SCIENTIFIC ARTICLE

The Natural History of Pediatric Trigger Thumb in the United States

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Purpose Surgical release of pediatric trigger thumbs has been recommended as definitive treatment, although controversy exists over the natural history of pediatric trigger thumb. This study sought to evaluate the incidence of spontaneous resolution of pediatric trigger thumb and the factors that may influence resolution.

Methods Pediatric patients were prospectively enrolled by a single surgeon from August 2009 to July 2015. All patients were initially treated with observation. They were followed annually and we collected pain scores (Parental visual analog scale), subjective dysfunction as perceived by parents, and physical examination information including the presence of flexion contracture of the thumb interphalangeal (IP) joint, thumb metacarpophalangeal joint laxity, and medial-lateral plane IP joint angular deformity. A competing risk framework was used to estimate the cumulative incidence at 5 years from the initial visit, and a subdistribution hazards model was used to compare patient characteristics with spontaneous resolution. Hazard ratios (HRs), 95% confidence intervals (95% CIs), and *P* values were reported.

Results Seventy-eight patients (93 thumbs) with an average age of 20 months ± 1 year (mean ± SD) were enrolled at the first clinic visit and followed for 4.3 years (interquartile range, 3.1−5.5 years). At 5 years from the initial visit, 32% (95% CI, 20%−43%) of thumbs had resolved spontaneously, and 43% (95% CI, 30%−54%) had elected to proceed to surgery. Among those who had surgery, the median time to surgery was 4.1 years (interquartile range, 2.9−5.3 years). Bilateral thumb involvement increased the risk of surgery (subdistribution HR, 2.38; 95% CI, 1.23−-4.6). Each degree increase in initial IP joint flexion decreased the occurrence of spontaneous resolution by 3% (subdistribution HR, 0.97; 95% CI, 0.94−0.99). Initial IP joint flexion 30° or less was associated with spontaneous resolution at 3 years (sensitivity, 0.73, 95% CI, 0.37−1.00; specificity, 0.70, 95% CI, 0.38−0.94; positive predictive value, 0.18, 95% CI, 0.13−0.41; negative predictive value, 0.76, 95% CI, 0.71−0.83; area under the curve, 0.78), whereas only 2.5% (95% CI, 0.4%−17%) of patients with an IP joint flexion greater than 30° resolved.

Conclusions A third of pediatric trigger thumbs resolved spontaneously, but most parents desired eventual surgical release. Patients with IP joint flexion contractures greater than 30° at baseline often lacked spontaneous resolution at 3 years and may be reasonable early surgical candidates. *(J Hand Surg Am. 2021;46(5):424.e1-e7. Copyright © 2021 by the American Society for Surgery of the Hand. All rights reserved.)*

Type of study/level of evidence Prognostic II.

Key words Congenital trigger, natural history, pediatric trigger, trigger finger, trigger thumb.

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Trigger thumb in the pediatric patient is usually first noticed by the parent as a lack of full extension.[1] The etiology of pediatric trigger thumb is unclear with many authors proposing both hereditary and acquired causes.[2–4] The ultimate result is a developmental size mismatch between the flexor pollicis longus tendon and its sheath. Surgical release of pediatric trigger thumbs has been recommended as definitive treatment, although controversy exists over the natural history of pediatric trigger thumb.[5–8] A number of studies have looked at the natural history of pediatric trigger thumb with spontaneous resolution rates ranging from 0% to 96% over a median duration of follow-up ranging from 6 to 48 months.[9–13] In a recent study by Baek and Lee,[14] 76% of the pediatric trigger thumbs resolved over an average follow-up of 5 years. We believe that a 76% resolution rate is not the experience of most pediatric hand surgeons in the United States because most are surgically treated prior to 5 years. However, the mostly nonsurgical cohort presented by Baek and Lee[14] with extended follow-up is compelling and warranted a reevaluation of our experience. Therefore, the primary goal of this study was to prospectively evaluate the natural history of pediatric trigger thumbs presenting at our institution to determine the incidence of spontaneous resolution. Our secondary goals included whether baseline measurements of thumb interphalangeal (IP) joint flexion contractures, IP joint angular deformity, metacarpophalangeal (MCP) joint laxity, and other baseline characteristics predict the incidence of spontaneous resolution.

