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Research Article – Basic and Applied Anatomy

The fourth slip of the flexor digitorum brevis muscle of the human foot. A systematic review and meta-analysis

Kaissar Yammine

The Foot and Hand Clinic and the Center for Evidence-Based Sport & Orthopedic Research, Division of Evidencebased Anatomy, Emirates Hospital, Dubai, UAE

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Abstract

The flexor digitorum brevis muscle, and in particular its fourth slip, has a significant clinical and surgical importance in medical practice. However, as for the majority of tendons destined to the little toe, the fourth slip is undergoing a phylogenetic degeneration. The aim of this meta-analysis is to conduct an evidence synthesis on the prevalence of FDB-5 and its variants in humans. Twenty-two studies met pre-defined inclusion criteria with a total of 2789 feet and 416 cadavers. Meta-analytical results of fourth slip agenesis were as follows; a) a true prevalence rate of 31.3%, b) a crude prevalence rate of 47%, c) a bilateral prevalence rate of 38.2%, d) a true prevalence rate of 77.3% in Indian populations, e) a true prevalence rate of $\approx 20\%$ in Japanese, Chinese, American, European and Turkish populations, f) an Odds Ratio of 1.5 significantly in favor for female gender, g) non-significance for hand side, h) a true prevalence rate of a thin fourth slip of 47.7%, and i) a true prevalence rate of the variations of its origin in 12.3%. The knowledge of the frequency of flexor digitorum brevis fourth slip agenesis and variations in relation to the demographic characteristics of patients would be of importance for tendon repair, tendon transfer or soft tissue reconstruction in foot surgery.

Keywords -

Flexor digitorum brevis, foot, anatomy, meta-analysis.

Key to abbreviations:

FDB = flexor digitorum brevis FDB-5 = flexor digitorum brevis slip to the little toe PPE = pooled prevalence estimate OR = Odds ratio

Introduction

The flexor digitorum brevis (FDB) is one of those muscles considered undergoing a phylogenetic degeneration (Lobo et al., 2008; Lakshmi, 2010) mainly owing to the various anatomical presentations, such as the absence or the hypotrophy of its fourth slip destined to the little toe.

^{*} Corresponding author. E-mail: kayseryam@yahoo.com.

The FDB muscle takes origin from the posteromedial calcaneal tuberosity, the posterior third of the deep surface of the plantar aponeurosis, and the lateral and medial intermuscular septa (Sarrafian and Kelikian, 2012). It lies between the abductor hallucis medially and the abductor digiti quinti laterally (Fig. 1). The body muscle then divides into four fascicles each followed by a tendon slip, superficial to the corresponding long flexor tendon and which divides into two slips at the level of the metatarso-phalangeal joint (Testut, 1884). These slips insert on the inferior aspect of the middle phalanx creating a groove for the passage of the long flexor tendon (Rosse and Gaddum, 1997; Williams et al., 2004).

Anatomical variations

The different variations affecting the tendon slips of the FDB may be in the form of agenesis, associated or not to the presence of a small muscular slip from the long flexor or flexor accessorius, or in the form of thin slips. (Nathan and Gloobe, 1974; Sarrafian and Kelikian, 2012). There is an agreement that the tendon slip to the little toe (FDB-5) is much more susceptible to variations compared with slips to other toes. Variations in FDB tendon were noted in 63% of the limbs, involving mostly the fifth toe and to a lesser degree the fourth toe (Nathan and Gloobe, 1974). The average absence of the fourth slip (FDB-5) was estimated at 21.5% (Sarrafian and Kelikian, 2012) and the absence of the slips for the fourth and fifth toes at 3% (Testut, 1884). In



Figure 1 – Slips of the flexor digitorum brevis (reprinted and modified from Wagenseil,, 1936, with permission by E. Schweizerbart'sche Verlagsbuchhandlung OHG (Naegele u. Obermiller), Stuttgart, Germany: www. schweizerbart.de.

case of absence, a substitute is sometimes observed in the form of a slender fusiform belly from the flexor digitorum longus (FDL) or from the inner tubercle of the os calcis (Wood, 1868; Macalister, 1875).

