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Case Report

Congenital bilateral trigger thumb in 3 years old girl: A case report

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1. Introduction

Neglected congenital bilateral trigger thumb is an uncommon anomaly in children. Its management is still controversial, ranging from observation to extensive surgical release. The incidence of pediatric trigger thumb is 3 per 1000 live births [1]. We report a rare case presentation of bilateral trigger thumb with a brief review of literature. Children with trigger thumb usually present after the parents notice the child unable to extend his thumbs. Untreated trigger thumb can cause serious functional and aesthetic deficits as the childs develops. Fortunately multiple treatment options exist. The optimal treatment window occurs from infancy to age three. Observation and splinting are usually indicated for infants and toddlers. If splinting does not provide resolution, or if the child remains symptomatic after age three, then surgery to release the A1 pulley is indicated.

2. Etiology and epidemiology

In a Japanese population, the reported incidence of trigger thumb by age 1 year is approximately 3.3 cases per 1000 live births [2]. Because evidence exists to suggest differences in incidence among ethnic groups, it is unclear whether the incidence among Japanese children is universally applicable [2]. Although trigger thumb is not believed to be a genetic condition, there have been reports documenting an autosomal dominant inheritance pattern with variable penetrance [1]. Historically, trigger thumbs in infants and toddlers were thought to be congenital based on interviews with patients' families that suggested that the deformities were present at birth. However, multiple studies support the belief that trigger thumb is an acquired not congenital condition. Slakey and Hennrikus reported that no cases of thumb triggering or Notta nodules were identified in a study of 4719 consecutive neonates [3]. Similarly, no congenital cases were identified by Rodgers and Waters in a study of 1046 infants during neonatal hospitalization [4]. Kikuchi and Ogino in a study of 1116 infants during the first two weeks of life [2], or by Moon et al. in a prospective study of 7700 infants [5]. Collectively, 14,581 newborns were examined, and the authors failed to identify a single case of congenital trigger thumb. Therefore, the term congenital trigger thumb is a misnomer and its use is discouraged. Currently, the etiology of acquired pediatric trigger thumb remains unknown. Some authors have suggested that the constant flexed position of the thumb during the prenatal and neonatal periods results in collagen degeneration and synovial proliferation, which produces a FPL nodule and thickening of the tendon sheath. Although a large systematic histological study has not yet been performed, Buchman et al. reported the results of electron microscopic analysis of longitudinal sections of the A1 pulley and Notta nodule in nine pediatric patients with trigger thumb [17]. The authors observed large amounts of mature collagen and fibroblasts but detected no degenerative or inflammatory changes in the tendon or sheath. They concluded that the presence of a large number of fibroblasts and abundant mature collagen without inflammatory or degenerative changes does not support an infectious, inflammatory, or degenerative etiology [1].

3. Natural history

The natural history of pediatric trigger thumb has been difficult to characterize [6]. Most published natural history studies are small retrospective case series in which the age at presentation and of management varies widely [7]. Dinham and Meggitt retrospectively reviewed 105 patients (131 trigger thumbs), with a mean age of 2 years. Twenty-six thumbs were treated nonsurgically, and 105 were treated with surgical release of the A1 pulley following variable periods of observation. In 19 of 26 thumbs in the nonsurgical group, the lesion resolved spontaneously within 12 months [8].

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Although our understanding of the natural history of pediatric trigger thumb has improved, limitations remain. Most of the available literature is composed of smaller retrospective cases with limited follow-up. In general, only a subset of patients presents for evaluation and treatment, raising concerns of selection bias, particularly if children with the most severe flexion contractures preferentially receive surgical treatment. Spontaneous resolution is often defined as full IP (Interphalangeal) joint extension rather than the hyperextension commonly present on the unaffected, contralateral side. Furthermore, loss of IP joint flexion, which may be the result of incarceration of the Notta nodule distal to the A1 pulley, and compensatory MCP (Metacarpo phalangeal) hyperextension have not been uniformly reported. Additional prospective evaluation of large numbers of children should help to address these lingering issues.