MATERIALS AND METHODS

This institutional review board–approved prospective study included all pediatric patients with a trigger thumb presenting to a single hand surgeon's (D.T.H.) office between August 2009 and July 2015 who were willing to be followed in this way. We explained the purpose of the study to the parents along with the risks and benefits of surgical release and nonsurgical treatment. Nonsurgical management for this cohort of patients included observation only. Participants had the option of leaving the study at any point during the study period and could choose other treatment options. They were assured that putting off surgery until later would not adversely affect the surgical result. Patients with less than 2 years of follow-up since the onset of symptoms were excluded.

The enrolled patients were followed annually during the study period. The age of the patient, sex, side involved, family history, age that deformity was first noticed, and any other conditions affecting the involved upper extremity were collected at the first visit. Findings in the history that were recorded included pain, dysfunction, and triggering of the thumb. A visual analog scale was used to capture the degree of patient discomfort and difficulty with daily activities over the last 6 months (parent proxy). The presence of triggering was recorded and defined as clicking and/or popping of the thumb with a reducible flexion contracture. Physical examination measurements, including passive motion of the IP joint of both thumbs with the MCP joint and wrist at neutral, thumb MCP joint laxity (hyperextension), and presence of IP joint angular deformity in the medial-lateral plane, were obtained at the first visit and all subsequent visits.

All patients were followed until they were either nearly resolved indicating a flexion contracture less than 5° and no pain, in which case parents and surgeon agreed that no surgery was needed, desired surgery, completed 5-year follow-up, or refused further follow-up.

Statistical methods

Baseline variables were summarized at both the patient (n = 78) and the thumb level (n = 93). Continuous variables were summarized as mean and SD, median, interquartile range (IQR), and range (minimum to maximum). Categorical variables were summarized as count and percentage. Patients were grouped based on their final resolution status into the following 3 groups: (1) spontaneously resolved: no triggering and at most minimal flexion contracture (<5°) and not requesting surgery, (2) surgery: patient/family elected to undergo surgical intervention, and 3) nonresolved: flexion contracture greater than 5° and not requesting surgery.

These data were analytically challenging for several reasons. First, our interest was the event of spontaneous resolution, which could take several years to occur, and a patient may have been lost to follow-up prior to resolution. Second, a patient may have received surgery to resolve the contracture at any time during our study. Finally, our sample size prohibited a rigorous multivariable analysis owing to a limited number of resolutions (n = 29). The first issue required the use of survival methods to handle loss to follow-up. However, in a conventional survival analysis, patients who are lost to follow-up are censored (their data are used until the point when they are lost to follow-up). The assumption for censored patients is that they are still at risk for the event to occur and their event occurrence rate is similar to the rate among those who are still observed. This assumption was

reasonable for subjects lost to follow-up, but potentially problematic for subjects who were treated with surgery. If surgery patients were treated as censored, then the probability of spontaneous resolution would potentially be overestimated as a result. Thus, to handle the second issue, competing risk models were used in which surgery was treated as an event that competed with spontaneous resolution. The start time for each patient was his or her first clinical visit and we compared patient characteristics collected at baseline with spontaneous resolution in univariable competing risk models, in which predictors included sex, age of onset, involved side, bilateral involvement, family history, follow-up time, triggering presence, patient discomfort over the last 6 months, difficulties with daily use over the last 6 months, IP joint angulation, IP joint flexion and MCP joint hyperextension. We used a Fine-Gray subdistribution hazard modeling approach for our competing risk analysis, which was implemented using the *crr* function in the cmprsk R package. Univariable hazards ratios (HRs), 95% confidence intervals (95% CIs), and *P* values were reported for both the subdistribution hazards of the spontaneous resolution event and the subdistribution hazards of surgery. The interpretation of these HRs is the relative change in the rate of spontaneous resolution among those who have not yet experienced resolution. Because spontaneous resolution was a positive outcome for the patient, we describe hazards of resolution as occurrence in discussing the results. The *cuminc* function in the cmprsk R package was used to plot the cumulative incidence curves for resolution and surgery in a competing risks framework. We also reported the cumulative incidence of resolution and surgery and corresponding 95% CIs at 1, 3, and 5 years from their initial visit.

