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Case Report

Neglected Congenital Bilateral Trigger Thumb: A Case Report

Thomas Erwin CJ Huwae[1], Johan Bastian[2] and Rudy[3]

[1]Staff of Orthopaedic and Traumatology Department, Faculty of Medicine Brawijaya University, Saiful Anwar General Hospital Malang, Indonesia
[2]Staff of Orthopaedic and Traumatology Department, Kanjuruhan General Hospital, Kepanjen, Malang, Indonesia
[3]Resident of Orthopaedic and Traumatology Department, Faculty of Medicine Brawijaya University, Saiful Anwar General Hospital Malang, Indonesia

Abstract

Neglected congenital bilateral trigger thumb is an uncommon anomaly in children. Its management is controversial, ranging from observation to extensive surgical release. We report a case presentation of bilateral trigger thumb with a brief review of literature.

Case Report: A 3 years old girl presented with fixed flexion deformity of interphalangeal joints of both thumb. We diagnosed as neglected bilateral trigger thumb and released of bilateral A1 pulleys.

Methods: The operation was performed under general anaesthesia. A transverse incision was made over the nodule in the tendon of flexor pollicis longus. The A 1 pulley was divided longitudinally. The tendon of flexor pollicis longus was then delivered into the wound to break down any adhesions. The thumb was assessed for full extension at the interphalangeal joint.

Results: All thumb MP (metacarpophalangeal) joints were stable with less than 20^0 of passive hyperextension.The range of motion of MP and IP was symmetric for both thumb. The children were allowed to mobilise the thumb freely within the dressings. There is no complication occur with our patient.

Keywords: Neglected; Congenital trigger thumb; Bilateral trigger thumb; A1 pulley; Case report

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 develop. 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.

Etiology and Epidemiology

In a Japanese population, the reported incidence of trigger thumb by age 1 year is approximately 3.3 cases per 1,000 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 4,719 consecutive neonates [3]. Similarly, no congenital cases were identified by Rodgers and Waters in a study of 1,046 infants during neonatal hospitalization [4]. Kikuchi and Ogino in a study of 1,116 infants during the first two weeks of life [2], or by Moon et al in a prospective study of 7,700 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 [1]. 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].

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 non surgically, 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].

Although our understanding of the natural history of pediatric trigger thumb has improved, limitations remain. Most of the available

***Corresponding author:** Rudy, Resident of Orthopaedic and Traumatology Department, Faculty of Medicine Brawijaya University, Saiful Anwar General Hospital Malang, Indonesia, Email: rudy_medstud@yahoo.com

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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 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 hyperextension have not been uniformly reported. Additional prospective evaluation of large numbers of children should help to address these lingering issues.

Treatment

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 to 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.

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 to 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 A1 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 21 pediatric patients (30 trigger thumbs) treated with open A1 pulley release, McAdams et al reported that 23% of patients experienced loss of IP joint motion by a mean 15.1-year follow-up, despite the fact that full motion was obtained in the immediate postoperative period. In addition, the investigators noted that 17.6% of patients experienced abnormal MCP hyperextension. No correlation was found between IP joint stiffness or MCP joint hyperextension with patient age, suggesting that there is no optimal time for surgical intervention [14].

Because a few small series have reported that the safety and efficacy of percutaneous techniques that use hypodermic needles is comparable to the safety and efficacy of open techniques, some surgeons have advocated the use of percutaneous techniques for management of trigger thumb. However, one study reported a recurrence risk of up to 10% associated with these techniques, suggesting that these small series may be insufficiently powered to detect differences in safety and efficacy [15]. Because of general anesthetic is required for percutaneous A1 pulley release in children, there may be no advantage to using these techniques to manage pediatric trigger thumb.

Case

A 3 years old girl presented with fixed flexion deformity of interphalangeal joints of both thumb (Figure 1). We diagnosed as neglected congenital bilateral trigger thumb and had been performed released of bilateral A1 pulleys.

Figure 1: Flexion deformity at interphalangeal joint (IPJ) and nodule at metacarpophalangeal joint (MPJ) of trigger thumb in a child.

Methods

The operation was performed under general anaesthesia. A transverse incision was made over the nodule in the tendon of flexor pollicis longus. The A 1 pulley was divided longitudinally. The tendon of flexor pollicis longus was then delivered into the wound to break down any adhesions (Figure 2 and Figure 3). The thumb was assessed for full extension at the interphalangeal joint.

Classification

Watanabe classification [9, 10]:

Stage 0: Only mass is palpable on MCP Joint without restriction of motion

Stage 1: The thumb IP Joint can actively flexed or extended with snapping

Stage 2: The locked IP Joint can be passively flexed or extended with snapping

Stage 3: The locked IP Joint cannot be passively flexed or extended

Result

After 1 month follow up, this patient had all thumb MP (metacarpophalangeal) joints were stable with less than 20^0 of passive hyperextension. The range of motion of MP and IP was symmetric for both thumb. The children were allowed to mobilise the thumb freely within the dressings.

Discussion

Trigger thumb in children is relatively uncommon and the etiology remains unclear. Some believe the condition to be congenital while others consider it to be acquired. Our patient undergoes operation for her bilateral trigger thumb. The result is that both of her thumbs were able to have complete active extension of IP joint post operatively (Figure 4).

