This is a provisional PDF only. Copyedited and fully formatted version will be made available soon.



ISSN: 0015-5659 e-ISSN: 1644-3284

Novel, bilateral, two-bellied muscles span the extensor forearm, thenar eminence to insert on the proximal phalanx of the thumb: clinical and embryological significance

Authors: Priti L. Mishall, Amber N. Marsh, Diana Perez, Xavier H. Quezada, Maximilian C. Stahl, Rachel E. Weinstock, Sherry A. Downie

DOI: 10.5603/FM.a2019.0067

Article type: CASE REPORTS

Submitted: 2019-03-05

Accepted: 2019-05-29

Published online: 2019-06-04

This article has been peer reviewed and published immediately upon acceptance. It is an open access article, which means that it can be downloaded, printed, and distributed freely, provided the work is properly cited. Articles in "Folia Morphologica" are listed in PubMed. Novel, bilateral, two-bellied muscles span the extensor forearm, thenar eminence to insert on the proximal phalanx of the thumb: clinical and embryological significance Running Head: Bilateral, novel muscles, forearm and hand

Priti L. Mishall¹, Amber N. Marsh¹, Diana Perez¹, Xavier H. Quezada¹, Maximilian C. Stahl, Rachel E. Weinstock¹, Sherry A. Downie²

¹Albert Einstein College of Medicine, Department of Anatomy and Structural Biology, Bronx, NY, United States ²Department of Anatomy and Structural Biology and Department of Physical Medicine and Rehabilitation, Bronx, NY, United States

Address for correspondence: Priti L. Mishall, MD, PGCertME, MBBS, Albert Einstein College of Medicine, Department of Anatomy and Structural Biology and Department of Ophthalmology and Visual Sciences, 1300 Morris Park Ave, F620SD, Bronx, NY 10461, United States, tel: 718 430 3423, e-mail: priti.mishall@einstein.yu.edu

Abstract

Muscle and tendon variations in the forearm, wrist and hand are commonly reported in the anatomical and surgical literature. They are frequently the source of inflammatory conditions such as de Quervain's tenosynovitis or carpal tunnel syndrome. During academic dissection, a cadaver presented with bilateral, additional muscles running parallel to the abductor pollicis longus muscles (APL) in the extensor compartment of the forearm. Both additional muscles had two bellies, one proximal and one distal, with an intervening tendon. The proximal bellies were separate and distinct from the adjacent APLs. The tendons traversed the first dorsal compartments with the tendons of the APLs and the extensor pollicis brevis muscles (EPB). The distal bellies lay adjacent to the abductor pollicis brevis (APB) muscles in the thenar compartments, and inserted onto the volar base of the proximal phalanges of the thumbs. Following a thorough search of the literature, we determined that these additional muscles constitute a previously unreported variation. This report details the variation, compares it with other reported variations, presents the related embryology, and reviews the significance of this variation as it relates to inflammatory conditions and surgical procedures.

Key words: novel muscles, extensor compartment forearm, de Quervain's syndrome

INTRODUCTION

Muscle and tendon variations in the forearm, wrist and hand are commonly reported. They are also frequently the source of inflammatory conditions, such as de Quervain's tenosynovitis [12] or carpal tunnel syndrome [10] that may cause dysfunction and require surgical intervention. Full exploration of anatomical variations identified during a surgical procedure is not usually feasible. However, variations found during academic dissection can be fully characterized and added to the body of information in the anatomical and surgical literature.

Reported variations in the extensor forearm include accessory heads of muscles (extensor carpi radialis longus and brevis [23, 24] and extensor indicis [35]), a wide variety of tendon anomalies (extensor digitorum [31], extensor indicis [11] abductor pollicis longus [1, 2, 6, 8, 26, 27, 30, 34]), the absence of muscles [33], and even the presence of novel muscles [16, 17].

Many of the muscles in the extensor forearm serve important functions in the hand such as moving the thumb or controlling position of the wrist to optimize grip. Variations in these muscles or their tendons can lead to inflammatory conditions that negatively impact function of the hand.

