes or syndromes, those that described only the surgical techniques, or those that did not report on the surgical procedures performed on the patients. This search yielded 62 results. The selected studies were published from April 1979 to July 2016, and they all had most of the following variables: gender, age, laterality, affected metatarsal, etiology, reason for consultation, and treatment applied.

Brachymetatarsia Research Findings

The studies included in this review were generally poor in quality. Only two were comparative studies (Choi, Chung, Baek, Cho, & Chung, 1999; Lee et al., 2009), and the rest were case series or case reports (see the Appendix). From all the articles selected ($N = 62$), a total of 407 patients (690 feet) affected with brachymetatarsia were identified; of these, most of the cases were of congenital or idiopathic etiology. The average age of patients was 20.52 years, with a range between 9 and

53 years. The data collated regarding sex revealed a higher prevalence in females, with an average female to male ratio of 10.53:1. Regarding laterality, unilateral presentation was slightly more common. Of all the feet analyzed, in more than one third of the cases, the fourth metatarsal was the one affected, followed by the first metatarsal. When more than one metatarsal in the same foot was affected, the most common combination was the first and fourth metatarsals. Regarding the reason for consultation, almost half of the individuals consulted for cosmetic problems, followed closely by those who consulted for pain or difficulty walking. Of the treatments administered, surgery was by far the most commonly used. More than 95% of the cases were treated by means of single-stage lengthening with bone graft or callus distraction with external distracters (see Table 1).

TABLE 1. DETAILS OF THE EPIDEMIOLOGICAL DATA OF THE SERIES OR CASE REPORTS

	<i>n</i> (%)	Total
Gender ^a		346
Female	316 (91.32)	
Male	30 (8.67)	
Patient age range ^a		407
10–20	193 (47.42)	
20–30	211 (51.84)	
30–60	3 (0.73)	
Etiology ^b		690
Congenital	685 (99.27)	
Traumatic	3 (0.43)	
Iatrogenic	2 (0.28)	
Bilateralism ^a		320
Unilateral	171 (53)	
Bilateral	149 (47)	
Reason for consultation ^a		415
Cosmetic	204 (49.15)	
Pain or difficulty walking	195 (46.98)	
Shoe wear irritation	16 (3.85)	
Affected ray ^c		447
M4	345 (77.18)	
M1	61 (13.64)	
M1 ± M4	13 (2.90)	
M3 ± M4	7 (1.56)	
Other associations	21 (4.69)	
Treatment ^c		711
Single-stage lengthening with bone graft	369 (51.8)	
Callus distraction	313 (44.0)	
Slide or transpositional osteotomy	3 (0.42)	
Others (orthotics)	27 (3.79)	

^aValues presented as no. of subjects.

^bValues presented as no. of feet.

^cValues presented as no. of metatarsals.

Discussion

The data regarding some of the variables analyzed were not available in all the articles reviewed. However, the data collected and subsequent analysis allowed to compile in-depth epidemiological and demographic aspects of major interest about the pathology.

Regarding the number of cases, it was observed that the oldest articles based their results on just one case. However, from the late 1990s, some authors began to publish case series that analyzed a higher number of patients, with an average of more than 10 patients per article, which allowed to compile a larger amount of data.

Regarding the etiology of the deformity, most cases corresponded to brachymetatarsia of congenital or idiopathic etiology. There were few cases of the pathology that were derived from syndromes or diseases, trauma, iatrogenic conditions, or infections.

Most individuals who consulted for the problem did so during adolescence or youth and always over the age of 9 years (no case of surgery in children under this age was collated). This could be explained by the fact that the growth plate of metatarsals closes definitively around the age of 14 years and also it is during adolescence when the existence of deformity often causes body image disorders. This is likely largely due to the psychological and emotional component entailed by the pathology and which is especially marked during adolescence. This does not mean that this is not visible at earlier ages, but in most cases, it is unusual that up until adolescence the pathology leads to the need to consult because of aesthetic or functional problems or both.

