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Presentation and aetiology of paediatric trigger finger: a systematic review

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Abstract

Paediatric trigger finger is a rare condition distinct from paediatric trigger thumb and adult trigger digits. We performed a systematic review of paediatric trigger finger presentation and aetiology in order to guide workup and management. Fifty-one studies with 193 patients and 398 trigger fingers were included. Most patients had a single, unilateral trigger finger (54%). Fifty-five patients (29%) had an underlying condition, such as mucopolysaccharidosis; these cases appeared to be associated with multiple or bilateral trigger fingers or with carpal tunnel syndrome. All patients with mucopolysaccharidosis were treated surgically. Conservative management was reported in 33% of all patients, and two-thirds of these did not need further intervention. Patients undergoing surgical release infrequently had recurrence of triggering (6%). We propose an algorithmic approach for patients presenting with paediatric trigger finger. Presence of bilateral or multiple trigger digits or concomitant carpal tunnel syndrome should raise suspicion for an atypical underlying pathology.

Keywords

Paediatric, trigger finger, systematic review, review

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Introduction

In contrast to trigger digits in adults and paediatric trigger thumb, paediatric trigger finger is a rare entity, and its pathophysiology is poorly understood (Shah and Bae, 2012). This makes it difficult to determine appropriate management, including whether further investigations and surgical intervention are needed. In paediatric trigger thumb, triggering is believed to be due to a size mismatch between the flexor pollicis longus tendon and the A1 pulley, either from tendon swelling or a thickening of the pulley (Bauer and Bae, 2015). In adult trigger finger, high pressures at the proximal edge of the A1 pulley gradually results in hypertrophy and fibrocartilaginous metaplasia at the tendon–pulley interface, eventually leading to a size mismatch, especially if the tendon becomes inflamed (Akhtar et al., 2005). In both paediatric trigger thumb and adult trigger digit, many cases resolve spontaneously and do not need surgical intervention (Akhtar et al., 2005; Bauer and Bae, 2015).

The proposed aetiology of paediatric trigger finger may be associated with trauma, anatomic anomalies or underlying conditions, such as mucopolysaccharidosis and diabetes (Hansen and Battista, 2007; Holt et al., 2013; Yosipovitch et al., 1990). Particularly in such cases, the aetiology of the trigger fingers should be taken into consideration and management tailored accordingly (Schaverien and Godwin, 2011; Shah and Bae, 2012).

We conducted a systematic review of paediatric trigger finger to determine its key features and associated pathophysiology to streamline investigations, conservative management and surgical intervention.

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Methods

This systematic review was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA). We included English-language case reports, case series, clinical trials, cohort studies and comparative studies reporting trigger finger in patients 16 years of age or younger. An experienced information specialist (RP) developed comprehensive searches for the concepts of paediatric populations and trigger finger (Online Table S1). We searched MEDLINE through PubMed, Embase, Cochrane Central Register of Controlled Trials and Cumulative Index of Nursing and Allied Health from inception to January 2020. We scanned the reference lists and citations of relevant studies identified for further references. Our search was completed 23 January 2020.

Two authors (ALW, MEW) identified studies based on title and abstract screening. Disagreements were resolved by discussion with a third author (MJW). Articles were independently assessed for inclusion based on full-text screening by two authors (ALW, MJW), and disagreements were resolved through discussion with another author (MEW). Exclusion criteria included adults, non-English language presentations, conference abstracts, review articles, papers reporting on trigger thumb only, forearm fractures and other hand trauma.

We included studies in accordance with existing tools for determining case reports' and case series' methodological quality (Murad et al., 2018), as well as validity and educational value (Pierson, 2009) (Online Table S2). The data extracted included: study setting; methodology; population, with participant demographics and baseline characteristics; aetiology of trigger finger, affected digit(s); details of the intervention and control conditions; suggested mechanisms of intervention action; recruitment and study completion rates; and outcomes and times of measurement.

Results

The PRISMA diagram shows the screening process (Online Figure S1). Fifty-one studies were included in the final review, including 26 case reports, 20 case series and five retrospective cohort studies. Given the nature of the included studies, quality was generally low (Online Table S2). Full details of the included case reports are summarized in Online Table S4, and details of case series and retrospective cohort studies are summarized in Online Table S5. The included studies varied greatly in terms of which specific details were presented.

In total, 398 trigger fingers were reported in 193 patients. The age of presentation ranged from birth to 16 years old. The reported total duration of triggering also had a range from birth to 16 years. The most commonly involved fingers were the long (44%) and ring (30%) fingers. Four publications did not adequately report which specific fingers were affected, accounting for 56 (14%) of the trigger fingers (Brinkman et al., 2019; Cardon et al., 1999; Grigull et al., 2011; Van Heest et al., 1998).

