CASE SERIES

Case series on Linburg-Comstock Variation in Sri Lanka

E A A M Wickramarathna[1], C S Wijayabandara[1], K M Chandimal[1], A Wijayasekara[2]

[1] *Department of Anatomy, Faculty of Medicine, Wayamba University of Sri Lanka*

[2] *Department of Parasitology, Faculty of Medicine, Wayamba University of Sri Lanka*

Abstract

The case series present a rare anatomical variation involving an anomalous connection between the flexor pollicis longus and flexor digitorum profundus tendons in the forearm called Linburg-Comstock variation. These three cases were identified in Sri Lankan population, highlighting the presence of this variation in the local population. The unique anatomical details and potential implications for hand function in individuals with Linburg-Comstock variation are discussed.

DOI: https://doi.org/10.4038/slaj.v8i1.230

Sri Lanka Anatomy Journal 2024; 8(I): 91–96

Corresponding author
EAAM Wickramarathna
E-mail: amilaw@wyb.ac.lk

https://orcid.org/0009-0005-5105-1697

Introduction

Linburg-Comstock variation, an anatomical variant in the distal upper limb, is a tendinous connection between the flexor digitorum profundus (FDP) and the flexor pollicis longus (FPL) (1). The FPL flexes the interphalangeal joint of the thumb, and the FDP flexes the distal interphalangeal (DIP) joint of other four fingers (2). Embryologically, both FDP and FPL muscles in humans originate from the same mesodermal mass (3). However, electromyographic studies have revealed that independent functions of the FPL and FDP muscles enable the FDP to provide strong flexion without activation of the FPL muscle. Strong independent function of FDP and FPL muscles is a unique evolutionary characteristic in humans which allows efficient manipulation of tools (4). In non-hominoid primates, the FPL tendon connects with the FDP tendons, resulting in simultaneous flexion of the other digits and the thumb (5).

Tendinous interconnection in Linburg-Comstock variation prevents independent flexion of the thumb at the interphalangeal (IP) joint and the index finger at the distal interphalangeal (DIP) joint. Occasionally, the middle and ring fingers are also affected by this connection (6). Typically, the interconnection with varying in size is concealed within a thickened tenosynovium (7).

Although this variant is usually asymptomatic, it sometimes leads to a variety of symptoms such as median nerve neuropathy, restrictive thumb-index flexor tenosynovitis, carpal tunnel syndrome, traumatic rupture of the anomalous tendon (7-10). The presence of symptoms associated with this variation is called Linburg-Comstock syndrome. Symptomatic patients often complain of tearing pain with attempts of independent movements of the thumb at interphalangeal (IP) joint and the index finger at distal interphalangeal (DIP) joint. They may also have challenges with everyday tasks like writing, or tying shoelaces (7, 11).

The first report of this variant was published in 1979 with the participation of 194 patients showing 31% of prevalence (1). Following that, a number of case reports and studies have been published across the globe including Israel, China, Spain, Brazil, and Saudi Arabia (3,6,12-14). Most of the studies show a female preponderance and high frequency of unilateral presentations. However, a recent metanalysis reveals no difference in laterality but confirms the female preponderance. While this condition has been widely studied in many other populations, there is limited data available in the Sri Lankan population.

The clinical diagnosis of the variation is assessed using two clinical tests developed by Linburg and Comstock. The first test involves asking the participant to flex their thumb actively while their forearm is in a supinated position and their hand is spread out. The presence of the anomaly is indicated if any of the fingers also flex concurrently. If the anomaly is observed in the first test, a second test is administered to determine pain in the distal volar region of the forearm and wrist. There the participant is asked to flex their thumb actively while the examiner manually extends the long fingers passively (1).

Nowadays, magnetic resonance imaging (MRI) is also used as an aid in confirmation and localization of the variant connection (15).

Case Presentations

First case is a 28-year-old, right-hand-dominant male from Rambukkana working at the same department of authors'. He was incidentally noted to have simultaneous motion of right index finger with motion of thumb. Upon physical examination, active flexion of the interphalangeal (IP) joint of the thumb of right hand was always accompanied by simultaneous flexion of the index finger at its distal interphalangeal (DIP) joint (Figure 01). He complained of no pain or discomfort when the thumb is actively flexed while the examiner maintains the passive extension of the other four fingers (figure 02). He denies any challenges with everyday tasks like writing or tying shoelaces. None of his living family members were having the variation.