The data recorded according to the metatarsal affected concur with the information provided in this regard by different authors. All the studies of case series revealed that the fourth metatarsal was by far the most affected with 77.18% of the cases recorded, followed by the first metatarsal. However, the involvement of more than one metatarsal was rare and the most common combination was the first and fourth metatarsals.

The most interesting part of our review was the contradictory data between the results of our review and the results reported in the literature regarding the prevalence of bilaterality and male to female ratio. Regarding laterality, most authors estimated the prevalence of bilaterality above 70%, well above the bilaterality obtained in this review and more in line with recent reviews (Jones et al., 2015). Regarding the prevalence of the pathology according to sex, it was observed that although the pathology was considerably more prevalent in women, the estimated male to female ratio shown by the results of this review is approximately half of that reported by most studies (Urano & Kobayashi, 1978). Although, in principle, it is worth considering the possibility that the pathology could be related to genetic markers linked to sex, it cannot be ruled out that this high prevalence may be due, in part, to the fact that the deformity is commonly associated with a high component of abnormality of body image and the social rejection far removed from the standards of beauty established that may have led women to consult more frequently than men.

Regarding the treatment used, surgery was by far the most common option. Specifically, single-stage

lengthening with a bone graft was the most commonly used treatment, followed by callus distraction. A recent review compared both treatments and concluded that single-stage lengthening with a bone graft was associated not only with fewer complications and faster healing times than callus distraction but also with lesser gains in length (Jones et al., 2015). Given that most patients with brachymetatarsia opt for surgery, healthcare professionals involved in managing these patients should individually recommend the best surgical option considering not only physical and demographic aspects but also psychological and social aspects.

The fact that some studies set out exclusion criteria to select the metatarsal affected, the age of patients or the kind of treatment provided may have caused some bias in some of the results obtained, which are some limitations of this review.

Nursing Implications

Given the psychoemotional component frequently associated with the deformity, orthopaedic nurses are essential when empathizing with patients with brachymetatarsia. Therefore, it is important to consider not only physical aspects associated with the pathology but also demographic factors such as age and gender in addition to social factors regarding the environment and profession, among others. Knowledge of the epidemiological factors linked to the pathology is key when establishing the most indicated therapeutic management in each case. However, it is important to perform a correct psychological evaluation of the individuals that allows identifying the presence of psychological distress and whether they may be candidates for surgery, which would allow providing them with the best surgical alternative. Orthopaedic nurses must be able to detect the existence of a body dysmorphic disorder associated with the pathology, for which they should recommend the patients to consult a psychiatrist before considering surgery and, if this is indicated, recommending the most suitable surgical procedure. Body dysmorphic disorder is a rare psychiatric condition defined by an obsession or preoccupation with a minor or nonexistent flaw in physical appearance that causes functional impairment or significant distress that is not explained by another psychological disorder. This disorder has been found to be up to 15 times more prevalent in young patients seeking plastic surgery. Patients with this disorder often have poor insight and frequently seek plastic surgery consultation over psychiatric consultation. Because of their habits, patients with body dysmorphic disorder often have broken social relationships, live alone, and avoid social situations where their perceived defect will be noticed.

Brachymetatarsia treatment must be focused not only on resolving the physical distress but also on handling the psychological aspects associated with the pathology. Nursing procedures should be aimed at improving aspects that enable accepting the deformity with patient education and social support and, in this case, providing counsel on all the beneficial surgical aspects to help this patient population.