The majority of patients (54%) presented with a single triggering digit on one hand (Online Table S6). Ten patients (5%) had a history of trauma. Five patients (3%) presented with a calcific mass. Fifty-five (29%) patients had an underlying condition, such as mucopolysaccharidosis or other genetic syndrome. Such cases tended to have bilateral trigger fingers, concomitant carpal tunnel syndrome or dysmorphic features and rarely had an isolated unilateral trigger finger. However, given differences in reporting it was not possible to quantify this association. Studies including patients with multiple or bilateral trigger fingers are shown in Online Table S7.

Radiographs of paediatric trigger fingers often failed to reveal any abnormality but were used in cases with a history of trauma. At least 64 cases (33%) were initially treated with splinting, physiotherapy, observation or other conservative management. For 26 of these 64 patients, triggering failed to resolve and later required surgical release. Multiple triggering digits appeared to be associated with failed conservative management. There was a single case report where a steroid injection improved the patient's symptoms (Hamada et al., 2011). One study showed a higher incidence of spontaneous resolution with splinting than without (Shiozawa et al., 2012). No study directly compared splinting with immediate operative treatment.

Among all patients and regardless of underlying aetiology, 122 (63%) had a surgical release of the A1 pulley. Less commonly, there was additional excision of visible masses, abnormal flexor digitorum superficialis (FDS) slips or adhesions between the FDS or flexor digitorum profundus (FDP); but these details were inconsistently reported. The most common intraoperative findings were the presence of a fibrous nodule (20 patients; 16%) or thickening of the FDS or FDP tendons (12 patients; 10%). Also, adhesions between FDS or FDP (four patients; 3%), abnormal slips of FDS (two patients; 2%) or thickened A1 pulley (seven patients; 6%) were reported. There were seven recurrences described among patients treated surgically (6%), but the included studies had inconsistent follow-up periods, ranging from 1 month to 10 years, or did not report follow-up at all.

Discussion

Despite its low incidence, there have been numerous reports describing paediatric trigger finger. A previous review by Womack et al. (2018) did not offer guidance regarding which features should raise concern for associated conditions or when to initiate further investigations and referrals. It also had important methodological limitations. It only searched a single database (MEDLINE) and did not review the noteworthy body of literature consisting of paediatric trigger finger case reports (Womack et al., 2018). Though often excluded from reviews due to 'poor quality' because by their nature there is no randomization or controls for treatment, case reports remain useful in understanding rare conditions and complications.

Including all case reports and case series, we synthesized all available information to provide suggested management (Table 1). Most patients had idiopathic triggering of a unilateral single digit. Only 29% of patients had a genetic abnormality, most commonly mucopolysaccharidosis and related lysosomal storage diseases. Such cases appeared to present at a younger age and often with multiple trigger digits, concomitant carpal tunnel syndrome or dysmorphic features. Only one study focused on paediatric trigger finger with concomitant diabetes (Paaske et al., 1995). In one series of 250 primarily adult patients with a history of juvenile diabetes, trigger finger was present in 13 patients, who ranged in age from 14 to 38 years old (Yosipovitch et al., 1990). A link between juvenile idiopathic arthritis and paediatric trigger finger has been previously suggested (Schaverien and Godwin, 2011; Shah and Bae, 2012), however, we only found this reported in a single case series (Paaske et al., 1995) and a conference abstract (Batticciotto et al., 2016).

Paediatric trigger finger was reported following relatively minor, repetitive trauma in 5% of patients. This was usually associated with an older age at presentation of at least 7 years and in an isolated digit. Regardless of a history of trauma, in patients where there was no associated genetic condition, physical examination was generally unremarkable apart from catching of the digit, though a nodule could sometimes be palpated.

Though not included in this review, multiple reports described reduced digit range of motion associated with hand or forearm fractures, a presentation that mimics trigger finger (Fernandez and Segal, 2007; Furuya et al., 2019; Harryman and Jordan, 1990; Lee et al., 2015; Ooi and Toh, 2001; Piquilloud et al., 2011; Rayan and Hayes, 1986; Rodríguez-Vega et al., 2013; Shaw and Murphy, 1996; Shively and Lesnick, 1982; Song et al., 2012; Walker et al., 2012).