An unsplit fourth tendon slip has been seen in 5% of subjects and a fusion of this slip with the long flexor was estimated, not counted, at 2% (Testut, 1884; 1892). Smaller slips to the fifth toe were found with a frequency up to 64.5% (Schwalbe and Pfitzner, 1891), and an additional slip from the FDL up to 20% (Nathan and Gloobe, 1974) or from the intermuscular septum in 3% (Yalcin and Ozan, 2005;Gugapriya, 2012). A recent study by Becerro de Bengoa Vallejo et al. (2011) found no cases of variation in the insertion of the FDB tendon into the second, third, or fourth digits in 45 feet.

Clinical relevance

The FDB is related to toe deformities such as the congenital curly toe (Tokioka et al., 2007) and hammer toes. Transfer of the FDB to the proximal interphalangeal joint has been found effective in the prevention of floating toes after Weil osteotomy (Lee and al., 2013) and in toe ulcers of claw- or hammer-toes in diabetic patients (Rasmussen et al., 2013). FDB transfer to the interosseous and lumbrical muscles has been effectively used in treating dynamic claw toe deformity (Errichiello et al., 2012). Transposition of the flexor digitorum brevis tendon has been described for flexible hammer toes (Becerro de Bengoa Vallejo et al., 2008; 2011). Its clinical relevance extends to soft tissue reconstruction; the FDB flap has been used to cover heel and distal plantar defects (Sakai et al., 2001; Attinger et al., 2002).

Aim of research

The aim of this systematic review is to provide more accurate data on the prevalence of FDB-5 tendon slip and its variants.

Methods

The Checklist for Anatomical Reviews and Meta-Analysis (CARMA) guidelines were followed while conducting this review (Yammine, 2014).

Search strategy and identification of studies

A systematic literature search was conducted through a number of electronic databases such as Medline, Embase, Scielo, EBSCO, and Google Scholar from inception to Feb 2014, using the Boolean combination of broad terms such as ("flexor brevis" OR "flexor digitorum brevis" OR "4th slip^{*}" OR "fourth slip^{*}" OR "flexor digitorum pedis brevis" OR "flexor digitorum communis brevis" OR "flexor digitorum sublimis brevis" OR "flexor digitorum perforatus brevis" OR "flexor digitorum sublimis brevis" OR "flexor digitorum perforatus brevis" OR "flexor digitorum sublimis articles. We also searched the websites of the following journals: *Acta Anatomica, Anatomical Record (A and B), Anatomical Sciences International, Annals of Anatomy, Clinical Anatomy, European Journal of Anatomy, European Journal of Morphology, Folia Morpholog-* ica, Foot and Ankle Surgery, Foot and Ankle Specialist, International Journal of Anatomical variations, International Journal of Morphology, Italian Journal of Anatomy and Embryology, Journal of the American Podiatric Medical Association, Journal of Anatomy, Journal of Bone and Joint Surgery [Br and Am], Journal of Morphology, Okajimas Folia Anatomica (Japan), Romanian Journal of Morphology and Embryology, Surgical and Radiological Anatomy, The Foot. All included articles were citation-tracked using Google Scholar to ensure that all relevant articles were identified. Duplicates were deleted.

Criteria for study selection

Literature concerning the prevalence of the variants of the FDB-5 is infrequent, so all published or unpublished studies reporting prevalence rates were included in the review. The primary outcomes are the true or the crude prevalence of FDB-5 or its variants in cadaveric studies. The true FDB-5 prevalence rate is defined as the number of feet affected compared to the number of feet available for study. The crude FDB-5 prevalence is the number of individuals who have either one or two FDB-5 compared to the number of individuals available for study. Secondary outcomes are the prevalence in relation to ancestry, gender, laterality and side, the interactions between those variables, and the variant types of FDB-5. To ensure unbiased selection of included studies, abstracts from conferences were not included. No restriction was imposed on date, language or age. Titles and abstracts were initially screened and full-text articles were obtained when at least one primary outcome was thought to be reported.

Data extraction and analysis

Data extracted included sample size, sample details, type of investigation (clinical or cadaveric), and the results. Analysis was performed using StatsDirect v2.7.8 (Stats-Direct, Altrincham, UK). Proportion meta-analysis was used to calculate the pooled prevalence estimate (PPE), and odds ratio (OR) meta-analysis was used to establish potential associations with other variables such as ancestry, gender, laterality or side. The "two independent proportion test" was used to look for significant proportion differences between studies reporting FDB-5 frequencies in different ancestry populations.Descriptive analysis was conducted when the data were not amenable to meta-analysis. We examined heterogeneity amongst studies using I² statistics; whenever I²> 50%, the random-effect estimate was reported.

