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REVIEW

The prevalence of extensor digitorum brevis manus and its variants in humans: a systematic review and meta-analysis

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Abstract Extensor digitorum brevis manus (EDBM) is a rare variant extensor muscle of the dorsum of the hand, which constitutes a diagnostic challenge in clinical practice. The aims of the review are to provide a better estimate of the frequency of EDBM and its association with variables such as ancestry, gender, laterality and side. Twenty-six studies met the inclusion criteria. The pooled rates of the meta-analyses yielded the following values: (a) an overall crude cadaveric prevalence of 4 %, (b) an overall true cadaveric prevalence of 2.5 %, (c) a true cadaveric prevalence of 2.6 % in European ancestry, (d) a true cadaveric prevalence of 2.3 % in Asian ancestry (2.07 % in Japanese and 4.2 % in Indian), (e) a bilateral occurrence in 26.3 %. Non-significant association was found between EDBM presence and ancestry, gender or side. The EDBM muscle was inserted on the index in 77 % of cases and on the long finger in the remaining 23 %. This is the first evidence-based anatomical review, which addresses the frequency of EDBM in humans.

Keywords Extensor digitorum brevis manus · Extensor indicis brevis · Anatomy · Evidence-based medicine · Hand

Introduction

Several variations of the extensors of the dorsum of the hand have been reported in the literature. Extensor digitorum brevis manus (EDBM) is one of those which presents a diagnostic challenge. The first description goes back to Bernhard Siegfried Albinus in 1758 with the appellation of "extensor brevis digiti indicis vel medii" [1]. Many anatomical terms have been used since; it has also been called the "m. extensor anomalus" [42] and "le muscle manieux" [27]; the latter was based on an analogy between the "extensor digitorum brevis" of the foot, named "le muscle pédieux" ("pied" meaning foot in French), and the extensor digitorum brevis of the hand named "le muscle manieux" (main meaning hand in French). Depending on where it inserted, EDBM has also been named extensor indicis brevis [3, 10], extensor digiti III brevis [22], extensor medii brevis, extensor medii and annular brevis [11]. However, after Macalister [28], coined it as "extensor digitorum brevis manus" many authors have used this term in later years.

Embryology and anatomy

In the human embryo, the precursor extensor muscle of the forearm differentiates into three parts; radial, superficial and deep. The deep portion, which is innervated by the posterior interosseous nerve, gives rise to the abductor pollicis longus and the extensor pollicis brevis on the radial side and the extensor pollicis longus and the extensor indicis proprius (EIP) on the ulnar side [48]. While comparative anatomy studies showed a marked stability of the superficial portion, the deep portion has undergone considerable evolutionary change and appears to be highly variable, as is observed by the significant variation in its expression in different species of primates [54]. Therefore, the EDBM may have been developed in the deep unstable part of the forearm extensor muscle mass where most of the variations occur. Some authors considered that EDBM is an atavistic muscle which may represent a failure of proximal migration of ulnocarpal elements of the extensor muscle mass in

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humans [2, 43, 46]. In fact, this muscle is present in nearly all amphibians where it primarily controls the extension of the fingers of the forelimb [56].

The distal insertion of EDBM and EIP is often joined and the two muscles share the same blood supply via the posterior branch of the anterior interosseous artery, and the same innervation via the posterior interosseous branch. It is the reason why some consider EDBM as an extrinsic muscle, a variant of EIP and naming it "extensor indicis brevis" [32, 49]. Furthermore, Ogura et al. found that EDBM muscle was present in 10 out of 20 (50.0 %) human upper limbs in which the extensor indicis proprius muscle did not occur, and only in 5 out of 534 (0.9 %) upper limbs in which that muscle occurred [32]. Therefore, it seems that the EDBM muscle compensates for the EIP muscle in man [34, 55]. In such a case, the EDBM muscle is the only muscle responsible for independent extension of the index finger [35].