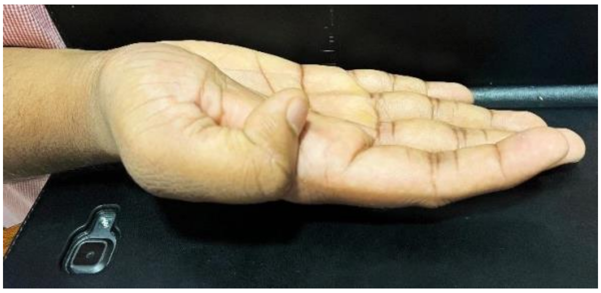

Figure 01: shows simultaneous flexion of the index finger at its distal interphalangeal (DIP) joint with the active flexion of thumb at interphalangeal (IP) joint in the first case.

Figure 02: shows the pain identification test. Individual may complain of pain with active thumb flexion while the examiner maintains the passive extension of the long fingers.

Additionally, a 29-year-old female from Polgahawela, known to case one but not genetically related was found to have the same observation of being unable to independently move her fingers on her right dominant hand. On examination, she was also found to have simultaneous flexion of the index finger at DIP joint with the active flexion the thumb at IP joint (Figure 03). She was also asymptomatic after testing for pain as shown in figure 02 and denies any family history of the presentation. No abnormalities were found in grasping or using tools or challenges in previously mentioned everyday tasks.

Figure 03: shows simultaneous flexion of the index finger at its distal interphalangeal (DIP) joint with the active flexion of thumb at interphalangeal (IP) joint in the second case.

Another case of a 29-year-old, right-hand-dominant male from Matara was also noted to have the same abnormal motion of the right index finger and thumb. He was also known to case one but not genetically related and reported to authors in the same month. Active flexion of the interphalangeal (IP) joint of the thumb of the right hand was followed by simultaneous flexion of the index finger at its DIP joint (Figure 04). After testing for pain, as shown in figure 02, he was also found to be asymptomatic and there were no other individuals in the family with the variation.

Figure 04: shows simultaneous flexion of the index finger at its distal interphalangeal (DIP) joint with the active flexion of thumb at interphalangeal (IP) joint in the third case.

Discussion

The embryological basis of the origin of FPL and the FDP describes the possible existence of tendinous connection between these two muscles, leading to this anatomical variation (5). The first report of this variant by Linburg and Comstock in 1979 with participation of 194 patients shows 31% of prevalence at least in one extremity (1). To our knowledge, these are the first reported cases of Linburg-Comstock variation in Sri Lankan population. All three of these case reports are unilateral presentations. Studies from various countries have reported a predominance of unilateral presentations (16-18). However, discrepancies in laterality have also been noted, with some studies showing no significant difference between bilateral and unilateral cases. Though female predominance has been observed in some of the studies (16-19), two out of the three reported cases were males. However, the findings of the three case reports are inadequate to comment on gender-based variations in the Sri Lankan population.

Having the variant involving only the index fingers in the reported cases also following the typical finding of the variant (6). Absence of challenges with everyday tasks like writing or tying shoelaces or pain in wrist or distal forearm in moving the thumb and index fingers independently in the dominant hand in both reported cases confirm the fact that vast majority with the variant are asymptomatic (7). Interestingly, two of the reported cases were from the close geographical areas (Polgahawela and Rambukkana) suggesting potential genetic or environmental factors that may influence the development of the Linburg-Comstock variation among people from the same geographic origin. However, further studies need to be conducted to explore potential factors that influence the development of this variation.

Despite data on this variant being available in other populations (3, 6, 12-14), there is a lack of data on the prevalence and clinical manifestations of this variant in the Sri Lankan population. Understanding the prevalence of Linburg-Comstock variant and syndrome in the Sri Lankan population could enhance clinical practice and enable healthcare providers to more effectively manage and treat individuals with this anatomical variation.

Conclusion

The reported case series describe the rare observation of Linburg-Comstock variation in three Sri Lankan individuals. Further, research to explore the prevalence of Linburg-Comstock variant and exploring the clinical significance of Linburg-Comstock syndrome is crucial to expand the knowledge of the condition among the Sri Lankan population.

