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CASE REPORT

Bilateral Reversed Palmaris Longus Muscle and the Connection to Down Syndrome: A Case Report

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Abstract

Objective: The palmaris longus muscle is located in the forearm's anterior or flexor compartment and exhibits a wide range of anatomical variations. Reversed palmaris longus muscle, with the muscle belly at the distal attachment, is rare, and the bilateral reversed variant is even more uncommon. Here, we present a case of bilateral reversed palmaris longus muscle in a donor with Down syndrome and explore its genetic, morphological, clinical, and surgical implications.

Methods: This case details the discovery of a bilateral reversed palmaris longus muscle in a donor with a history of Down syndrome. Images of the variation were obtained and measurements were recorded.

Results: A reversed palmaris longus muscle was observed in the anterior or flexor compartment of both forearms. On the right forearm, a proximal tendon was followed by a distal muscle belly in the wrist, with two distal tendon attachments. The left reversed palmaris longus muscle had a proximal tendon followed by a distal muscle belly and a single distal tendon attachment. No other anatomical abnormalities were observed in the anterior forearm bilaterally.

Conclusion: This case, to the knowledge of the authors, details the first documented instance of bilateral reversed palmaris longus muscle in an individual with Down syndrome. Although the relationship between Down syndrome and palmaris longus muscle variants has rarely been explored, abnormal gene expression may contribute to this association. This case aims to increase awareness and offer insight into an underrecognized variant of the palmaris longus muscle, highlighting the importance of identifying such variants in clinical evaluation and surgical planning.

Key Words: *Anatomic variation; Bilateral; Down syndrome; Palmaris longus; Reversed*

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Introduction

The palmaris longus muscle (PLM) lies in the superficial layer of the anterior forearm. Its proximal attachment is the medial epicondyle of the humerus, extending medially to travel in tandem with the flexor carpi radialis muscle. Typically, the muscle belly arises from the proximal attachment and becomes tendinous in the mid-forearm. The PLM tendon passes anterior to the flexor retinaculum, with some fibers attached to the retinaculum itself. Most fibers fan out, attaching broadly to the palmar aponeurosis. In terms of neurovasculature, the PLM is supplied by the median nerve and a branch of the anterior ulnar recurrent artery. The PLM tendon often serves as a gross landmark for locating the median nerve because of its close relationship. The PLM contributes to wrist flexion and prevents displacement of the skin and fascia from horizontal shearing forces [1].

The PLM is widely considered the most variable muscle in the human body, most commonly presenting as agenesis in 20.25% of the population [2]. A reversed PLM is defined as having a proximal tendinous origin, with the muscle belly reaching the palmar aponeurosis. Other variants include duplicate, triplicate, intermediate muscle belly, and digastric [3]. These variants have a combined incidence of 9% [4].

Despite significant progress in learning anatomy using other methods, including modern imaging techniques, the use of donors remains of great value in identifying anatomical variations. Discovering these differences in anatomy, even if rare, is essential for the proper diagnosis and management of related clinical disorders [5].

Here, we present a case of bilateral reversed PLM in a donor with a history of Down syndrome identified during a medical school anatomy lab dissection. While variations such as PLM agenesis are well documented, bilateral reversed PLM is rarely reported. One study identified only five cases in the literature, all in males [6]. Additionally, the association between Down syndrome and variants of the PLM has not yet been explored to the author's knowledge. Awareness of the diverse presentations of the PLM among clinicians, anatomists, and surgeons is essential for surgical planning, proper identification, and addressing symptomatic presentations. By documenting this case and examining previous occurrences, we aim to improve recognition and understanding of this rare variant.

Methods

In November 2024, during the routine gross anatomy dissection at the Creighton University School of Medicine, a bilateral reversed PLM was identified in a 71-year-old male donor with a history of Down syndrome. A single palmar crease was observed on the palmar aspect of both hands. The PLM bellies were located distally near the wrist bilaterally, with two distal tendons identified on the right forearm, and only one distal tendon identified on the left. Both muscles terminated distally, inserting into the flexor retinaculum and palmar aponeurosis. Approximately one month after dissection, both arms were abducted to photograph the variant muscles (Figure 1).

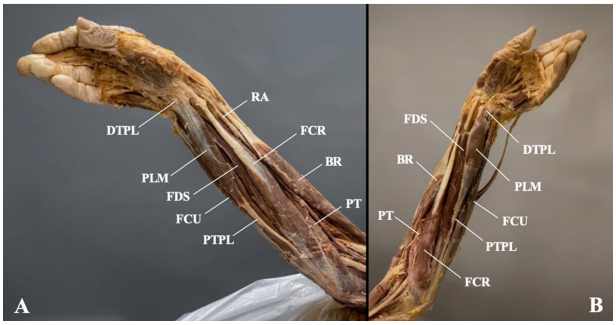

Figure 1) *Donor photographs showing the reversed palmaris longus muscles. A) Right upper limb. B) Left upper limb.*

BR: brachioradialis muscle, DTPL: distal tendon of palmaris longus muscle, FCR: flexor carpi radialis muscle, FCU: flexor carpi ulnaris muscle, FDS: flexor digitorum superficialis muscle, PLM: palmaris longus muscle, PT: pronator teres muscle, PTPL: proximal tendon of palmaris longus muscle, RA: radial artery

A brief review of the literature was performed to assess the presence of bilateral reversed PLM in patients with Down syndrome. Multiple databases were used, including Google Scholar, PubMed, Scopus, Medline, Embase, and CINAHL, using the search query ((“reversed palmaris longus”) OR (“palmaris longus inversus”) OR (“palmaris inversus longus”) OR (“inverse palmaris longus”) OR (“reverse palmaris longus”) AND (“down syndrome”) AND (“trisomy 21”)), which returned zero reported cases. To our knowledge, this case details the first documented occurrence of a reversed bilateral PLM in an individual with Down syndrome.