Anatomical variations of EDBM

Usually the EDBM muscle takes origin from the dorsal aspect of the wrist joint capsule, the distal end of the radius, the dorsal metacarpal surface, or from the proximal portion of the radiocarpal ligament in the area of the fourth compartment of the extensor digitorum communis [4, 10, 12, 34, 39, 55]. The EDBM muscle could have up to four tendons; the variant with a single tendon to the index or to the middle finger is the most frequent [27].

Variations of EDBM were categorized by Ogura et al. [34] and Yoshida [55] with regard to its distal insertion; the former has been the most commonly used. The classification of Ogura et al. [34] was mainly based on the relationship of EDBM with the EIP. Type I: insertion of EDBM on the dorsal aponeurosis of the index where the EIP was absent. Type II: both EDBM and EIP were inserted on the index; type IIa is where a vestigial EIP was confluent with the EDBM belly and inserted on the index, type IIb is where the distal end of EDBM was joined to the EIP tendon and type IIc is where EDBM was inserted on the ulnar side of EIP tendon. Type III is where the EDBM was inserted on the long finger.

On the other hand, the anomaly could be bilateral [12, 31, 41] and less often found in familial cases [17, 33, 47].

Clinical relevance

Dunn and Evarts stated an EDBM frequency varying from 1 to 9 % [9]. However, the incidence of EDBM in cadaveric studies has been reported with a range between 2–4 % [22, 25, 34, 39, 55]. Surprisingly, few clinical cases have been reported out of more than 300 clinical and cadaveric case reports [7]; it is likely that the condition is usually symptomless. Hayashi et al. [15] coined the term "The fourth-compartment syndrome" to describe chronic dorsal wrist pain with possible five causes; the presence of EDBM has been considered as one etiology. When present, EDBM might cause a painful tumefaction which could easily be mistaken with other dorsal wrist pathology such as synovial cyst and lipoma [34, 36, 40, 44]. Sometimes, a ganglion could be associated to or embedded in an EDBM which adds to the diagnostic difficulty [8, 34]; 17 ganglions were found to be associated with the 68 cases of EDBM reported by Ogura et al. [34]. Based on a review of previous clinical cases, manual work and hand dominance have been incriminated as potential factors in the clinical expression of EDBM [42]. Gama randomly examined 3,404 adults and found 38 cases of EDBM for an incidence of 1.1 %; 19 (50 %) required surgery because of pain precluding work [13]. Out of 29 patients seen by Ogura et al. [34], 5 (17.2 %) requested surgery. Pain may be produced by asking the patient to push the palm against the table with the wrist in an extension position [13, 37].

Usually discovered during surgery [6, 33], ultrasound and MR scans can be of great assistance to clinicians when the diagnosis is suspected on physical examination [30, 34]. However, even magnetic resonance imaging can be of no help if the anomaly is not known or looked for by investigators [12]. The statement made by Reef and Brestin [40] less than three decades ago seems to stand still: "The most consistent single finding was the inability of the surgeon to make a correct preoperative diagnosis". EDBM could be one of those conditions where one should think of it to make the diagnosis; it should be included in the differential diagnosis of "dorsal ganglions".

Conservative treatments such as paraffin baths, diathermy, immobilization, anti-inflammatory medication, and botulinum toxin have been used with limited success [22, 40, 42, 52]. A surgical treatment is often needed. Division of the extensor retinaculum is more likely to offer good results in the case of muscle hypertrophy [37, 42], whereas total ablation of the atavistic muscle usually yields complete recovery of the symptoms [12, 26].

It is worthy to note that, when present EDBM could be used as a transfer tendon to restore extension function [38].