During academic dissection, a digastric muscle was identified bilaterally. It originated in the extensor forearm adjacent to the APL and terminated in the thenar compartment by attaching to the base of the proximal phalanx of the thumb. The intertendon that connected the two bellies passed through the first dorsal compartment with the APL and EPB tendons. The other muscles in the forearms exhibited normal morphology as described in contemporary textbooks [22]. Therefore, this finding represents a novel bilateral muscle. The present case report describes the finding, reviews the potential developmental basis of the novel muscle, and considers its potential clinical and surgical implications.

CASE REPORT

During routine dissection of an 87-year-old male cadaver at the Albert Einstein College of Medicine, Bronx, NY, bilateral additional muscles, running parallel to typical APL muscles, were identified in the extensor compartment of the forearm. The additional muscles had two bellies, one proximal and one distal, connected with intermediate tendons. Both right and left proximal bellies were attached to the dorsal side of the radius and adjacent interosseous membrane, lateral and parallel to the origin of the respective APL. The right proximal belly extended across the interosseous membrane and attached to the radial side of the ulna at the level of the distal border of the supinator muscle (Figure 1A). The left proximal belly was somewhat smaller and attached only to the radius and adjacent interosseous membrane (Figure 2A). The distal muscle bellies were located in the thenar compartment, radial and parallel to typical APBs. Each had a small area of muscle fiber interdigitation with the lateral fibers of the respective APB. The distal attachments of the additional muscles were to the lateral aspect of the base of the proximal phalanx of each thumb. The distal belly of the right muscle was 9.84 mm in width and 39.45 mm in length. The distal belly of the left muscle was 7.14 mm in width and 35.73 mm in length. The intermediate tendons that connected the proximal and distal bellies ran parallel to the respective APL and EPB tendons, through the 1st dorsal compartment. The blood supply to the proximal and distal muscle bellies was derived from the posterior interosseous artery and radial artery respectively. The innervation of the proximal and distal bellies was derived from the posterior interosseous nerves and recurrent branches of the median nerves respectively.

In an effort to understand whether the distal bellies of the additional bilateral muscles were truly 'additional' or simply part of the typical APB, measurements were taken from the cadaver of interest, as well as eleven other male cadavers (ages 67 to 94 years) with typical thenar muscles. The right hand of one cadaver was unable to be measured because of previous dissection so the total number of muscles measured was 21. To control for the

3

different sizes of the hands of the cadavers, the ratio of width to length was used to compare muscle size rather than the width or length alone. If the additional muscle was a standalone muscle, then the ratio of the adjacent APB would be the same as the ratio of the APBs of the other cadavers. A one-tailed, one-sample T-test was performed to compare the ratio of the width and length of the right and left APB of the cadaver of interest to the ratio of the width and length of the right and left APB of the other cadavers. There was no significant difference (p=0.914). Thus, the additional muscles were indeed additional and not simply part of the APBs (Table 1).

DISCUSSION

Other novel muscles have been described in the forearm, wrist and hand [16]. Lee et al, [16] described two novel bilateral muscles, extensor digitorum brevis manus and extensor medii proprius, on the dorsum of the hand. Li and Ren [17] also reported bilateral extensor medii proprius. The true cadaveric prevalence of these muscles was reported as 2.5-4% and 0.8-10.4% respectively [16].

A novel abductor pollicis tertius (AP tertius) has been described that arises from the dorsal aspect of the radius with the APL [1]. No photograph was provided in this report, but the description of the proximal muscle sounds similar to the present case. The distal attachments, however, differed in two ways. Firstly, in the case of AP tertius, the distal muscle fused with the APB; in the present case, the distal muscle was a separate muscle. Secondly, the distal muscle belly of AP tertius inserted on the first metacarpal [1], while in the present case, the distal muscle belly inserted at the base of the proximal phalanx.

In addition to novel muscles, many variations have been reported in the literature and some have similarities to the muscles in this case report. A study by Fabrizio and Clemente [9] of 50 cadaver forearms described a variation of the APL in which an additional muscle belly (APL2) arose from the typical APL belly. In 4 cases the APL2 gave a tendon that paralleled the APL tendon [9]. This arrangement of tendons is similar to the present case, but the insertions of these four APL2 tendons were different. The most similar was one tendon that inserted onto the fascia of the APB. However, in the present case, the distal belly was distinct from the APB and inserted into the proximal phalanx.