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Authors	No. Subjects	Affected Ray	Reason for Consultation	Gender	Laterality	Age	Treatment	Etiology
D'Arrigo, Arenas Planellas, Erasó Lara, & Jiménez Samiento, 2011	1	M4	Metatarsalgia	Male	Unilateral	24	Callus distraction	Idiopathic
Berges Manfredi, Suárez Méndez, Gallart Ortega, & Valero Salas, 2000	1	M3, M4, and M5 (right foot) M3 and M4 (left foot)	Metatarsalgia	Male	Bilateral	13	Orthotic	Idiopathic
Munuera Martínez et al., 2004	20	M4	NA	NA	Unilateral (12; 60%) Bilateral (8; 40%)	NA	NA	Idiopathic (18) Congenital (1) Posttraumatic (1)
Marcinko, Rappaport, & Gordon, 1984	1	M4	Transfer metatarsalgia Shoe injuries	Female	Unilateral	12	Callus distraction	Posttraumatic
Page, Dockery, & Vance, 1983	1	M4	Metatarsalgia	Female	Unilateral	16	Calnan-Nicolle prosthesis	Congenital
Goforth & Overbeek, 2001	1	M3 and M4	Metatarsalgia	Female	Unilateral	20	Transversal-osteotomy + tubular plates	Idiopathic
Jimenez, 1979	1	M3	Transfer metatarsalgia	Female	Bilateral	14	Bone graft	Congenital
McGlamry & Fenton, 1983	1	M4	Transfer metatarsalgia	Female	Unilateral	12	Bone graft	Congenital
Solomon & Blackledge, 1995	1	M4	Transfer metatarsalgia	Female	Unilateral	13	Bone graft	Congenital
Froehlich & Wuenschel, 2014	1	M1	Disturbance of body image	Female	Bilateral	15	Callus distraction	Idiopathic
Guizar-Cuevas, Mora-Rios, Mejia-Rohenes, López-Marmolejo, & Cortés-Gómez, 2010	7	NA	NA	NA	Unilateral (4) Bilateral (3)	8-15	Callus distraction	Congenital
Lee et al., 2010	48	M1 (19) M4 (29)	Disturbance of body image	NA	NA	18-20	Callus distraction	Idiopathic
Lee, Suh, Moon, & Kim, 2009	106	M4	NA	Female (100) Male (6)	NA	13-48 (26.3)	Bone graft (35) Callus distraction (71)	Congenital
Blankenhorn, Kerner, & DiGiovanni, 2010	1	M4	Transfer metatarsalgia Shoe injuries	Female	Bilateral	25	Bone graft	Congenital

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APPENDIX. RESEARCH STUDIES ABOUT BRACHYMETATARSIA (Continued)

Authors	No. Subjects	Affected Ray	Reason for Consultation	Gender	Lateralism	Age	Treatment	Etiology
Lee, Yoo, & Moon, 2009	30	NA	NA	NA	NA	20–36	Bone graft	Congenital
Lee et al., 2009	16	M1	Transfer metatarsalgia (14) Disturbance of body image (16)	Female (15) Male (1)	NA	12–34	Callus distraction	Congenital
Gilbody & Navagam, 2008	1	M1	Transfer metatarsalgia	Female	Bilateral	13	Callus distraction	Congenital
Yamada, Yasuda, Hashimoto, Iwashiro, & Uchinuma, 2005	1	M4	NA	NA	Bilateral	NA	Bone graft	Congenital
Wilusz, Van, & Pupp, 2007	4	M4	NA	Female (4)	Unilateral (3) Bilateral (1)	NA	Callus distraction	Congenital
Tomic, Krajcinovic, & Dakic, 2000	10	M4 (9) M3 (1)	Transfer metatarsalgia Disturbance of body image.	NA	Unilateral (1) Bilateral (9)	NA	Callus distraction	Congenital
Choi et al., 1999	15	M4 (12) M3 (2) M1 (1)	Transfer metatarsalgia Disturbance of body image	Female (15)	Unilateral (6) Bilateral (9)	10.8–14 (12.3)	Callus distraction (5) Bone graft (10)	Congenital
Baek & Chung, 1998	21	M4 (16) M1 (1) M1 and M4 (2) M1, M3, and M4 (1) M4 and PF (proximal phalanx) (1)	Transfer metatarsalgia (10) Disturbance of body image (21)	Female (21)	Unilateral (11) Bilateral (10)	10–36 (16)	Bone graft	Congenital
Alter, Feinman, & Rosen, 1995	1	M4	NA	Male	Unilateral	14	Bone graft	Congenital
Masuda, Match, Nakajima, Tomi, & Ohba, 1995	6	M2, M3, and M4	NA	Female (6)	Unilateral (3) Bilateral (3)	NA	Callus distraction	Congenital
Kawashima et al., 1994	3	M4	NA	NA	Unilateral (2) Bilateral (1)	NA	Callus distraction	Congenital
Steedman & Peterson, 1992	4	M1	NA	NA	Unilateral (2) Bilateral (2)	NA	Bone graft	latrogenesis (2) Congenital (2)
Glickman & Cornfield, 1990	1	M1	NA	NA	Bilateral	NA	Bone graft	Congenital