In paediatric trigger finger, appropriate workup should be based on suspicion of underlying pathology. Plain film radiography is often normal but may show joint abnormalities or bony masses (Bauer and Bae, 2015; Oliveira et al., 2017). Ultrasound is increasingly used in evaluating paediatric trigger thumb and similarly can be used in paediatric trigger finger to identify nodules or thickening of the flexor tendons or the pulleys (Batticciotto et al., 2016; Bauer and Bae, 2015). While MRI is an excellent modality for characterizing soft tissue lesions, it may require sedation in paediatric patients and is unlikely to alter management. If there is suspicion for mucopolysaccharidosis, urine should be screened for glycosaminoglycans and, if positive, should be followed by referral for formal genetic testing (Holt et al., 2013). To avoid missing a subtle median neuropathy, electromyography and nerve conduction testing should be ordered for these patients (Shah and Bae, 2012).

A trial of conservative management is reasonable if there is no suspicion for an underlying metabolic or anatomical cause. Though difficult to quantify, approximately one-third of conservatively managed cases either failed to resolve or later required surgical release (Bhaban et al., 2017; Cardon et al., 1999;

Table 1. Suggested management algorithm for paediatric trigger finger. Specific workup and referrals should be guided by suspected underlying condition (i.e. mucopolysaccharidosis, arthritis).

Child presenting with trigger finger	
Single, unilateral digit	Multiple or bilateral involved digits
1. History of trauma: consider imaging to rule out structural cause, treat any underlying structural abnormality or trial conservative management	1. Have a high suspicion for an underlying condition
2. No history of trauma: trial conservative management	2. Initiate workup and referrals
3. If no resolution with conservative management, proceed with surgical treatment	3. Surgical treatment

Case and Leslie, 1998; Hamada et al., 2011; Luna et al., 2013; Moon et al., 2001; Pargali and Habibzadeh, 2011; Salati, 2014; Shiozawa et al., 2012; Tsuyuguchi et al., 1983).

Multiple surgical treatment algorithms have been proposed, and all follow a similar strategy (Bauer and Bae, 2015; Schaverien and Godwin, 2011; Shah and Bae, 2012; Womack et al., 2018). Surgery should start with an incision designed over the A1 pulley. Following release of A1, the finger should be ranged by passive extension and flexion via traction on the flexor tendons proximal to the A1 pulley to see if triggering persists. Careful inspection of the flexor tendons at that level should be done to identify any nodules, abnormal FDS slips, abnormal lumbrical insertions or adhesions between the FDS and FDP tendons. Any abnormal structures or masses should be excised. If there is still triggering, the incision should be extended distally to inspect the A2 and A3 pulleys. In the case of mucopolysaccharidosis, it is still reasonable to start with a course of conservative management and then proceed to operative treatment if there is no improvement in 6 months (Holt et al., 2013). The surgical approach is the same, except that release of both A1 and A3 is done initially, a slip of FDS is excised and carpal tunnel release is performed concurrently.

Postoperative management usually involves a soft dressing and allows early motion since immobilization of the hand following can increase stiffness (Shah and Bae, 2012). Some surgeons, however, prefer a short course of splinting postoperatively (Fox et al., 2017; Luna et al., 2013; Vilai and Vechmamontien, 2019). Based on our included studies, the incidence of recurrence after surgical treatment appears low even when there is an underlying medical condition. However, since many of these studies had a follow-up period of less than 1 year or did not specify their duration of follow-up, it is unclear if longer term (i.e. greater than 1 year) incidences of recurrence are comparable.

The number of patients in this review is small; and assessment measures, treatments and follow-up periods were not standardized. Specific details reported in the included studies were very diverse, and useful features were oftentimes not reported. Often patient comorbidities, ages and treatment specifics were reported separately from presentation and thus difficult to correlate. Publication bias may be present. Comparing different treatment modalities or drawing robust conclusions is therefore difficult. However, since there are very few large series and no prospective trials on paediatric trigger finger, case reports remain a major part of the existing literature and point in a similar direction of treatment.

The underlying aetiology of paediatric trigger finger is usually idiopathic. However, awareness of other documented causes is essential for proper identification and treatment. We suggest that the presence of bilateral or multiple trigger fingers or concomitant carpal tunnel syndrome should raise suspicion for underlying pathology.

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Contributions ALW initiated this systematic review, drafted the research protocol, collected data, and wrote much of the manuscript. MJW collected data and wrote much of the manuscript. RP performed the literature search, and assisted with preparing the systematic review protocol and editing of the manuscript. MEW settled disputes around inclusion of articles in the systematic review, and was involved in preparing the research protocol and writing the manuscript.

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Supplemental material Supplemental material for this article is available online.

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