Methods

Search strategy and identification of studies

A systematic literature search was conducted through a number of electronic databases such as Medline, Embase, Scielo, AMED, AUSPORT, SPORTDiscus, and the Cochrane library from inception to Nov 2013, using the Boolean combination of broad terms such as "(extensor* AND tendon* AND hand AND brevis)" to locate the maximum number of relevant articles. We also searched the following journals: *Acta Anatomica*, *Annals of Anatomy*, *Clinical Anatomy*, *European Journal of Morphology*, *Folia Morphologica*, *Journal of Anatomy*, *Journal of hand Surgery* [*Br and Am*], *Journal Bone and Joint Surgery*[*Br and Am*], *Journal of Morphology*, *Surgical and Radiological Anatomy*, *The Anatomical Record* (*A and B*). 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 EDBM is scant, therefore all published or unpublished studies reporting prevalence rates were included in the review. However, case reports or prevalence ranges cited in textbooks were excluded. The primary outcome is the overall crude or true prevalence rate of EDBM in cadaveric, clinical or radiological studies. The crude EDBM prevalence is the number of individuals who have either one or two EDBM compared to the number of individuals available for study. The true EDBM prevalence rate is the number of hands affected compared to the number of hands available for study.

Secondary outcomes are the prevalence in relation to ancestry, gender, laterality and side, the interactions between those variables, and the variation types of EDBM. To ensure unbiased selection of included studies, abstracts from conferences were not included. However, unpublished but localizable data from articles or theses were included, and unpublished data from published articles treating the anatomy of the extensors of the hand were also considered; for the latter, the corresponding authors were contacted; whenever EDBM prevalence rate was calculated but not reported, the result had to be communicated via email for documentation.

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

Relevant data extracted included sample size, sample details, type of investigation (clinical or cadaveric), and the results. Analysis was performed using StatsDirect v2.7.8 (Altrincham, United Kingdom). Proportion meta-analysis (MA) 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 EDBM frequencies in different ancestry populations. Descriptive analysis was conducted when the data were not amenable to meta-analysis. We examined heterogeneity amongst studies using I^2 statistics; whenever $I^2 < 50$ %, the fixed-effect estimate was reported. When possible, sensitivity analysis was conducted by limiting inclusion to studies with sample size ≥100.

Results

Search results

The search strategy yielded a total of 516 studies; 24 duplicates were removed. Case reports constituted the majority of the excluded studies with a total of 301 papers. Seven papers were case reports with narrative reviews. One hundred fifty-eight papers were not related to the EDBM muscle. Finally, 26 studies reporting one of the predefined outcomes were included [3, 5, 10, 13, 14, 16, 18–20, 22–25, 28–30, 34, 39, 45, 49–51, 53, 55–57]. Four were located by searching cited references [14, 16, 24, 50]. All studies are cadaveric but one, that of Gama [13] is a clinical study. One study comprised fetuses [23], whereas all others were conducted on adults (Table 1). Only Wagenseil’s study comprised two sub-studies [51].

EDBM frequencies in individual studies are given in Table 2.

Crude overall cadaveric prevalence of EDBM

Seventeen studies reported the crude cadaveric prevalence rate of EDBM [3, 5, 14, 19, 20, 25, 26, 28–30, 34, 39, 41, 50–52, 55] with a total of 1,867 cadavers and a PPE of 4.0 % (95 % CI 0.032–0.049, $I^2 = 0$ %). A sensitivity analysis was conducted on eight large-sampled ($n \geq 100$ cadavers) studies [3, 23, 25, 29, 34, 41, 53, 55] with a total of 1,326 cadavers and a PPE of 4.3 % (95 % CI 0.033–0.055, $I^2 = 0$ %).

True overall cadaveric prevalence of EDBM

Twenty-five studies reported the true cadaveric prevalence rate of EDBM [3, 5, 10, 14, 16, 18–20, 22–25, 28–30, 34, 39, 41, 45, 50, 51, 53, 55–57] with a total of 5,789 hands and a PPE of 2.3 % (95 % CI 0.017–0.029, $I^2 = 46.4$ %). A sensitivity analysis was conducted on 18 large-sampled ($n \geq 100$ hands) studies [3, 5, 16, 18, 19, 22–25, 28, 29, 33, 40, 49, 51, 54–56] with a total of 5,398 hands and a PPE 2.51 % (95 % CI 0.020–0.030, $I^2 = 0$ %).