(continues)

Authors	No. Subjects	Affected Ray	Reason for Consultation	Gender	Laterality	Age	Treatment	Etiology
Hwang, Song, & Kim, 2012	5	M1	NA	NA	Unilateral (1) Bilateral (4)	NA	Callus distraction	Congenital
Brown, Yeoman, Roberts, & Pillai, 2012	1	M4	Disturbance of body image	Male	Bilateral	NA	Bone graft	Congenital
Song et al., 2003	16	M4	NA	NA	Unilateral (10) Bilateral (6)	NA	Callus distraction	Congenital
Masada, Fujita, Fuji, & Ohno, 1999	4	M4	NA	NA	Unilateral (2) Bilateral (2)	NA	Callus distraction	Congenital
Kim, Baek, Chung, & Yoon, 2004	5	M1 and M4 (4) M1, M4, and M5 (1)	Disturbance of body image	Female (5)	Bilateral (5)	10-26 (15.8)	Bone graft	Congenital
Giannini et al., 2010	29	M4 (23) M3 (1) M3 and M4 (5)	Transfer metatarsalgia Disturbance of body image	Female (26) Male (3)	Unilateral (17) Bilateral (12)	12-42 (27)	Bone graft	Congenital
Scher, Blyakher, & Krantzow, 2010	1	M4	Disturbance of body image	Female	Unilateral	16	Callus distraction	Congenital
Shim & Park, 2006	17	M4 (9) M1 (7) M5 (1)	Transfer metatarsalgia Disturbance of body image	Female (15) Male (2)	Unilateral (5) Bilateral (12)	11-41 (19.5)	Callus distraction	Congenital (16) Traumatic (1)
Kline, 2009	1	M4	Transfer metatarsalgia	Female	Unilateral	13	Bone graft.	Congenital
Wingenfeld, Arbab, & Abbata-Czardybon, 2013	1	M4	NA	Female	Unilateral	NA	Callus distraction	Congenital
Bartolomei, 1990	1	M4	Intractable plantar keratosis	Female	Unilateral	53	Bone graft	Congenital
Frankel & Fleishman, 1991	1	M4	Shoe injuries	Female	Unilateral	22	Bone graft	Congenital
Remón Davila et al., 2008	6	M4	NA	Female (6)	Unilateral (1) Bilateral (5)	10-15 (13.2)	Callus distraction	Congenital
Rock, Gaspari, & Mancuso, 1993	1	M4	Disturbance of body image	Female	Bilateral	44	Bone graft	Congenital
Houshian, Skov, & Weeth, 2002	2	M4 (2)	Disturbance of body image (2)	Female (2)	Unilateral (1) Bilateral (1)	14 10	Callus distraction	Congenital
Hosokawa & Susuki, 1987	1	M4	NA	Female	Bilateral	14	Bone graft.	Congenital