Variations in the APL have been studied extensively [2, 15]. Baba [2] studied 143 wrists and found tendon variations in all but two cadavers. Lacey et al. [15] studied APL tendons in 38 human forearms and found seven to be normal.

More recently additional bellies of the APL have been described. In one case the authors reported bilateral APL tendons that continued beyond the extensor retinaculum as muscle bellies on the dorsolateral side of the first metacarpal. The APL tendons were accompanied by the EPB tendons to the insertion onto the base of the proximal phalanx [30]. The similarities to the present case include the presence of a distal muscle belly and sharing the first dorsal compartment with the EPB. However, in the present case, the distal belly was positioned volarly in the thenar eminence and there were three tendons in the first dorsal compartment: EPB, APL and the novel muscle.

In a second case, the authors reported a unilateral APL that split into two tendons: medial and lateral. The medial tendon inserted typically at the base of the metacarpal, while the lateral tendon gave rise to a muscle belly that inserted at the base of the proximal phalanx [27]. Again, the distal muscle belly was part of the APL. In the present case, it is part of the novel muscle. In a third case, a unilateral APL split into two bellies that each gave a tendon [36]. One tendon inserted onto the base of the first metacarpal, while the other divided into three slips that inserted into the thenar muscles. The slip to the APB "acquired muscular characteristics prior to insertion". The authors' illustrations and photograph do not show a tendon dividing into three slips and distributing to the three thenar muscles. Therefore it is difficult to understand what is meant by their description [36].

In the present case, the two-bellied, novel muscles were completely separate entities, not part of the APL muscle. The distal muscle belly was also a separate muscle that ran parallel to the APB and inserted at the base of proximal phalanx lateral to the APB insertion.

The embryological basis for variations in the limb has been well studied. Limb skeletal elements have been explored for many years [7, 19, 25, 29] but only recently has

the technology been available to observe and document the soft tissues – tendon, ligaments and muscle [3, 18].

During development, cartilage and muscle progenitors appear at about the same time at the base of the presumptive limb bud. Cartilage arises from limb mesenchyme while muscle precursors originate from the dermamyotome. Tendon progenitors also arise from limb mesenchyme but appear at a later time. Ultimately, individuated tendons and muscles connect with each other forming functional units of movement [5]. Surprisingly, myoblasts are not preprogrammed to become a specific muscle, or go to a specific place in the limb, or express a specific myofiber type. These processes of specification are regulated by signals from a unique component of the limb mesenchyme called muscle connective tissue (MCT) [4].

MCT is formed by fibroblasts of the lateral plate mesoderm. It is the connective tissue that integrates into muscle as endomysium, perimysium and epimysium. MCT regulates muscle patterning, splitting and orientation [4, 18]. Manipulation of transcription factors that affect MCT has been shown to cause muscle patterning defects, positioning defects, and orientation defects [32]. It is likely that these factors are also responsible for the extra muscles identified in the present case report.

Tendon variations, such as multiple tendons or a novel muscle with a tendon, as in the present case may predispose patients to de Quervain's stenosing tenosynovitis [14, 20]. The exact pathophysiology of de Quervain's tenosynovitis has not been elucidated, but the current thought is that overuse or overcrowding in the tendon sheath leads to irritation of the sheath and resistance to gliding of the tendons in their canal. De Quervain's presents as swelling and pain on the dorsolateral side of the wrist [21]. Although the initial management of de Quervain's tenosynovitis is nonsurgical, patients with very serious inflammation who have recurrent or chronic symptoms are treated with surgical correction [13]. Proper identification of the tendons is critical for the operational technique, as the surgery involves decompression of the osseo-fibrous canal in which the tendons lie. The importance of decompression is highlighted by the fact that incomplete release of the tendon subcompartment has been linked to long-term post-surgical complications [28]. The importance of recognition of anatomical variations in surgical correction of de Quervain's syndrome has been known for many decades, and multiple authors [2, 12] have reported the impact that accessory APL tendons have on the procedure. The findings in the present case are of equal importance because failure to recognize this muscle and its tendon could have led to incomplete tendon decompression and surgical failure.