Ethical approval

Informed written consent was obtained from all three individuals to publish their data and images. Individuals' anonymity and the confidentiality was strictly maintained.

References

1. Linburg RM, Comstock BE. Anomalous tendon slips from the flexor pollicis longus to the flexor digitorum profundus. The Journal of hand surgery. 1979 Jan 1;4(1):79-83.
2. Drake RL, Vogl AW, Mitchell AW. Gray's anatomy for students, 3rd edn Philadelphia. PA: Churchill Livingstone Elsevier. 2015.
3. Stahl S, Stahl S, Calif E. Failure of flexor pollicis longus repair caused by anomalous flexor pollicis longus to index flexor digitorum profundus interconnections: a case report. The Journal of hand surgery. 2005 May 1;30(3):483-6.
4. Williams EM, Gordon AD, Richmond BG. Hand pressure distribution during Oldowan stone tool production. Journal of Human Evolution. 2012 Apr 1;62(4):520-32.
5. Chavez TJ, Morrell NT. The evolution of the human hand from an anthropologic perspective. The Journal of Hand Surgery. 2022 Feb 1;47(2):181-5.
6. Yoon HK, Kim CH. Linburg–Comstock syndrome involving four fingers: a case report and review of the literature. Journal of Plastic, Reconstructive & Aesthetic Surgery. 2013 Sep 1;66(9):1291-4.
7. Gancarczyk SM, & Strauch RJ. Linburg-Comstock Anomaly. The Journal of Hand Surgery. 2014.39(8), 1620–1622.
8. Lombardi RM, Wood MB, Linscheid RL. Symptomatic restrictive thumb-index flexor tenosynovitis: incidence of musculotendinous anomalies and results of treatment. The Journal of hand surgery. 1988 May 1;13(3):325-8.
9. Slater RR. Flexor tendon anomalies in a patient with carpal tunnel syndrome. Journal of Hand Surgery. 2001 Aug;26(4):373-6.
10. Old O, Rajaratnam V, Allen G. Traumatic correction of Linburg–Comstock anomaly: a case report. The Annals of the Royal College of Surgeons of England. 2010 May; 92(4):e1-3.
11. Badhe S, Lynch J, Thorpe SK, Bainbridge LC. Operative treatment of Linburg-Comstock syndrome. The Journal of Bone & Joint Surgery British Volume. 2010 Sep 1;92(9):1278-81.
12. Aguado AR, del Pino Paredes V. Flexor digitorum profundus common to thumb and

index finger, associated with a post-traumatic distal adherence of both tendons. The Journal of Hand Surgery: British & European Volume. 1988 Feb 1;13(1):72-4.

13. Barreto LC, Fernandes CH, Nakachima LR, Santos JB, Fernandes M, Faloppa F. Prevalence of the Linburg-Comstock anomaly in a Brazilian population sample. Revista Brasileira de Ortopedia. 2020 Jul 22;55(3):317-22.

14. Alzahrani MT, Almalki MA, Althunayan TA, Almohawis AH, Almehaid FF, Umedani LV. Functional anatomy of the hand: prevalence of the linburg–Comstock anomaly in a young saudi population. Journal of Musculoskeletal Surgery and Research. 2018 Jan 1;2:21.

15. Karalezli N, Haykir R, Karakose S, Yildirim S. Magnetic resonance imaging in Linburg-Comstock anomaly. Acta Radiologica. 2006 May;47(4):366-8.

16. Hamitouche K, Roux JL, Baeten Y, Allieu Y. Linburg-Comstock syndrome. Epidemiologic and anatomic study, clinical applications. Chirurgie de la Main. 2000 May 1; 19(2):109-15.

17. Karalezli N, Karakose S, Haykir R, Yagisan N, Kacira B, Tuncay I. Linburg–Comstock anomaly in musicians. Journal of plastic, reconstructive & aesthetic surgery. 2006 Jul 1; 59(7):768-71.

18. Low TH, Senan NA, Ahmad TS. The Linburg-Comstock anomaly: incidence in Malaysians and effect on pinch strength. The Journal of hand surgery. 2012 May 1; 37(5):930-2.

19. Yammine K, Erić M. Linburg–Comstock variation and syndrome. A meta-analysis. Surgical and Radiologic Anatomy. 2018 Mar; 40:289-96.