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APPENDIX. RESEARCH STUDIES ABOUT BRACHYMETATARSIA (Continued)

Authors	No. Subjects	Affected Ray	Reason for Consultation	Gender	Lateralism	Age	Treatment	Etiology
Kashuk et al., 1991	3	M5 M4 (2)	Transfer metatarsalgia	Female (3)	Bilateral (1) Unilateral (2)	22 34 40	Bone graft.	latrogenesis (1)
Boike, Gerber, & Snyder, 1993	1	M2	Transfer metatarsalgia	Female	Unilateral	16	Callus distraction	Congenital
Trujillo Pérez et al., 2007.	25	M4	NA	Female (19) Male (6)	Unilateral (13) Bilateral (12)	NA	Orthotic	NA
Robinson & Ouzounian, 1998	1	M3 and M4	Transfer metatarsalgia Hallux abducto valgus	Female	Unilateral	15	Bone graft	Congenital
Handelman, Perlman, & Coleman, 1986	1	M4	Transfer metatarsalgia	Female	Bilateral	27	Bone graft	Congenital
Kim et al., 2003	12	M4 (5) M4 and M5 (1) M1 and M4 (5) M1, M3, M4, and M5 (1)	Hygiene alterations Transfer metatarsalgia Disturbance of body image	Female (9) Male (3)	Unilateral (6) Bilateral (6)	NA	Bone-graft	NA
Ferrández, Yubero, Usabiaga, & Ramos, 1993	3	M4 (2) M3 (1)	Plantar keratosis (3) Disturbance of body image (2)	Female	Bilateral (1) Unilateral (2)	11 13 20	Callus distraction	Congenital
Desai, Lidder, Armitage, Rajaratnam, & Skyrme, 2013	1	M4	Disturbance of body image	Female	Unilateral	16	Bone graft	Idiopathic
Cortés-Rodríguez, Romano-Juárez, Castañeda-Pichardo, & Tercero-Quintanilla, 2009	14	NA	Transfer metatarsalgia Disturbance of body image	Female (12) Male (2)	Unilateral (12) Bilateral (2)	10–18	Callus distraction	Idiopathic
Fox, 1998	6	M4	Metatarsalgia. Disturbance of body image	Female (6)	Unilateral (6)	NA	Callus distraction	Congenital
Hamada, Sakamoto, Nagasao, & Kishi, 2016	1	M4	Disturbance of body image	Female	Bilateral	16	Callus distraction	Idiopathic
Montero-Quijano et al., 2015	15	M4 (13)	Disturbance of body image; transfer metatarsalgia	Female (11) Male (4)	Unilateral (5) Bilateral (3) NA (7)	13	Callus distraction	Congenital

(continues)

APPENDIX. RESEARCH STUDIES ABOUT BRACHYMETATARSIA (Continued)

Authors	No. Subjects	Affected Ray	Reason for Consultation	Gender	Lateralism	Age	Treatment	Etiology
Hosny & Ahmed, 2016	6	M4	Disturbance of body image	Female	Unilateral (1) Bilateral (5)	23	Callus distraction	Congenital
Barbier et al., 2015	30	NA	Transfer metatarsalgia	NA	Unilateral (22) Bilateral (8)	20.5	Callus distraction	Congenital
Smolle, Scheipl, Leithner, & Radl, 2015	4	M4	Transfer metatarsalgia	NA	NA	24.8	Bone graft	Congenital
Haleem, Mintz, & Rozbruch, 2014	1	M4	Disturbance of body image; transfer metatarsalgia	Female	Unilateral	39	Callus distraction	Congenital
Oh, Satish, Lee, & Song, 2004	8	M1 (5) M1 and M4 (2) M1 and M5 (1)	Disturbance of body image; transfer metatarsalgia	Female (4) Male (4)	Unilateral (3) Bilateral (5)	18.8	Callus distraction	Congenital (7) Traumatic (1)

Note. NA = not available.