CONCLUSIONS

This case documents a novel, bilateral muscle that arose adjacent to the APL in the extensor forearm. Its tendon occupied the first dorsal compartment along with the APL and EPB tendons. Its distal muscle belly resided in the thenar compartment, adjacent to the APB, and inserted onto the volar base of the proximal phalanx. The right and left muscles were identical in their morphology, only the left was a bit smaller than the right.

Musculoskeletal variations are common, and failure to consider and identify variations could lead to insufficient surgical relief and an eventual recurrence of inflammatory conditions and dysfunction [30]. Knowledge of this novel muscle will be interesting to the anatomist and useful for the practicing clinician.

Acknowledgements

The authors acknowledge and gratefully thank the individual whose body and tissues were used in this study for the advancement of physician education and patient care. No funding sources to report.

REFERENCES

1. Akita K, Nimura A. Bergman's Comprehensive Encyclopedia of Human Anatomic Variation. In: Tubbs RS, Shoja MM, Loukas M (eds.). Forearm muscles. Wiley Blackwell, New Jersey 2016: 298-314.

2. Baba M. The accessory tendon of the abductor pollicis longus muscle. The Anatomical Record. 1954;119(4):541-547.

3. Charles JP, Cappellari O, Spence AJ, Hutchinson JR, Wells DJ. Musculoskeletal Geometry, Muscle Architecture and Functional Specialisations of the Mouse Hindlimb. PLoS One. 2016;11(4):e0147669. doi: 10.1371/journal.pone.0147669. PubMed PMID: 27115354; PubMed Central PMCID: PMCPMC4846001.

 Chevallier A, Kieny M. On the role of the connective tissue in the patterning of the chick limb musculature. Wilhelm Roux's archives of developmental biology. 1982;191(4):277-280. doi: 10.1007/bf00848416.

5. Chevallier A, Kieny M, Mauger A. Limb-somite relationship: origin of the limb musculature. J Embryol Exp Morphol. 1977;41:245-258. PubMed PMID: 591873.

6. Dos Remedios C, Chapnikoff D, Wavreille G, Chantelot C, Migaud H, Fontaine C. The abductor pollicis longus: relation between innervation, muscle bellies and number of tendinous slips. Surgical and radiologic anatomy : SRA. 2005;27(3):243-248. doi: 10.1007/s00276-004-0286-3. PubMed PMID: 15549300.

 Downie SA, Newman SA. Morphogenetic differences between fore and hind limb precartilage mesenchyme: relation to mechanisms of skeletal pattern formation. Dev Biol. 1994;162(1):195-208. Epub 1994/03/01. doi: 10.1006/dbio.1994.1078. PubMed PMID: 8125187.

8. El-Beshbishy RA, Abdel-Hamid GA. Variations of the abductor pollicis longus tendon: an anatomic study. Folia morphologica. 2013;72(2):161-166.

 9. Fabrizio PA, Clemente FR. A variation in the organization of abductor pollicis longus. Clinical anatomy (New York, NY). 1996;9(6):371-375. doi: 10.1002/(SICI)1098-2353. PubMed PMID: 8915615.

10. Franklin GM, Evanoff B. Treatment of carpal tunnel syndrome: Surgery or more conservative management? Muscle Nerve. 2019. doi: 10.1002/mus.26502. PubMed PMID: 31049992.

11. Georgiev GP, Tubbs RS, Iliev A, Kotov G, Landzhov B. Extensor indicis proprius muscle and its variants together with the extensor digitorum brevis manus muscle: a common classification. Clinical significance in hand and reconstructive surgery. Surgical and radiologic anatomy : SRA. 2018;40(3):271-280. doi: 10.1007/s00276-018-1981-9. PubMed PMID: 29383419.

Giles KW. Anatomical Variations Affecting the Surgery of De Quervain's Disease.
 The Bone & Joint Journal. 1960;42 B(2):352-355.

 Huisstede BM, Coert JH, Friden J, Hoogvliet P, European HG. Consensus on a multidisciplinary treatment guideline for de Quervain disease: results from the European HANDGUIDE study. Phys Ther. 2014;94(8):1095-1110. doi: 10.2522/ptj.20130069.
 PubMed PMID: 24700135.

