PEDIATRIC TRIGGER THUMB: A TALE OF TWIN SISTERS
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ABSTRACT
Although very rare in twins, pediatric trigger thumb demands early identification and treatment for successful resolution. The results are better in younger patients as it allows for motor learning and functional skills. However, for older or refractory patients surgical release is indicated. Recurrence is also common so follow up is needed. A case report of twin sisters is presented.

Keywords: Flexor pollicis longus tendon, Pediatric trigger thumb.

INTRODUCTION
Though prevalence of pediatric trigger thumb (PTT) is reported to be 2.2% of all upper extremity pediatric anomalies; it is an infrequent problem to present with, in pediatric population, to a physiatrist. The etiology of the condition is by far unclear. Recent studies have supported the opinion that it is an acquired condition caused by trauma. However, some authors have proposed that it is primarily congenital or hereditary.

The pathology, however, is the same i.e. a disparity in the size of flexor retinaculum and the surrounding sheath mostly at the level of first annular ring (A1) producing resistance to the gliding of flexor pollicis longus (FPL). The PTT mostly occurs unilaterally and is extremely rare in twins. Only a few case reports of identical twins with congenital PTT have been described. The typical presentation is at the age of two years with the interphalangeal (IP) joint locked in flexion and a palpable nodule in FPL tendon. We report here, two identical twins who presented in their preschool age with PTT. To the best of our knowledge, it is the first report of its kind from Pakistan.

CASE REPORT
Two 2½-year-old identical twin sisters were brought by their mother to the outpatient rehabilitation department with complaints of deformed thumbs. One girl had unilateral while the other had bilateral deformities. The deformity was noticed by the mother at the age of five months and on visiting a clinic she was told by a general practitioner that the deformity would settle with the children’s growth. Both the girls were right handed. They were finding it difficult to do tasks involving thumb movements and grasp like holding pencils. The deformity did not produce pain nor interfere with routine daily activities e.g. playing or self-feeding. Their past medical, birth, family and social histories were unremarkable. The mother denied any history of drug intake during pregnancy.

On examination, the hand examination was unremarkable except for the flexion contractures of the IP joints of the thumbs.

Figure: Showing flexion contracture at the interphalangeal joint of both thumbs of the first twin (a), resolution of flexion contracture after surgery in the first twin (b), thumb of the second twin before and after surgery (c).

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making active or passive IP extension impossible. (Fig-a) A non-tender hard nodule was present at the IP joints along with thickening of flexor pollicis longus (FPL) tendon. Triggering was absent. Distal sensations and capillary refill were normal bilaterally. No other apparent physical abnormality was found. Based on history and examination, a diagnosis of congenital PTT was made. Initial trial of physiotherapy and occupational therapy was given for 2-4 weeks, but due to lack of improvement and concern of the mother, the children were referred to a plastic and reconstructive surgeon for surgical release of A1 pulley of FPL tendon. Surgery was performed at the age of three years. Postoperatively, the patients’ thumbs were protected with thumb-spica splints for five days and an extensive occupational therapy program was started the next day to improve active extension and motor skills. On follow-up after three months, the patients had full active extension of the IP joints and no sensory deficit in their thumbs (Fig-b, c). At present, they are four years of age and do not have any residual deformity or movement restriction. Both are studying in play group and doing well with peers in all art activities.

DISCUSSION

Bilateral PTT is a rare disorder affecting 1 in 1,000 live births according to the international statistics. It typically presents as a small, firm and tender swelling at the IP joint of the thumb around the first annular pulley (A1) supporting the FPL tendon. The swelling is named “Notta’s node” and is characterized by hypertrophy of the FPL tendon along with variable contracture of the IP joint. PTT is differentiated from fracture with or without dislocation, congenital absence of the extensor pollicis longus tendon, and arthrogryposis. A simple diagnostic test for PTT involves flexing the metatarsophalangeal joint and passively extending the IP joint. If IP joint can be extended, in combination with the above-mentioned symptoms, the diagnosis is definitely a PTT. The majority of patients with trigger-locked thumbs present with locked thumbs in flexion rather than triggering and do not present at birth but later in infancy.

The understanding of the natural history and management of PTT has improved over the last two decades. It represents a distinct condition and should not be treated like adult trigger thumb. The treating options lie between observation, serial splinting, and surgical release. Steroids are not indicated for use in children due to hormonal alterations which may inhibit growth. Observation alone may resolve PTT, although resolution may take several years. Splinting corrects contracture in 33% of patients after 12 weeks, however, pediatric patient's compliance with splinting is difficult to maintain and should be monitored frequently. If extension splinting does not resolve the contracture within 3 months, surgery is indicated. Surgical release of the A1 pulley resolves trigger thumb in more than 95% of patients. If there is a non-reducible contracture at the outset, surgical release of the pulley is deemed necessary as the first mode of management.

Surgical management of PTT with isolated release of the A1 pulley has been associated with high recurrence rates. Awareness of the anatomic factors that may contribute to triggering in the pediatric finger and willingness to explore and address other involved components of the flexor mechanism can prevent surgical failure. Other complications include nerve injury, incomplete release, and bowstringing of the tendon. Patients achieve the best results when triggering is resolved before three years of age. Older patients require more time and therapy to develop functional skills. Long term follow-up following surgical release has shown a small degree of hyperextension and decreased movement at the IP joint in a few patients; however, functional capabilities were not affected.

CONFLICT OF INTEREST

The authors of this study reported no conflict of interest.

REFERENCES