We used the competing risks model for IP joint flexion along with receiver operating characteristic curves to estimate whether baseline IP joint flexion could predict the probability of achieving spontaneous resolution of trigger thumb at various time points. The optimal threshold for baseline IP joint flexion to predict spontaneous resolution of trigger thumb was picked by maximizing Yudon's index (sensitivity + specificity − 1) across 10° increments of flexion ranging from 10° to 50°. Point estimates of sensitivity, specificity, positive predictive value (PPV), and negative predictive value (NPV) at this threshold were reported with 95% CIs that were estimated from 2,000 bootstrap samples. Sensitivity and specificity were considered our primary measures of diagnostic accuracy because they are relatively independent of the prevalence of spontaneous resolution and surgery. However, PPV and NPV are reported as well to assess the IP joint's flexion's performance in a clinical diagnostic setting that has a similar rate of resolution to what was observed here.

TABLE 1. Patient Level Descriptive Summary at Baseline (n = 78 Patients)

Variable	Count
Sex, n (%)	
Male	35 (45)
Involved side, n (%)	
Both	18 (23)
Left	34 (44)
Right	26 (33)
Family history, n (%)*	20 (26)
Maximum follow-up (y)†	
Mean (SD)	4.5 (1.7)
Median (IQR)	4.3 (3.1−5.5)
Range	2−8.7
Patient discomfort over last 6 mo (VAS)	
Mean (SD)	0.9 (1.7)
Median (IQR)	0 (0−1.8)
Range	0−8
Difficulties with daily use over last 6 mo (VAS)	
Mean (SD)	0.2 (0.7)
Median (IQR)	0 (0−0)
Range	0−5

VAS, visual analog scale.
*Missing values: Family history = 2.
†Total years of follow-up since date of onset. If both thumbs were involved, we report the maximum follow-up of the 2.

We compared the average IP joint flexion at baseline between our cohort and the Baek and Lee study's cohort[14] using a 2-sided, 2-tailed *t* test at the thumb level. Statistical significance was assessed at the .05 level using 2-tailed tests for all analyses. Analyses were conducted in R v 3.6.0.

RESULTS

From August 2009 to July 2015, 102 patients were enrolled at the first clinic visit (only 4 refused to be part of the study and preferred only surgery). Seventeen patients were excluded for follow-up of less than 2 years and 7 patients withdrew from the study. Thus, 78 patients (76.5%) with an average age of 20 months ± 12 months (mean ± SD) were included in the final analyses (Table 1). Eighteen

TABLE 2. Thumb Level Summary at Baseline (n = 93 Thumbs)

Variable	Resolved Naturally (n =29)	Surgery (n = 36)	Not Resolved (n = 28)	P1*	P2*
Sex, n (%)					
Male	13 (45)	21 (58)	9 (32)	.37	.15
Age of onset (y)					
Mean (SD)	1.7 (1.1)	1.5 (1.0)	1.9 (1.1)	.36	.43
Median (IQR)	1.5 (0.9–2.2)	1.3 (1.0–2.0)	1.8 (1.1–2.1)		
Range	0.1–4.5	0.0–4.0	0.0–4.3		
Involved side, n (%)					
Right	14 (48)	16 (44)	11 (39)	.68	.96
Bilateral (yes)	8 (28)	17 (47)	8 (29)	.23	.01
Family history (yes), n (%)	10 (36)	6 (17)	8 (29)	.11	.06
Follow-up (y), n (%)†					
Mean (SD)	4.7 (1.7)	4.4 (1.8)	4.4 (1.5)	.43	.05
Median (IQR)	4.4 (3.2–5.5)	4.1 (2.9–5.3)	4.4 (3.3–5.5)		
Range	2.2–8.0	2.0–8.7	2.3–7.2		
Triggering present (yes), n (%)	9 (31)	17 (47)	14 (50)	.14	.61
Patient discomfort over last 6 mo (VAS), n (%)	1.2 (2.0)	0.6 (1.1)	1.1 (1.8)		
Any patient discomfort over last 6 mo	10 (34.5)	9 (25)	13 (46.4)	.75	.1
Difficulties with daily use over last 6 mo	0.2 (0.8)	0.2 (0.9)	0.1 (0.4)	.52	.75
IP joint angulation (yes)	2(7%)	5(14%)	1(4%)	.26	.2
IP joint flexion (°)					
Mean (SD)	26.6 (16.5)	37.3 (16.4)	36.4 (15.9)	.01‡	.05
Median (IQR)	25 (20–35	35.0 (25–45)	37.5 (25.0–46.2)		
Range	0–60	13–75	0–60		
MCP hyperextension (yes), n (%)§	3 (14)	4 (2)	6 (30)	.37	.81
MCP hyperextension, n (%)				–	–
Mean (SD)	0.5 (6.7)	5.5 (12.6)	6.4 (16.4)		
IP flexion/MCP laxity Correlation	$P = .5$	rho = 0.1			