14. Kulthanan T, Chareonwat B. Variations in abductor pollicis longus and extensor pollicis brevis tendons in the Quervain syndrome: a surgical and anatomical study. Scand J Plast Reconstr Surg Hand Surg. 2007;41(1):36-38. PubMed PMID: 17484184.

15. Lacey T, Goldstein L, Tobin C. Anatomical and clinical study of the variations in the insertions of the abductor pollicis longus tendon, associated with stenosing tendovaginitis. The Journal of Bone & Joint Surgery. 1951;33(2):347 - 350.

16. Lee DH, Lee JH, Woo RS, Song DY, Baik TK, Yoo HI. A rare bilateral variation on the dorsum of the hand: extensor digitorum brevis manus and extensor medii proprius.
Anat Cell Biol. 2019;52(1):97-99. doi: 10.5115/acb.2019.52.1.97. PubMed PMID: 30984461; PubMed Central PMCID: PMCPMC6449591.

17. Li J, Ren ZF. Bilateral extensor medii proprius with split tendon of extensor indicis proprius, a rare anatomical variant. Romanian journal of morphology and embryology = Revue roumaine de morphologie et embryologie. 2013;54(3):639-641. PubMed PMID: 24068417.

Mathew SJ, Hansen JM, Merrell AJ, Murphy MM, Lawson JA, Hutcheson DA, et al.
 Connective tissue fibroblasts and Tcf4 regulate myogenesis. Development.

2011;138(2):371-384. doi: 10.1242/dev.057463. PubMed PMID: 21177349; PubMed Central PMCID: PMCPMC3005608.

19. McWhinnie DJ, Saunders JW, Jr. Developmental patterns and specificities of alkaline phosphatase in the embryonic chick limb. Dev Biol. 1966;14(2):169-191. Epub 1966/10/01. PubMed PMID: 4382022.

Mehta V, Arora J, Suri R, Rath G. A rare quadruplicate arrangement of abductortor pollcis longus tendons - anatomical and clinical relevance. Clinics. 2009;64(2):153-155.
 Moore JS. De Quervain's tenosynovitis. Stenosing tenosynovitis of the first dorsal compartment. J Occup Environ Med. 1997;39(10):990-1002. PubMed PMID: 9343764.
 Moore K.L, Dalley A.F, Agur A. M. R. Essential Clinical Anatomy. Upper Limb. Wolters Kluwer, Philadelphia 2015: 398-482.

23. Nayak SR, Krishnamurthy A, Pai MM, Prabhu LV, Ramanathan LA, Ganesh kumar C, et al. Multiple variations of the extensor tendons of the forearm. Romanian Journal of Morphology and Embryology. 2008;49(1):97-100.

24. Nayak SR, Krishnamurthy A, Prabhu LV, Rai R, Ranade AV, Madhyastha S.
Anatomical variation of radial wrist extensor muscles: a study in cadavers. Clinics (Sao Paulo). 2008;63(1):85-90. PubMed PMID: 18297212; PubMed Central PMCID: PMCPMC2664178.

25. Newman SA, Frisch HL. Dynamics of skeletal pattern formation in developing chick limb. Science. 1979;205(4407):662-668. PubMed PMID: 462174.

 Rai R, Ranade AV, Mamatha T, Jiji PJ, D'Costa S, Maheshwari C. A rare origin of abductor pollicis longus. Romanian Journal of Morphology and Embryology.
 2010;51(2):399-400.

27. Ranade AV. Atypical insertion of the abductor pollicis longus muscle, an anatomical case report. Italian Journal of Anatomy and Embryology. 2017;122(2):147-150.

28. Rogozinski B, Lourie GM. Dissatisfaction After First Dorsal Compartment Release for de Quervain Tendinopathy. J Hand Surg Am. 2016;41(1):117-119. doi:

10.1016/j.jhsa.2015.09.003. PubMed PMID: 26481556.