Results

Search results

The electronic search strategy yielded 197 hits; abstracts of 43 potentially relevant studies were scanned and 10 studies were retained for inclusion. Reasons for exclusion were as follows; 22 case reports and 11 clinical studies with no data on FDB prevalence. Reference checking of the included studies yielded additional 7 studies and their reference checking added another five. In total, 22 studies met our inclusion criteria (Table 1 and 2).

Studies	Population	Age	Sample size: cadavers	Male	Female	Sample size: feet	Right	Left
Turner, 1865	British	Adults?	1	ı	ı	50	ı	ı
Wood, 1866	British	Adults?	34	ı	ı	68	ı	ı
Wood, 1868	British	Adults?	102	68	34	204	102	102
Schwalbe and Pfitzner, 1889	German	Adults	ı	ı	ı	132	ı	ı
Schwalbe and Pfitzner, 1891	German	Adults	ı	ı	ı	214	ı	ı
Schwalbe and Pfitzner, 1894	German	Adults?	540	367	173	1080	540	540
Le Double, 1897	French	Adults?	100	50	50	200	100	100
Adachi, 1900	Japanese	·	ı	ı	ı	226	ı	ı
Koganei et al., 1903	Japanese	ı	ı	ı	ı	292	ı	ı
Adachi, 1909	Japanese	ı	ı	ı	ı	141	ı	ı
Kurz, 1923	Chinese	Adults & children	ı	116	30	146	ı	ı
Wagenseil, 1936	Chinese	Adults	ı	111	29	140	ı	ı
Nathaniel, 1954	Indian	Adults	ı	ı	ı	60	ı	ı
Mori, 1964	Japanese	Adults	ı	ı	ı	50	ı	ı
Nathan and Gloobe, 1974	American	ı	ı	ı	ı	100	ı	ı
Chaney et al., 1996	American	ı	ı	ı	ı	284	ı	ı
Kura et al., 1997	American	50-80	11	8	Э	11	4	г
Yalçin and Ozan, 2005	Turkish	33-74	15 cadavers + 3 legs	9 cadav.	7cadav.	33 (unknown side for 3)	15 out of 30	15 out of 30
Becerro de Bengoa Vallejo et al., 2008	Spanish	Adults	ı	ı	ı	45	ı	ı
Lobo et al., 2008	Indian	Adults	30	20	10	60	30	30
Locke et al., 2010	British	71-88	4 (random from 15)	H	ß	4	ı	ı
Gugapriya 2012	Tamil (Indian)	25-70	15	12	ß	30	15	15
llayperuma 2012	Sri Lankan	48-67	135	81	54	270	135	135
Bernhard et al., 2013	American	·	57 (51 Caucasians, 6 Africans)	33	24	26	48	49

63

 Table 1 – Characteristics of the studies included in the present analysis.

The fourth slip of the flexor digitorum brevis

Studies	Sample size (cad./feet)	Absence (Crude/True)	Crude prevalence	True prevalence	Variants
Turner, 1864	50 cad. / 100 feet	5 (10%)		45 (90%)	1
Wood, 1866	34 cad.	7 cad. (20.6%)	27 (79.4%)	ı	1
Wood, 1868	102 cad. / 204 feet	10 (10%) / 25 (12.2%)	92 (90.2%)	179 (87.7%)	,
Schwalbe and Pfitzner, 1889	132 feet	25 (19%)	ı	107 (81%)	78 (59%): thin slips
Schwalbe and Pfitzner, 1891	214 feet	41 (19.2%)	ı	173 (80.8%)	138(64.5%)
Schwalbe and Pfitzner, 1894	540 feet	135 (25%)	ı	405 (75%)	335 (62%): thin slips
Le Double, 1897	100 cad. / 200 feet	3 (3%) / 19 (9.5%)	87 (87%)	181 (90.5%)	,
Adachi, 1900	226 feet	42 (18.6%)		184 (81.4%)	ı
Koganei et al., 1903	292 feet	66 (22.6%)	·	226 (77.4%)	1
Adachi, 1909	141 feet	23 (16.3%)		118 (83.7%)	1
Kurz, 1923	146 feet	19 (3%)		127~(87%)	ı
Wagenseil, 1936	140 feet	36 (25.7%)		104 (74.3%)	ı
Nathaniel, 1954	60 feet	23 (38.3%)	·	37 (61.7%)	21 (35%): slip from a deep head
Mori, 1964	50 feet	8 (16%)	·	42 (84%)	ı
Nathan and Gloobe, 1974	100 feet	23 (23%)		77 (77%)	Additional slip to 5th (20%) or to 4th-5th toes (3%); origin from FDL (5%), intermuscular septum (1%), tibialis posterior (1%)
Chaney et al., 1996	284 feet	181 (63.7%)		103 (36.2%)	ı
Kura et al., 1997	11 feet	4 (36.4%)	ı	7 (63.6%)	ı
Yalçin and Ozan, 2005 (*)	33 feet	6 (18.2%)	ı	27 (81.8%)	12 (36%): thin slips; 1 (3%) separate belly; 1 (3%) slip from intermuscular septum