Interaction with ancestry

Eleven studies reported the true cadaveric prevalence rate of EDBM in European populations [3, 14, 22, 28, 29, 41,

Table 1 Characteristics of the included studies and EDBM prevalence

Studies	Population	Age	Sample size: cadavers	Sample size: hands
Cauldwell et al. [3]	British	Adults	140	243
Dass et al. [5]	Indian	Adults	–	100: 47 R, 53 L
El-Badawi et al. [10]	Saudi	Adults	–	181
Gama [13]	Brazilian	Adults	3,404 Living subjects	–
Godwin and Ellis [14]	British	Adults	25	50
Hirai et al. [16]	Japanese	Adults	–	548: 276 R, 272 L
Inoue [18]	Japanese	Adults	50	100
Jacobina et al. [19]	Brazilian	Adults	86	172: 86 R, 86 L
Jadhav et al. [20]	Indian	Adults	48	96: 48 R, 48 L
Kadanoff [22]	German	Adults?	–	300
Koh [23]	Japanese	Fetuses	100	200
Komiyama et al. [24]	Japanese	Adults	78 Pairs + 8 unpaired limbs	164
Kosugi et al. [25]	Japanese	Adults	193: 90 M, 103 F	375: 180 R, 195 L
Macalister [28]	British	Adults?	15	30
McGregor [29]	South African (Bantu)	Adults	100	200
Moriya [30]	Japanese	Adults	82	164
Ogura et al. [34]	Japanese	Adults	286: 158 M, 128 F	559: 282 R, 277 L
Ranade et al. [39]	Indian	Adults	36: 18 M, 18 F	72
Rodríguez-Niedenführ et al. [41]	Spanish	Adults	128: 59 M, 69 F	256: 128 R, 128 L
Smith [45]	British	Adults	–	50
von Schroeder and Botte [50]	American	Adults	–	43
Wagenseil [51]	Chinese	Adults	75	131
Wagenseil [51]	German	Adults	–	124
Wood [53]	British	Adults	102: 68 M, 34 F	204: 102 R, 102 L
Yoshida et al. [55]	Japanese	Adults	277	554
Yoshida [56]	Japanese	Adults	416: 217 M, 199 F	832
Zilber and Oberlin [57]	French	Adults	–	50

EDBM extensor digitorum brevis manus, *M* male, *F* female, *R* right, *L* left

45, 50, 51, 53, 57] with a total of 1,550 hands and a PPE of 2.6 % (95 % CI 0.016–0.036, $I^2 = 32$ %).

Thirteen studies reported the true cadaveric prevalence rate of EDBM in Asian populations [5, 16, 18, 20, 23–25, 30, 34, 39, 51, 55, 56] with a total of 3,895 hands and a PPE of 2.3 % (95 % CI 0.015–0.031, $I^2 = 59.5$ %). Subgroup analyses revealed the following: nine studies reported the true cadaveric prevalence rate of EDBM in Japanese populations [16, 18, 23–25, 30, 34, 55, 56] with a total of 3,496 hands and a PPE of 2.07 % (95 % CI 0.020–0.032, $I^2 = 68.5$ %), and three studies reported the true cadaveric prevalence rate of EDBM in Indian populations [5, 20, 39] with a total of 268 hands and a PPE of 4.2 % (95 % CI 0.021–0.069, $I^2 = 0$ %). A two-independent proportion test yielded non-significance between both ethnicities ($p = 0.08$). Another two-independent proportion test yielded non-significance between European and Asian populations ($p = 0.3$). One study [51] reported the true cadaveric prevalence of EDBM in a Chinese population; 2 EDBM out of 131 hands (1.52 %).