VAS, visual analog scale.
*P values are from a competing risk model in which surgery competes with natural resolution (HRs and 95% CIs are in Table 3).
†The time to event data starts at the first clinical visit for each patient because our analysis focused on how patient characteristics collected at baseline predicted surgery/resolution outcomes. The follow-up time starts at the date of initial presentation of symptoms, which in some cases was 2 years prior to the initial visit.
‡Significant.
§MCP joint hyperextension was not available if both thumbs were involved. Among thumbs with MCP joint hyperextension available (n = 60), 78% had a value of 0 (no difference from the other thumb). Thus, we dichotomized it for analysis to 1 if MCP joint hyperextension > 0, otherwise 0.

patients had bilateral thumb involvement of which 3 patients had a thumb with less than 2-year follow-up that was excluded from our analyses. Thus, 93 thumbs were prospectively followed to determine the incidence of spontaneous resolution (Table 2). The median follow-up time was 4.3 years (IQR, 3.1–5.5 years), and the range was 2 to 8 years. Within 5 years from the initial visit, 32% (95% CI, 20%–43%) of thumbs resolved spontaneously, 25% did not resolve, and 43% (95% CI, 30%–54%) had elected surgical correction. Figure 1 shows the surgical and resolution groups as a function of time. Those choosing surgery did so mainly after a median follow-up of 4.1 years (IQR, 2.9–5.3 years).

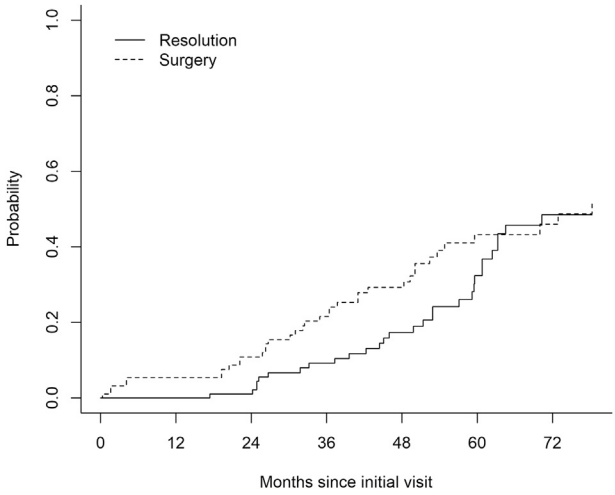

FIGURE 1: Survival curve from the surgical and resolution groups.

The IP joint flexion at initial presentation was the only variable found to be significantly associated with spontaneous resolution, where on average a 1° increase in IP joint flexion corresponded to a 3% decrease in the occurrence of spontaneous resolution (subdistribution HR, 0.97; 95% CI, 0.94−0.99; Table 3). Bilateral thumb involvement was associated with a 138% increase in the hazard of surgery (subdistribution HR, 2.38; 95% CI, 1.23−4.6). The area under the curve accuracy of IP joint flexion measured at baseline for predicting spontaneous resolution at the 3-year follow-up time point was 0.778. An optimal threshold for IP joint flexion was 30° for maximizing both the sensitivity and the specificity (Table 4). Thumbs with an IP joint flexion of 30° or less at baseline were predicted to resolve spontaneously within 3 years, whereas only 2.5% (95% CI,: 0.4%−17%) of patients with an IP joint flexion greater than 30° experienced spontaneous resolution at 3-year follow-up. The sensitivity of this test was 0.73 (95% CI, 0.37−1.00), a specificity of 0.70 (95% CI, 0.38−0.94), a PPV of 0.18 (95% CI, 0.13−0.41), and an NPV of 0.76 (95% CI, 0.71−0.83). Twenty-six percent of our patients had a family history of trigger thumb. Family history and presence of IP joint angulation (different from flexion) were not significantly associated with resolution ($P = .11$ and $P = .26$, respectively; Table 2).

