Congenital absence of abductor pollicis brevis

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Congenital thenar hypoplasia is a rare anomaly, and even more perplexing when it is isolated to a specific muscle with associated nerve and vascular anomalies. In the present article, the authors report a case involving a 14-year-old female with unilateral absence of the abductor pollicis brevis muscle, a bifid median nerve and persistent median artery presenting with wrist pain. Comprehensive assessment, including radiographic, electromyographic and magnetic resonance imaging studies, were used to evaluate and document this anomaly.

Key Words: *Abductor pollicis brevis; Bifid median nerve; Hand; Median artery; Thenar atrophy*

All individuals are unique and complex. Contributing to this are myriad genetic patterns and embryonic changes. Congenital anomalies affect 1% to 2% of newborns, in whom approximately 10% are upper limb abnormalities (1). While early insults commonly result in death of the embryo, later insults during the stage of growth and maturation typically result in minor functional deficits such as overgrowth or hypoplasia that may go unnoticed. A clear understanding and knowledge of these anomalies, from the common to the most severe or rarest deformity, aids in clinical judgment and patient reassurance when presenting with concomitant trauma or other symptoms. The present report details a case involving a 14-year-old girl with chronic wrist pain and associated thenar atrophy. Detailed workup revealed absence of the abductor pollicis brevis (APB), a bifid median nerve and persistent median artery (PMA).

CASE PRESENTATION

A healthy right-handed 14-year-old girl presented to hand clinic with a complaint of chronic right wrist pain from a fall on an outstretched hand five months previously. Examination showed significant thenar atrophy (Figure 1). Opposition was weak and static two-point discrimination measured 7 mm in the median nerve distribution. The volar-radial wrist pain was reproduced by hyperextension. The remainder of the upper extremity examination was unremarkable. The patient and her parents were aware of the deformity from three years of age but noted minimal functional impairment. No history of trauma, pregnancy complication or family history of congenital anomalies was presented.

X-rays of the hand did not reveal any osseous abnormalities. Magnetic resonance imaging of the wrist was negative for ligamentous injury, but demonstrated absence of the APB muscle with a small tendon remnant. The other intrinsic muscles were present and of normal configuration (Figure 2). There was a high median nerve division with PMA coursing in between (Figure 3). The radial duplicated median nerve became the recurrent nerve coursing toward the thenar musculature. An electromyogram confirmed the absence of APB; however, no evidence of median neuropathy was present.

With conservative nonsurgical management, the patient's wrist pain subsided and static two-point discrimination was measured at 4 mm on subsequent follow-up. She reported occasional discomfort from overuse when playing basketball and volleyball. Her thenar anomaly was assessed to be congenital without significant functional compromise that did not require intervention.

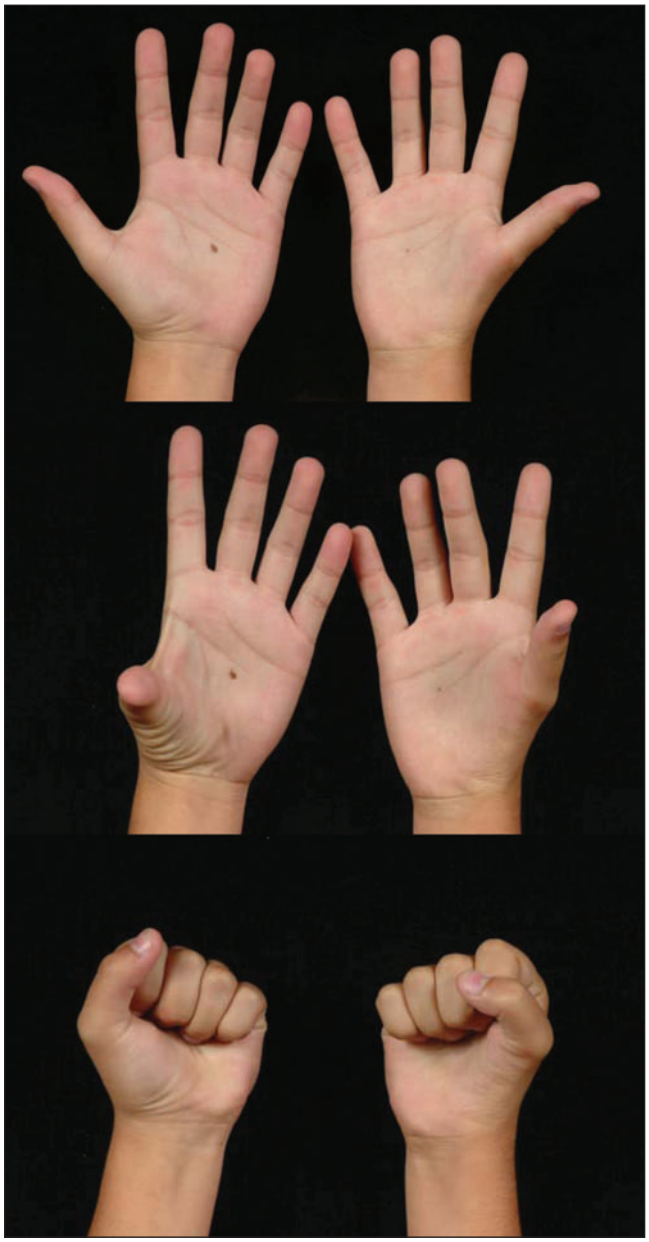

Figure 1) *Patient's hands in radial and palmar abduction and full-fisted positions*