One study reported the true cadaveric prevalence rate of EDBM in a South African population of the Bantu tribe [29], which was 1.5 % (3 out of 200 hands). Two Brazilian studies reported EDBM prevalence in Brazilian populations; one cadaveric [41] showing a true prevalence of 1.1 % (2 out of 172 hands) and one clinical [13] reporting a crude prevalence of 1.1 % (38 out of 3,404 subjects).

Interaction with gender

Five studies reported the gender distribution of their samples along with gender frequencies [25, 34, 39, 41, 53] with a total of 393 males and 351 females yielding a pooled OR of 0.98 (CI 0.499–1.941, $p = 0.9$, $I^2 = 0$ %). Therefore, no statistically significant gender-based difference could be found.

Interaction with side

Eight studies reported the side distribution of their samples along with side frequencies [5, 19, 20, 25, 34, 39, 41,

Table 2 Prevalence of EDBM

Studies	Sample size: cadavers	Crude Nb (crude EDBM prevalence)	Bilateral prevalence	Sample size: hands	True Nb (true EDBM prevalence)
Cauldwell et al. [3]	140	5 (3.6 %)	1 (20 %)	243	6 (2.5 %)
Dass et al. [5]	–	–	–	100	3 L (3 %)
El-Badawi et al. [10]				181	2 (1.1 %)
Gama [13]	3,404 Living subjects	38 (1.1 %)	–	–	–
Godwin and Ellis [14]	25	2 (8 %)	–	50	2 (4 %)
Hirai et al. [16]	–	–	–	548	1 (0.2 %)
Inoue [18]	50	2 (4 %)	0	100	2 (2 %)
Jacobina et al. [19]	86	1 (1.1 %)	1 (100 %)	172	2 (1.1 %): 1 L, 1 R
Jadhav et al. [20]	–	–	–	96	4 (4.2 %): 2 R, 2 L
Kadanoff [22]	–	–	–	300	10 (3.3 %)
Koh [23]	100	4 (4 %)	0	200	4 (2 %)
Komiyama et al. [24]	–	–	–	164	2 (1.2 %): 1 R, 1 L
Kosugi et al. [25]	193	9 (4.7 %): 4 M, 5 F	3 (33.3 %)	375	12 (3.2 %): 5 R, 7 L
Macalister [28]	15	1 (6.7 %)	0	30	1 (3.3 %)
McGregor [29]	100	3 (3 %)	0	200	3 (1.5 %)
Moriya [30]	82	2 (2.4 %)	1 (50 %)	164	3 (1.8 %)
Ogura et al. [34]	286	11 (3.8 %): 4 M, 7 F	6 (54.5 %)	559	17 (3 %): 10 R, 7 L
Ranade et al. [39]	36	3 M (8.3 %)	0	72	3 L (4.2 %)
Rodríguez-Niedenführ et al. [41]	128	3 (2.3 %): 2 M, 1F	1 (33.3 %)	256	4 (1.6 %): 2 R, 2 L
Smith [45]	–	–	–	50	0
von Schroeder and Botte [50]	–	–	–	43	0
Wagenseil [51]	75	2 (2.6 %)	0	131	2 (1.5 %)
Wagenseil [51]	–	–	–	124	2 (1.6 %)
Wood [53]	102	8	–	204	12 (4.9 %): 8 M, 4F
Yoshida et al. [55]	277	12 (4.3 %): 7 M, 5 F	3 (25 %)	554	15 (2.7 %): 7 R, 8 L
Yoshida [56]	416	–	–	832	18 (2.2 %)
Zilber and Oberlin [57]	–	–	–	50	0

EDBM extensor digitorum brevis manus, *M* male, *F* female, *R* right, *L* left

55] with a total of 1,084 right and 1,100 left hands with a pooled OR of 0.89 ($p = 0.7$, $I^2 = 0$ %). Therefore, no statistically significant side-based difference could be found.

Interaction with laterality

Fifteen studies reported the laterality prevalence of EDBM [3, 5, 18–20, 22, 26, 28–30, 34, 39, 41, 51, 55] with a total of 1,690 cadavers (Table 2); 16 out of 62 EDBM were bilateral with a pooled rate of 26.3 % (95 % CI 0.171–0.366, $I^2 = 11.2$ %).