Compensatory MCP joint hyperextension, potentially a result of the IP joint flexion contracture, is an often-cited rationale for surgical release.[15] Sixty unilateral patients had MCP joint hyperextension measures and 78% (n = 47) had 0° difference between their triggering and their contralateral thumbs. We did not find a significant association between MCP joint hyperextension and either spontaneous resolution of trigger thumb or the decision for surgery ($P = .37$ and $P = .81$, respectively; Table 2). A plot of MCP joint hyperextension over time shows that it is relatively stable (Fig. 2). In addition, we found no correlation between IP joint flexion and MCP joint hyperextension. Baseline measurement of discomfort, difficulty with daily use, and visual analog scale pain were not associated with surgery or spontaneous resolution (all $P > .05$; Table 2).

DISCUSSION

The literature on the resolution of pediatric trigger thumb with nonsurgical therapy shows a wide variation with rates ranging from 0% to 96% over a median duration of follow-up that ranged from 6 months to 5.3 years.[1−5,16,17] The median follow-up in our study was 4.3 years (IQR, 3.1−5.5 years), and we found that 32% (95% CI, 20%− 43%) of trigger thumbs had spontaneous resolution within 5 years from the initial visit. This differs from recent studies such as Baek and Lee,[14] which showed that 76% of pediatric trigger thumbs resolved with a median time to resolution of 4.1 years, and by Jung et al,[17] which showed 80% of pediatric trigger thumbs completely resolved by mean follow-up of 5.3 years. One reason for this difference is that surgery is a competing event with the natural resolution process—that is, subjects who receive surgery are no longer able to naturally resolve. These other studies did not report the cumulative incidence of natural resolution in a competing risk framework, which may overestimate the natural resolution incidence. Another possible reason is that the average IP joint flexion in those that did not resolve in 4 years was 27.6° ± 9.3° in the study by Baek and Lee[14] versus 36.9° ± 6° at baseline among those who had surgery in our study. The average IP joint flexion at baseline of the entire cohort also differed, being 33.8° ± 16.8° in our study versus 26.3° ± 9.9°. Thus, it is possible that our cohort had a larger initial flexion contracture of the IP joint, accounting for some of the difference in spontaneous resolution rates. Jung et al[17] did not record IP joint flexion in degrees and so the 2 cohorts could not be compared in this regard. Cultural differences and a potential difference in the definition of resolution in these other studies as well as the availability of surgery as an alternative may also contribute to the differing results.

TABLE 3. Association Between Baseline Variables and Time to Resolution/Surgery Estimated From Univariate Competing Risk Models

Variable	HR1 (95% CI)*	*P*1*	HR2 (95% CI)†	*P*2†
Sex, n (range)				
Male	0.73 (0.37−1.46)	.37	1.6 (0.84−3.05)	.15
Age of onset (y), n (range)	1.18 (0.83−1.66)	.36	0.89 (0.66−1.2)	.43
Involved side, n (range)				
Right	1.16 (0.58−2.32)	.68	0.99 (0.52−1.88)	.96
Bilateral (yes/no)	0.64 (0.3−1.34)	.23	2.38 (1.23−4.6)	.01
Family history (yes), n (range)	1.85 (0.88−3.88)	.11	0.46 (0.2−1.05)	.06
Follow-up (y), n (range)	0.92 (0.74−1.14)	.43	0.8 (0.64−1)	.05
Triggering present (yes), n (range)	0.56 (0.26−1.21)	.14	1.18 (0.62−2.24)	.61
Patient discomfort over last 6 mo (yes), n (range)‡	1.12 (0.54−2.31)	.75	0.56 (0.28−1.11)	.1
Difficulties with daily use over last 6 mo (VAS), n (range)	0.9 (0.64−1.25)	.52	0.96 (0.72−1.27)	.75
IP joint angulation (yes)	0.52 (0.17−1.63)	.26	1.8 (0.74−4.39)	.2
IP joint flexion (°)	0.97 (0.94−0.99)	.006§	1.02 (1−1.04)	.05
MCP joint hyperextension (yes)	0.6 (0.2−1.8)	.37	1.14 (0.4−3.22)	.81

VAS, visual analog scale.
*Natural resolution.
†Surgery.
‡Patient discomfort over last 6 mo variable ranged from 0 to 8 on the VAS with most people being 0. Therefore, it was dichotomized as > 0 (yes) versus 0 (no).
§Statistically significant at $P < .05$.