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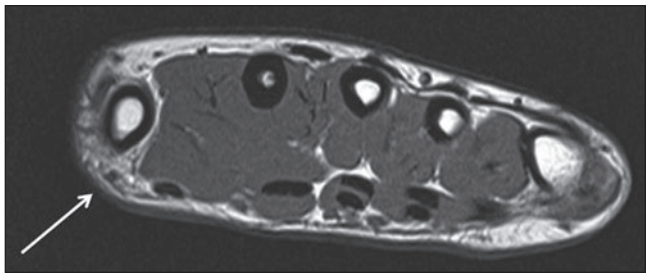

Figure 2) *A T1-weighted axial magnetic resonance image at the level of the shaft of the first metacarpal demonstrating absence of the abductor pollicis brevis muscle. Instead, there is a thin tendon in its expected position indicated by the arrow*

DISCUSSION

There are numerous case reports of thumb hypoplasia and its variances, from the most severe deformity of radial club hand with completely absent radius and thumb to the mild diminution of thenar muscle bulk. Described within these reports are findings of other syndromes that are highly associated with radial anomalies, most commonly Holt-Oram syndrome and VACTERL (2). However, to our knowledge, there have not been any reported cases of isolated APB absence with associated nerve and vascular anomalies. This may be due to the technological differences that have evolved in the recent decades. Past cases reported atrophy without further distinguishing specific muscles within the thenar musculature. Thus, lacking among these reports are advance imaging, enabling further differentiation of specific anatomical differences in these rare anomalies. Technological advances will undoubtedly facilitate understanding of the congenital deformities and their pathogenesis to aid in surgical planning if warranted.

We can only speculate whether the three abnormalities identified in the present case are manifestation of a syndrome or a congenital sequence resulting in absence of the APB muscle. This fundamental defect could simply be from the failure of differentiation of the muscles (the APB muscle primordium was already deficient before any interconnection to the median nerve motor branch). In contrast, this could be due to a very specific localized insult to the inductive tissue during the sixth week of embryogenesis when the intrinsic muscle mass appears.

The presence of the PMA and bifid median nerve strengthen the congenital sequence theory in the differential of this interesting anomaly. The median artery is a transitory vessel providing the main blood supply to the hand in the embryo. The artery usually regresses during the eighth week of gestation as the ulnar and radial arteries develop, becoming a small vestigial vessel accompanying the median nerve (3). Its presence into adulthood has been associated with numerous complications related to median nerve compression. Several reported cases of carpal tunnel syndrome (CTS) caused by PMA secondary to thrombosis (4), dilation (5) and trauma (6) have been documented. Its presence may have also contributed to our patient's wrist discomfort during sports. One reported theory is that temporal compression of the median nerve occurs from vasodilation and increased pressure in the PMA during strenuous exercise (3). Unfortunately, we did not have the capacity to test this hypothesis by measuring flow and diameter of the PMA before and after exercise.

Thenar hypoplasia has generally been described as sequelae of median nerve neuropathy – a common problem in the adult population; however, it has rarely been reported in children (7,8). The patient's initial symptoms and interesting examination findings were

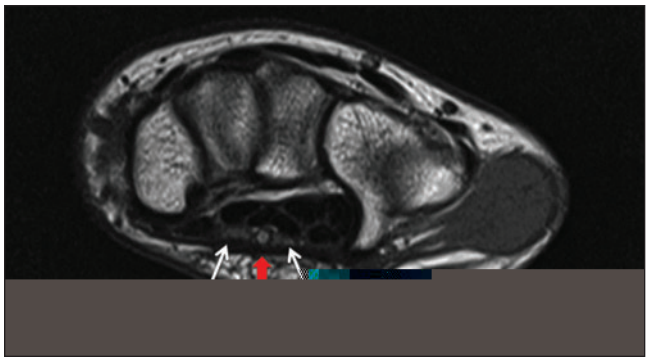

Figure 3) *A T1-weighted axial magnetic resonance image at the wrist at the level of the hook of the hamate is depicted. The white arrows indicate the duplicated median nerves. The red arrow indicates the persistent median artery, which is located between the median nerves*

suggestive of CTS; however, radiographic and nerve testing were not confirmatory. Perhaps the presence of the PMA and bifid median nerve were contributing factors lowering the threshold for CTS, with the wrist trauma serving as an inciting insult causing transient median nerve neurapraxia.

The present case is a rare, detailed description of congenital APB absence with associated nerve and vascular anomalies and, to our knowledge, previously unpublished. It highlights the need for thorough evaluation in young patients presenting with pain secondary to trauma. In the present case, a routine workup for wrist pain led to the discovery of unique congenital anomalies of initial uncertain significance. We believe these unique neurovascular anomalies may have contributed to the patient's transient CTS, with the absence of the APB muscle being a clinical clue to the underlying pathology. Fortunately, the patient recovered well without surgical intervention, reaffirming that not every congenital abnormality needs surgical correction, and may simply contribute to unique genetic composition.

DISCLOSURES: The authors have no financial disclosures or conflicts of interest to declare.

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