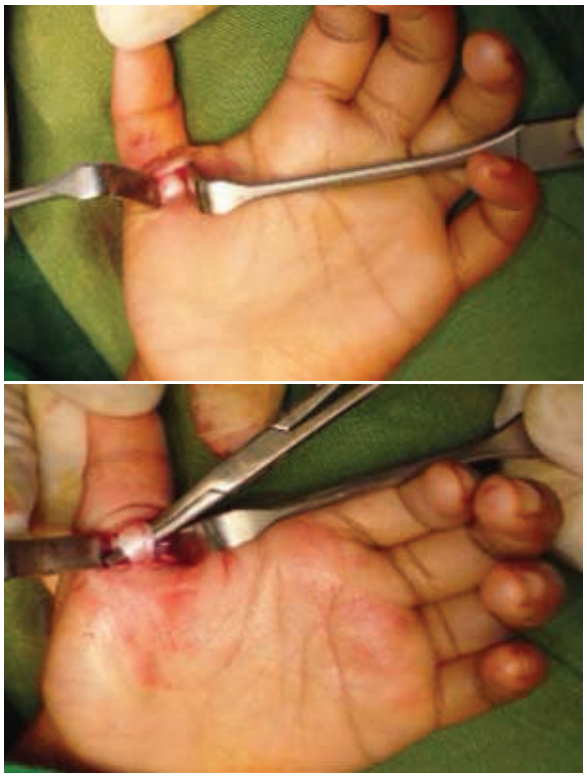

Figure 2: The FPL tendon of left thumb at surgery shows definitive enlargement (arrow) proximal to the A1 pulley.

Figure 3: The FPL tendon of right thumb at surgery shows definitive enlargement (arrow) proximal to the A1 pulley.

Conclusion

- It is very important to identify trigger thumb in children as earlier as possible.
- In children, operative treatment has been found to be satisfactory.

Ethical Approval

Institutional review board approval was exempt from our institution because all data were collected from clinical records and imaging system for routine pre-post-operative planning and follow up.

Author Contribution

Thomas Erwin CJ Huwae, Johan Bastian and Rudy wrote this paper; Johan Bastian and Rudy performed the surgery; Thomas Erwin, Johan Bastian decided for study design and data analysis. All authors read this paper.

Conflict of Interest

The authors declare that there is no conflict of interest regarding the publication of this paper.

Guarantor

Thomas Erwin CJ Huwae and Johan Bastian

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

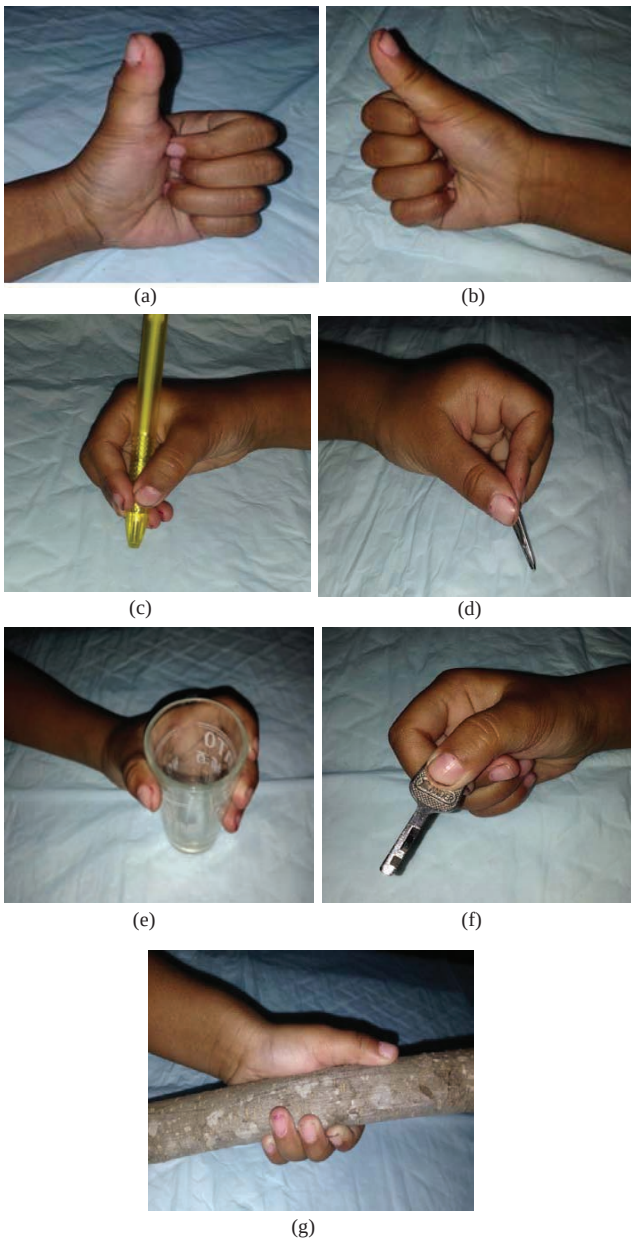

Figure 4: Patient able to use both of her thumb for daily activities without difficulties. (a,b) extend thumb, (c) tripod, (d) pinch, (e) grasping, (f) key, (g) power grip

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