TABLE 4. Evaluation of IP Joint Flexion as a Diagnostic Measure of Natural Resolution of Pediatric Trigger Thumb*

Cut-Off (°)	Sensitivity (95% CI)	Specificity (95% CI)	PPV (95% CI)	NPV (95% CI)
10	0.25	0.96	0.48	0.7
20	0.37	0.88	0.2	0.73
<30	0.73 (0.37−1)	0.7 (0.38−0.945)	0.18 (0.13−0.41)	0.76 (0.71−0.83)
40	0.87	0.43	0.13	0.74
50	1	0.23	0.11	0.88

*Optimum threshold for the natural resolution outcome according to Yudon's index (sensitivity + specificity − 1) was 30° with an area under the curve of 0.778 95% CIs for optimal threshold were estimated from 2,000 bootstrapped samples.

We found that baseline IP joint flexion contracture measurements were significantly lower in the thumbs that spontaneously resolved. This is in contrast to the study by Baek and Lee,[14] which showed no difference with regards to this measurement. The establishment of an IP joint flexion threshold at initial presentation associated with lack of trigger thumb resolution is a finding unique to our study. In our study, only 2.5% (95% CI, 0.4%−17%) of patients with an IP joint flexion greater than 30° experienced spontaneous resolution at 3-year follow-up. However, the accuracy of this IP joint flexion threshold in predicting spontaneous resolution was limited with a PPV of 0.18 and an NPV of 0.76.

Age was not associated with spontaneous resolution or surgical intervention and this is consistent with prior literature.[13,14,17] Hyperextension of the MCP joint should not be used as an indication for surgical release. Bilateral thumb involvement was associated with a poorer prognosis in our cohort, as was found to be the case by Jung et al[17] as well. This could be due to the chance of 1 thumb failure leading to surgical intervention being done on both thumbs simultaneously.

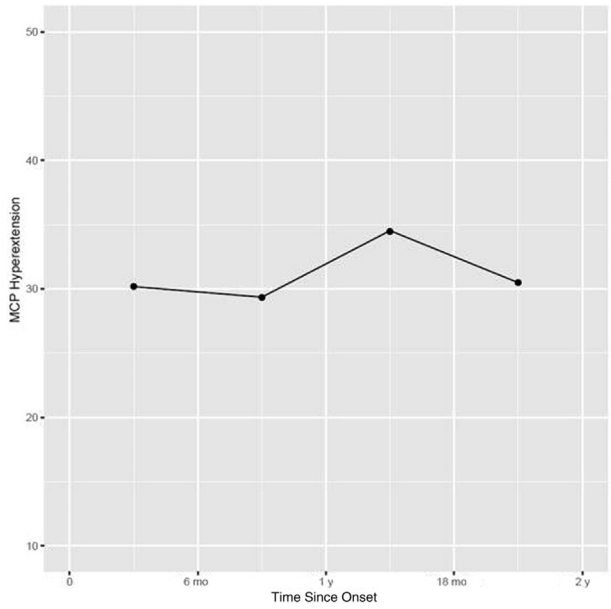

FIGURE 2: All thumbs, MCP joint hyperextension versus time.

Although our study represents the largest prospectively followed cohort of pediatric trigger finger patients, our follow-up time is similar to other large pediatric trigger thumb studies.[13,14,17] The median time to resolution has been shown to average about 5 years in these studies, and therefore, an ideal duration of follow-up would be closer to 10 years. Furthermore, our study population is reflective of the practice patterns at our institution in the United States, limiting the external validity of these results to patients in other regions of the world. We believe that insurance issues and patient choice in the United States leads to a higher surgical rate in general.

This study shows that the outcomes for nonsurgical management of pediatric trigger thumb are variable. However, there is a relationship between higher baseline IP joint flexion and failure to spontaneously resolve. Baek and Lee[14] and Jung et al[17] found that the natural history of pediatric trigger thumb favors spontaneous resolution; however, our study does not confirm their results. We believe the difference in our findings may be the result of multiple factors including the initial severity of IP joint flexion in our cohort and cultural differences. We do agree with Baek and Lee[14] that nonsurgical care of pediatric trigger thumbs is appropriate in many cases. We know of no deleterious effects to delaying surgery and there is benefit from waiting to identify late-presenting bilateral thumb involvement. It is also consistent with the suggestion of our anesthesia colleagues to delay elective pediatric surgery beyond 3 years of age.[18–21] Furthermore, a year of observation even in the more severe flexion contractures allows the physician and family to observe for improvement. If there was an improvement, we would recommend continued observation. However, surgery is safe and effective.

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