Variant types of EDBM

Full anatomical details of the EDBM were found in 11 studies [3, 5, 20, 21, 26, 29, 34, 39, 41, 55, 56]. We attempted to assign each of the distal insertion variation of EDBM to one of the types described in the classification of Ogura et al. [34]. Out of 87 detailed variants of EDBM, 27 (31 %) were of type I, 4 (4.6 %) of type IIa, 13 (15 %) of type IIb, 23 (26.4 %) of type IIc, and 20 (23 %) of type III (Table 3).

Discussion

Summary of main findings

Our results showed that EDBM was present in nearly 4 % of the dissected cadavers and in nearly 2.5 % of the dissected hands. Where such values were in accordance with the rates reported in the two Japanese large-sampled studies of the review [34, 55], it is likely that our pooled estimates reflect the global EDBM frequency more accurately, while taking into account different ancestral populations.

Pooled results showed no association with variables such as ancestry, gender and side. They also showed a bilateral occurrence of EDBM in 26 % of cases. This would suggest

Table 3 Prevalence of anatomic types of EDBM (based on Ogura et al.'s classification)

	Type I	Type IIa	Type IIb	Type IIc	Type III	Total
Caldwell et al. [3]		0	2	3	1	6
Dass et al. [5]	0	0	0	3	0	3
Jacobina et al. [19]	0	0	0	0	2	2
Jadhav et al. [20]	0	0	0	4	0	4
McGregor [29]	0	0	0	3	0	3
Kosugi et al. [25]	2	1	2	3	4	12
Ogura et al. [34]	4	3	2	3	5	17
Ranade et al. [39]	0	0	0	0	3	3
Rodríguez-Niedenführ et al. [41]	0	0	0	1	3	4
Yoshida et al. [55]	10	0	2	2	1	15
Yoshida [56]	11	0	5	1	1	18
Total	27 (31 %)	4 (4.6 %)	13 (15 %)	23 (26.4 %)	20 (23 %)	87

that the presence of EDBM could have a genetic basis. Additionally, while conducting the review, we found some reports demonstrating the occurrence of familial EDBM. In contrast, clinical case reports showed that painful EDBM is more encountered in manual workers and on the dominant hand.

We found that EDBM inserted much more on the index (types I and II) than onto the medius (type III): 77 versus 23 %. This is in accordance with the majority of publications. On the other hand, when EIP was absent as in type I, EDBM had the highest frequency when compared to subtypes II and type III where the EIP was present. This finding would favor the theory of extension substitution by the EDBM muscle in the case of EIP agenesis.

Potential limitation and bias

Despite an extensive search strategy, no confirmation could be provided that this review located all relevant articles. However, the pooled samples of 1,867 cadavers and 5,989 hands could be fairly considered as representatives to draw prevalence estimates for this rare condition. On the other hand, we were able to compare ancestry-related associations only between few groups or subgroups of ethnicities; European versus Asian and Japanese versus Indian populations. We could locate only few single studies reporting EDBM rates in other ancestries, which were not amenable to MA. Another possible limitation could be that the five largest sample-sized studies were all Japanese [24, 25, 34, 55, 56]. This could introduce bias to our results related to ancestry association; however, MA is usually used to counter such bias.

In conclusion, this evidence-based anatomical review attempted to provide a more accurate frequency of EDBM in humans. Pooled estimates were provided for the global overall prevalence and those of some ancestral subgroups such as European, Asian, Japanese and Indian populations as well. Our results corroborated the fact that EDBM prevalence is not associated with ancestry, gender or side. Including EDBM presence as a differential diagnosis of dorsal wrist tumefaction is of major importance in clinical practice; the awareness of its prevalence would be an added value for hand surgeons when facing this diagnostic challenge.

Conflict of interest The authors declare that they have no conflict of interest.

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