4. Treatment

4.1. Nonsurgical

Nonsurgical management of pediatric trigger thumb includes passive extension exercises; however, the efficacy of these exercises has not been established. Watanabe et al. reported results of passive thumb IP joint extension exercises performed by the mothers of 48 children with 60 trigger thumbs. Mean age of the children at initial diagnosis was 26 months. Although the investigators reported that 56 of 58 thumbs (96%) achieved a "satisfactory" result, motion remained abnormal in 34 thumbs (59%) at final follow-up. In addition, because there was no control group, it is unclear whether passive stretching produced more improvement than did observation alone [9].

Published outcomes of extension splinting for pediatric trigger thumb are also inconclusive.

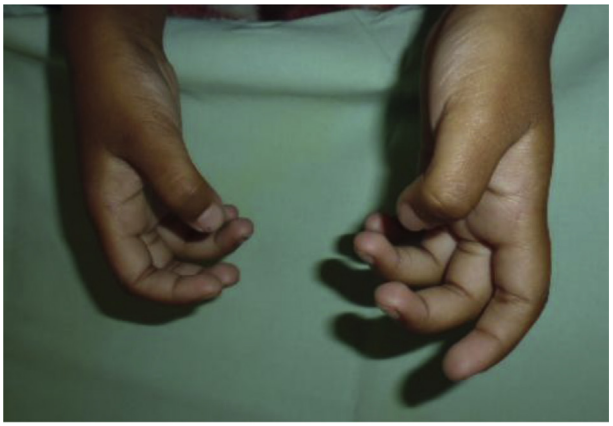
The splints were applied continuously for 6–12 weeks (mean, 11.7 weeks) before transition to night time splinting. The authors reported improvement in 71% of splinted thumbs compared with 23% of thumbs in the control group ($P < 0.05$). Although this difference was statistically significant, normal motion was restored in only 39% of splinted thumbs [10]. Currently, the role of nonsurgical management remains unclear, and the generalizability of these studies to other geographic areas and ethnic groups is unknown.

4.2. Surgical

Open surgical release of the A1 pulley of the thumb is effective in restoring IP joint motion with minimal risk of neurovascular injury, infection, and persistent or recurrent triggering. Most studies estimate that the risk of inadequate flexor tendon sheath release or recurrence is low [11]. Dunsmuir and Sherlock reported a recurrence rate of 4% in a study of 200 trigger thumbs treated surgically [12]. The authors reported that younger patients (eg, aged <36 months) may be at highest risk of recurrence. Although rare, surgical site infection is a potentially devastating complication of surgical treatment. Dinham and Meggitt reported on a series of 105 children (131 trigger thumbs) treated with open A1 pulley release [8].

One hundred thumbs regained full IP joint motion following one operation. Two cases required reoperation due to inadequate release of the A1 pulley (one patient) and surgical site infection (one patient). Three patients experienced residual IP flexion contracture >15° despite "what appeared to have been an adequate release." These patients underwent surgery at 4–6 years of age, prompting the authors to recommend surgical release by age 3 years. However, this recommendation has not been universally supported. Han et al. 26 retrospectively reported the results of open Al pulley release in 23 children (31 trigger thumbs) with a mean age of 7.5 years [13].

The authors noted that patients with unilateral involvement had no evidence of asymmetric MCP joint hyperextension. In a study of



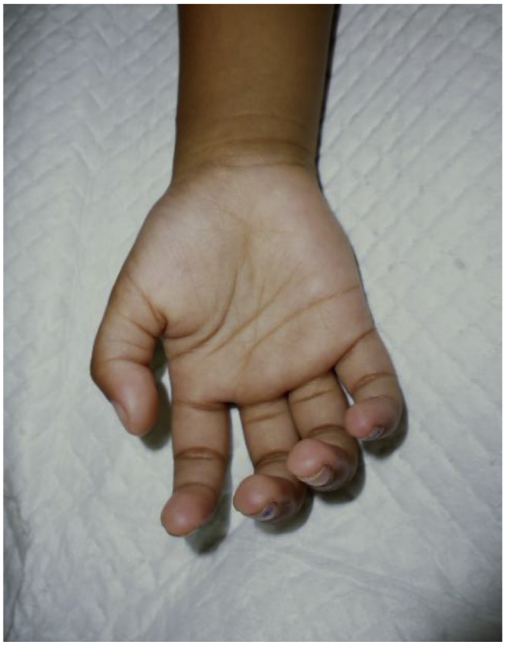


Fig. 1. Flexion deformity at interphalangeal joint (IPJ) and metacarpophalangeal joint (MCPJ) of trigger thumb in a child.