Results

The PLM measurements are summarized in Table 1. On the right forearm, the muscle belly of the PLM measured 126.6 mm, and the proximal tendon measured 124.4 mm. The lateral distal tendon measured 15.3 mm, and the medial distal tendon measured 10.1 mm. The total length of the right PLM measured 266.3 mm. On the left forearm, the muscle belly of the PLM measured 116.5 mm, the proximal tendon measured 135.9 mm, and the distal tendon measured 23.0 mm. The total length of the left PLM measured 275.4 mm. The left PLM was cut at the distal attachment as part of the routine dissection protocol. No additional variations or abnormalities were observed in adjacent structures.

Discussion

Embryologic and genetic considerations of the palmaris longus muscle

The embryologic development of the PLM is more easily understood within the context of the superficial forearm flexor muscle group. Development of these muscles begins as a common flexor mass composed of superficial and deep layers. In response to factors produced by limb mesenchyme, delamination and migration of upper limb progenitor cells occur. The chemoattractant SDF1, secreted by the core of the limb mesenchyme, promotes migration of the muscle progenitor cells to their proper location. Ephrin 5A, another chemoattractant located in specific areas of the limb mesenchyme, determines proper positioning of migrating muscle progenitor cells [7].

In cases of PLM agenesis, evidence suggests that the muscle fails to form entirely, rather than forming and later regressing [8]. This suggests that abnormalities are a result of early disruptions in embryonic signaling pathways, rather than late-stage developmental degeneration. While the specific embryological pathogenesis of a reversed PLM is unknown, a connection could exist between abnormal delamination and migration of muscle progenitor cells and PLM variants; however, this requires further investigation.

Currently, no single gene has been explicitly linked to the presence, absence, or reversal of the

Table 1: Summary of palmaris longus muscle measurements.

Measurements (mm)	Right	Left
Total Length	266.3	275.4
Muscle Belly	126.6	116.5
Proximal Tendon	124.4	135.9
Distal Tendon	Lateral: 15.3 Medial: 10.1	23.0

PLM. However, population studies suggest that PLM agenesis follows an autosomal dominant inheritance pattern with incomplete penetrance and variable expressivity [9,10]. Whether a bilateral reversed presentation is coincidental or influenced by specific genetic mechanisms warrants further investigation; however, the explanation likely involves complex interactions between genetic and environmental factors.

Palmaris longus muscle variation in down syndrome

The following report, to our knowledge, details the first documented occurrence of a bilateral reversed PLM in an individual with Down syndrome. Trisomy 21 is well-established as being associated with increased instability in normal developmental processes and a higher frequency of musculoskeletal anomalies [11,12]. While direct evidence of an association between Down syndrome and PLM variants is limited, several mechanisms could link them. The presence of an extra chromosome 21 may increase developmental instability, leading to higher frequencies of anatomical malformations, including muscle variants. This mechanism has been demonstrated for other anatomical structures in individuals with Down syndrome [12]. Additionally, Boyle et al. [13] hypothesize that reduced fetal apoptosis results in increased incidence of supernumerary muscles in individuals with Down syndrome, supporting the notion of a genetic association. Altered collagen metabolism is also observed in patients with Down syndrome, with the type VI collagen genes COL6A1 and COL6A2 being located on chromosome 21 [14]. Since type VI collagen is crucial for muscle function and stability, altered expressions of these genes could theoretically influence muscle development and variant formation. Trisomy 21 also affects muscle development through altered gene expression patterns involving muscle metabolism and mitochondrial function, resulting in hypotonia [15]. These developmental disruptions could potentially influence the formation of muscle variants. Although prevalence studies on supernumerary muscles in individuals with Down syndrome are lacking, prior research has noted a greater occurrence of primitive muscular structures in those with trisomies, which could explain our finding [16]. Future investigations should consider the incidence and morphology of bifid, accessory, or reversed PLM forms more broadly in populations with chromosomal abnormalities, which may help clarify the relationship between genetic factors and evolutionary development involving limb bud formation.

Clinical and surgical implications

The presence of a reversed PLM presents unique challenges in the clinical management and surgical planning of these patients. A reversed PLM can present as a soft tissue mass, which requires thorough radiological evaluation for complete characterization and diagnosis [17]. The distal positioning of the muscle belly occupies more space within the carpal tunnel than its typical presentation, potentially increasing pressure on the median nerve and leading to pain, swelling, and paresthesia [18]. In such cases, the symptomatic PLM can usually be successfully treated by surgical excision, decompressive fasciotomy, median nerve release, or a combination of these [19].

The PLM is commonly utilized in tendon grafting procedures due to its expandability and anatomical accessibility. The loss of the tendon of the PLM produces no functional deficit and is thus often used by surgeons for tendon and joint reconstruction procedures. The PLM is considered an ideal autogenous donor due to its superficial location in the anterior forearm and its ability to be stretched without tearing [20].

The tendon of a reversed PLM may be shorter, thicker, or abnormally positioned, making it a less ideal donor site. Thus, preoperative imaging is essential when a reversed PLM or other variants are suspected in order to characterize the muscle for appropriate tendon graft usage.

Conclusion

To the author's knowledge, this study presents the first documented case of a bilateral reversed PLM identified in an individual with Down syndrome. While the exact mechanism for this abnormal development is unknown, associations with chromosomal instability and altered gene expression in trisomy 21 could explain a predisposition to such morphological variations. Our findings highlight the importance of radiological assessment and precise surgical planning when evaluating wrist and forearm symptoms in individuals with Down syndrome. Continued documentation of similar cases is essential to determine if this is an isolated finding or a reflection of a broader anatomical pattern among this population.

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