Saunders JW, Jr., Gasseling MT, Errick JE. Inductive activity and enduring cellular constitution of a supernumerary apical ectodermal ridge grafted to the limb bud of the chick embryo. Dev Biol. 1976;50(1):16-25. Epub 1976/05/11. PubMed PMID: 1269827.
 Shirpa P, Srijit D. Variant Abductor Pollicis Longus Muscle: a Case Report. Acta Medica (Hradec Kralove). 2007;50(3):213-215. doi: 10.14712/18059694.2017.85.
 Suwannakhan A, Tawonsawatruk T, Meemon K. Extensor tendons and variations of the medial four digits of hand: a cadaveric study. Surgical and radiologic anatomy : SRA. 2016;38(9):1083-1093. doi: 10.1007/s00276-016-1673-2. PubMed PMID: 27056052.
 Swinehart IT, Schlientz AJ, Quintanilla CA, Mortlock DP, Wellik DM. Hox11 genes are required for regional patterning and integration of muscle, tendon and bone. Development. 2013;140(22):4574-4582. doi: 10.1242/dev.096693. PubMed PMID: 24154528; PubMed Central PMCID: PMCPMC3817943.

 Trivedi S, Siddiqui AU, Sinha TP, Sinha MB, Rathore M. Absence of extensor indicis: A rare anatomical variant. International Journal of Biomedical Research.
 2014;05(01):61-62. doi: https://doi.org/10.7439/ijbr.v5i1.442.

34. van Oudenaarde E. Structure and function of the abductor pollicis longus muscle. Journal of Anatomy. 1992;174:221-227.

35. Yammine K. The prevalence of the extensor indicis tendon and its variants: a systematic review and meta-analysis. Surgical and radiologic anatomy : SRA.

2015;37(3):247-254. doi: 10.1007/s00276-014-1352-0. PubMed PMID: 25096501.

36. Yuksel M, Onderoglu S, Arik Z. Case of an abductor pollicis longus muscle: variation or differentiation? Okajimas folia anatomica Japonica. 1992;69(4):169-171. PubMed PMID: 1436958.

Table 1: Reports the mean ratio of the right and left APB of the cadaver of interest, the mean ratio of the 21 muscles from the control cadaver, the alpha value, and the calculated p-value for a one tailed sample t test with 20 degrees of freedom.

		Mean of 21	STD of 21		
	Mean of Cadaver	muscles from	Muscles from		
Ratio of	of Interest (2	Control	Control	Alpha	
Width to	muscles)	Cadavers	Cadaver	Value	P-Value*
Length of					
APB	0.43	0.46	0.10	0.05	0.9141057

*This result is indicative that the ratio of width to length of the APB muscles of the cadaver of interest is not significantly different than the average ratio of the width to length of the 21 APB muscles of the 11 control cadavers.

FIGURE LEGENDS

Figure 1. (A) Dorsal aspect of the right forearm illustrating the proximal belly (PB) of the novel muscle running lateral and parallel to, but independent of the abductor pollicis longus (APL). IM = interosseous membrane, S = supinator muscle, EPB = extensor pollicis brevis. (B) Radial view of the right wrist showing the tendon of the novel muscle running parallel to the APL tendon. The distal belly (DB) of the novel muscle is visible in the thenar compartment. (C) Palmar view of the right hand. Flexor pollicis brevis muscle (FPB) and abductor pollicis brevis (APB) muscle with two distinct muscle bellies (APB1 and APB2) are visible. The DB of the novel muscle sits lateral to APB. Its tendon marked with an asterisk (*).

Figure 2. (A) Dorsal aspect of the left forearm illustrating the proximal belly (PB) of the novel muscle running lateral and parallel to, but independent of, the abductor pollicis longus (APL). IM = interosseous membrane, S = supinator muscle, EPB = extensor pollicis brevis (EPB). (B) Radial view of the left wrist showing the tendon of the novel muscle running parallel to the APL tendon. The distal belly (DB) of the novel muscle is visible in the thenar compartment (C) Palmar view of the left hand. Flexor pollicis brevis muscle (FPB) and abductor pollicis brevis (APB) muscle with two distinct muscle bellies (APB1 and APB2). The distal belly (DB) of the novel muscle sits lateral to the APB. Its tendon marked with an asterisk (*).