Table 2 – Prevalence of the fourth slip of flexor digitorum brevis (FDB-5).

Studies	Sample size (cad./feet)	Absence (Crude/True)	Crude prevalence	True prevalence	Variants
Becerro de Bengoa Vallejo et al., 2008	45 feet	3 (6.7%)	ı	42 (93.3%)	ı
Lobo et al., 2008	30 cad./60 feet	60 (100%)	0	0	I
Locke et al., 2010	4 feet	1 (25%)	ı	3 (75%)	I
Gugapriya 2012	15 cad. / 30 feet	12 (80%) / 25 (83.3%)	3 (20%)	5 (16.7%)	1 (3.3%) separate belly; 1 (3.3%) slip from intermuscular septum
llayperuma 2012	135 cad. / 270 feet	97 (71.8%) / 194 (71.8%)	38 (28.1%)	76 (28.2%)	ı
Bernhard et al., 2013	57 cad. / 97 feet	47 (48%)	28 (49%)	50 (52%)	25 (26%): thin slips
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Cad.: cadavers, IMS: intermuscular septum, TP: tibialis posterior, FDB: flexor digitorum brevis, FDL: flexor digitorum longus. (*) Microscope aided study.

Primary outcomes

True prevalence of FDB-5 absence

Twenty-two studies reported the true prevalence of FDB-5 absence (Turner, 1865; Wood, 1868; Schwalbe and Pfitzner, 1889; Schwalbe and Pfitzner, 1891; Le Double, 1897; Adachi, 1900, 1909 Koganei et al., 1903; Kurz, 1923; Wagenseil, 1936; Nathaniel, 1954; Mori, 1964; Nathan and Gloobe, 1974; Chaney et al., 1996; Kura et al., 1997; Yalcin and Ozan, 2005; Becerro de Bengoa Vallejo et al., 2008; Lobo et al., 2008; Locke et al., 2010; Gugapriya, 2012; Ilayperuma, 2012; Bernhard et al., 2013) with a total of 2789 feet and a PPE of 31.3% (95% CI = 0.208 to 0.427, $I^2 = 97.4\%$).

Crude prevalence of FDB-5 absence

Six studies reported the crude prevalence of FDB-5 absence (Wood, 1866, 1868; Le Double, 1897; Lobo et al., 2008; Gugapriya 2012; Ilayperuma 2012) with a total of 416 cadavers and a PPE of 47% (95% CI = 0.176to 0.773; $I^2 = 97.5\%$).

Secondary outcomes

Bilateral prevalence of FDB-5 absence

Eight studies reported the bilateral prevalence of FDB-5 absence (Wood, 1868; Schwalbe and Pfitzner, 1894; Le Double, 1897; Adachi, 1900, 1909 Lobo et al., 2008; Gugapriya, 2012; Ilayperuma, 2012) with a total of 776 cadavers and a PPE of 38.2% (95% CI = 0.174 to 0.614, I² = 97.6%).

Side-based prevalence of FDB-5 absence

Six studies reported the side of FDB-5 absence (Wood, 1868; Adachi, 1900, 1909 LeDouble, 1897; Lobo et al., 2008; Bernhard et al., 2013) with a total of 469 right and 469 left hands. The pooled OR of 1.1 yielded no significance between hand side (95% CI = 0.766 to 1.602, $I^2 = 0\%$, p = 0.5).

Gender-based prevalence of FDB-5 absence

Seven studies reported such occurrence (Wood, 1868; Schwalbe and Pfitzner, 1894; Le Double, 1897; Adachi, 1900, 1909 Lobo et al., 2008; Bernhard et al., 2013) with a total of 790 males and 406 females; the pooled OR result showed that FDB-5 was absent 1.5 times more in females than in males (95% CI = 1.196 to 2.099, $I^2 = 0\%$, p = 0.002).

