

Isolated Flexor Pollicis Longus Hypoplasia With Absence Of The Interphalangeal Crease: A Case Report And Review Of The Literature

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Abstract

This report represents the first documented case in history of a patient with a combination of congenital hypoplasia of the flexor pollicis longus, in the absence of other muscular, skeletal or soft tissue anomalies, and the absence of an interphalangeal flexor crease of the first digit. This case report displays a unique presentation in the limited literature representing the disease spectrum associated with flexor pollicis longus aplasia and hypoplasia. The astute orthopedic physician should be aware of the variable presentations of this pathology, including absence or presence of an interphalangeal crease, hypoplasia or aplasia of neighboring osseous or muscular structures, as well as limited or absent thumb interphalangeal joint motion and the associated functional findings.

INTRODUCTION

The intricate nature of the digits and the ability to grasp, pinch, and hold, has allowed for the advancement of man through history. No other digit in the hand has the importance of the thumb. Approximately 50% of the function of the hand is attributed to the actions of the thumb, and it is credited as the most important member of the upper extremity in terms of function. Thus, it stands to reason that congenital aberrations of the thumb should be studied and better understood to prevent loss of function. This report represents the first documented case in history of a patient with a combination of congenital hypoplasia of the flexor pollicis longus, in the absence of other muscular, skeletal or soft tissue anomalies, and the absence of an interphalangeal flexor crease of the first digit.

Froment in 1895 was the first to report agenesis of the FPL, which was associated with lumbrical anomalies, absent thenar muscles and absent flexor digitorum superficialis. Isolated aplasia or hypoplasia of flexor pollicis longus (FPL) is an extreme anatomical rarity, only 13 documented in history (1-12). Aplasia or hypoplasia of the FPL in history has been commonly associated with a company of anomalies related to median nerve innervated structures, but rarely seen

in isolation. Of the 13 documented cases, 2 are bilateral, 1 of which is associated with craniofacial abnormalities. Eleven are unilateral, 2 associated with hypoplasia of either the muscular of the thenar eminence or of the osseous structures of the first ray of the hand. The remaining 9 have isolated FPL aplasia or hypoplasia. Eight of these cases all have distinct flexor creases of the interphalangeal joint of the effected thumb. This patient represents the first documented case in history of a patient with the combination of congenital hypoplasia of flexor pollicis longus, in the absence of thumb hypoplasia or aplasia in terms of soft tissue, musculature or osseous structures of the thenar eminence or first ray, and an associated absent interphalangeal flexor crease of the first digit. Other cases were noted to have a paucity of an interphalangeal crease, although they also displayed aplasia or hypoplasia of the FPL. One other case has been documented which is similar in nature, but there was documented aplasia, and therefore absence of the FPL.

CASE REPORT

The patient reported to the orthopedic clinic with complaints of discomfort and profoundly limited ability to actively flex the interphalangeal joint of the thumb. It was immediately

noted the child had an absence of a flexor crease at the thumb's interphalangeal joint. Imaging and clinical exam confirmed symmetrical osseous anatomy consistent with that of an 11 year old child, with no skeletal or muscular hypoplasia noted on the bilateral hand, but the workup did lead to the diagnosis of a hypoplastic FPL as compared to the contralateral upper extremity. The patient had an unremarkable medical history, and was essentially a healthy child. Discomfort with active range of motion is suspected to be caused by soft tissue contractures and joint stiffness secondary to long-term severe limited motion at the effected joint.

DISCUSSION

The patient was an 11 year old male at the time of diagnosis and it was an incidental finding, as the discovery was made when the patient reported to the hospital regarding a fractured phalanx on the contralateral hand. The majority of cases in the literature follow a similar theme, in which the anomaly is discovered in children and are usually coincidental in nature. Most of the patients discovered with such defects have adapted to their disabilities to some degree, but the vast majority have received orthopedic intervention in the form of tendon transfers to correct for functional deficits (13). The ability of the patient to adapt to the disability correlates to the observation most of these discoveries are coincidental in nature and are thus not the reason the patient initially sought the medical care which eventually lead to the chance finding of the anomaly. The father of the patient claims he has noticed the child has some difficulty with feeding himself with the effected hand, as well as holding writing instruments such as pencils, pens and crayons. Upon proposing the possibility of surgical intervention in the form of a tendon transfer, the father denied entertaining the notion. He claimed the child had no notable debility which would warrant surgery at this time. Surgical intervention would allow for direct visualization of FPL and the associated tendon, which would provide definite evidence of this finding, superior to any imaging study or physical exam. The vast majority of similar cases underwent surgical intervention via tendon transfers, with good results. Long-term follow-up is not available for the 3 cases in the literature whom denied surgical treatment.

This case report displays a unique presentation in the limited literature representing the disease spectrum associated with flexor pollicis longus aplasia and hypoplasia. The astute orthopedic physician should be aware of the variable

presentations of this pathology, including absence or presence of an interphalangeal crease, hypoplasia or aplasia of neighboring osseous or muscular structures, as well as limited or absent thumb interphalangeal joint motion and the associated functional findings.

HUMAN AND ANIMAL RIGHTS

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008. Informed consent was obtained from all patients for being included in the study.

DECLARATION OF CONFLICTING INTEREST

The Authors declares that there is no conflict of interest in regards to the content or publication of this paper.

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INFORMED CONSENT

Informed consent was obtained from all individual participants included in the study.

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