Ancestry-based prevalence of FDB-5 absence

Four studies reported their results in Japanese populations (Adachi, 1900, 1909 Koganei et al., 1903; Mori, 1964) and two studies in Chinese populations (Kurz, 1923; Wagenseil, 1936). The total pooled sample in both populations was 995 feet with a PPE of 19.1% (95% CI = 0.156 to 0.229, $I^2 = 52.3\%$). No significant differences were found between the pooled prevalence values of both populations.

Four studies reported their results in Indian populations (Nathaniel, 1954; Lobo et al., 2008; Gugapriya 2012; Ilayperuma 2012) with a total of 420 feet and a PPE of 77.7% (95% CI = 0.489 to 0.965, $I^2 = 96.6\%$).

Ten studies reported their results in American/European populations (Turner, 1865; Wood, 1868; Schwalbe and Pfitzner, 1889, 1891; Le Double, 1897; Nathan and

Gloobe, 1974; Chaney et al., 1996; Kura et al., 1997; Becerro de Bengoa Vallejo et al., 2008; Locke et al., 2010) with a total of 1244 feet and a PPE of 21.2% (95% CI = 0.097 to 0.356, $I^2 = 96.6\%$).

Only one study reported the frequency of FDB-5 in a Turkish population (Yalcin and Ozan, 2005) with an absence rate of 18.2%.

The two independent proportion test yielded a difference of $\approx 45\%$ (p <0.0001) between Indians and all other studied populations.

True prevalence of thin slips

Four studies reported the occurrence of a thin slip of FDB-5 in their samples (Schwalbe and Pfitzner, 1889, 1891; Yalcin and Ozan, 2005; Bernhard et al., 2013) with a total of 476 feet and a PPE of 47.7% (95% CI = 0.294 to 0.662, I² = 93.5%).

True prevalence of the variations in the origin of FDB-5

Four studies reported the variation in the origin of the muscle (Nathaniel, 1954; Nathan and Gloobe, 1974; Yalcin and Ozan, 2005; Gugapriya 2012) with a total 223 feet and a PPE of 12.7% (95% CI = 0.028 to 0.283, $I^2 = 87.7\%$). The observed variations include a separate belly from FDB, a slip from flexor digitorum longus, and a slip from the intermuscular septum.

Discussion

Summary of main findings

Sarrafian and Kelikian (2012) reported an average absentee true rate of FDB-5 of 21.5%. However, their calculation was done as a mean value from five studies where one value, that of Le Double (1897), was in fact a crude, not a true frequency value. Our pooled results yielded higher prevalence values; the true and crude prevalence of the agenesis of FDB-5 were of 31.3% and 47%, respectively. A thin slip was found in half cases; meaning that only in about 1 foot out of 5 (20%) the slip for FDB-5 was considered by authors as having a "normal" size. Bilateral agenesis of FDB-5 was found to be high, 38.2%. While no significance was found for hand side, FDB-5 was 1.5 times significantly less present in females than in males. Ancestry-based results demonstrated that Indian populations have significantly higher rate (\approx 77%) of FDB-5 absence when compared to Japanese/Chinese, American/European and Turkish populations (\approx 20%). This review reveals that there is a genetic basis for the involution of the fourth slip of the FDB muscle. While the variation of the distal insertion of FDB-5 was exceptionally reported, four studies reported the prevalence of variations in its proximal origin to be \approx 13%.

Limitations and possible bias

Despite an extensive search strategy, no confirmation could be provided that this review located all relevant articles. However, the pooled sample sizes of 2789 feet and 416 cadavers used in meta-analyses could be fairly considered as representative to draw prevalence estimates for the agenesis of the fourth slip of the FDB muscle. On

the other hand, due to the lack of available reported data, our analyses did not cover all populations, mainly the African and Middle Eastern ancestries. Future research should be oriented to investigate the frequency of fourth slip to the little toe in those populations.

In conclusion, this evidence-based anatomical review attempted to provide an accurate frequency of the fourth slip of the FDB muscle and its variants in the human foot. Surgeons should be aware of the high rates of agenesis and smaller size of this slip when considering it for tendon repair, tendon transfer, or flaps soft tissue reconstruction.

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Conflict of interest: none to declare.

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