

ORIGINAL ARTICLE

Hand

A Retrospective Review of Patient-reported Outcomes after Postaxial Polydactyly Ligation and Surgical Excision

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Background: Interventions for type B postaxial polydactyly include suture ligation and surgical excision, yet there is a paucity of literature comparing the outcomes of these procedures. This study sought to compare patient-reported long-term outcomes of postaxial digit excision.

Methods: A six-question survey was distributed from January 2021 to March 2022 to patients who underwent treatment for type B postaxial polydactyly at a single pediatric institution from 2010 to 2016. Patients were queried about the incidence of pain sensitivity, keloid healing, and/or persistent presence of bump ("nubbin") at the treatment site.

Results: A total of 158 responses accounting for 258 digits were attained for a 53% response rate. The majority of digits (67.4%, n = 174) were surgically excised. Median age at procedure was 49 days: 13.0 days for ligation, 63.0 days for surgical excision. Median age at survey was 8 [IQR 5.4–10.2] years. Short-term (<30 days after procedure) complications rate was 1.6%. The rate of a raised or sensitive scar was 39.5% (ligation 51.5% versus surgery 35.4%, P < 0.05). The likelihood of postoperative sensitivity (P = 0.80) was similar among groups. However, the odds of a residual bump or raised scar at the surgical site was significantly higher in the ligation group (P = 0.001). These findings remained significant in the adjusted analysis. **Conclusion:** This study suggests that suture ligation can be used in select cases without increasing the prevalence of long-term pain or sensitivity, albeit with greater risk of a bump or raised scar at the excision site compared with surgical excision. (*Plast Reconstr Surg Glob Open 2024; 12:e5557; doi: 10.1097/GOX.000000000005557; Published online 23 January 2024.*)

INTRODUCTION

Postaxial polydactyly is one of the most common congenital upper extremity abnormalities, affecting almost four in every 10,000 newborns annually.^{1,2} It is characterized by a supernumerary digit on the ulnar aspect of the small finger, which can present as an articulated, fully formed digit with osseous components (type A), or as a digit or "nubbin," attached by a narrow skin bridge without the presence of any bone parts (type B).^{3,4}

Interventions to remove the nonfunctional digit in patients with type B postaxial polydactyly include

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Copyright © 2024 The Authors. Published by Wolters Kluwer Health, Inc. on behalf of The American Society of Plastic Surgeons. This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal. DOI: 10.1097/GOX.00000000005557 suture/clip ligation and surgical excision. As each technique has different advantages and disadvantages, specific patient characteristics dictate which type of procedure is indicated. Suture ligation is a simple procedure that can be performed in an office by either surgeons or pediatricians, requires no anesthesia, and is easiest to perform in young infants with a narrow digit stalk. However, it has been associated with higher rates of raised scar and/or a neuroma when the digital nerve does not retract appropriately into the subcutaneous tissue.^{5–7}

Surgical excision can be performed using local anesthesia in very young infants (usually <4 months) or can be delayed until a later age under general anesthesia.⁸ This procedure can be performed for digits with larger soft tissue stalks and has been touted to have a reduced risk of neuroma⁵ and better aesthetic outcomes.^{9,10} The current

Disclosure statements are at the end of this article, following the correspondence information.

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literature comparing postoperative outcomes of suture ligation and/or excision is limited to case reports and small case series.^{18,11} Therefore, the purpose of this study was to compare the long-term complications and patient-reported outcomes between suture ligation and surgical excision of type B postaxial polydactyly.

PATIENTS AND METHODS

Study Design

Following approval from our institutional review board, the authors performed a retrospective review of patients who underwent primary suture ligation or surgical excision for type B postaxial polydactyly at our tertiary care institution between 2010 and 2016. The study cohort was identified by performing a search from the billing department for the division of plastic and reconstructive surgery using International Classification of Diseases, version 9 and 10 (ICD 9 and 10) codes for polydactyly diagnosis (ICD 10: Q69.9 and ICD 9: 755). Patients with inadequate follow-up, contradictory responses, or those not interested in participating were excluded from the study.

Study Variables

The information recorded included baseline demographic characteristics, age at the time of surgery, type of treatment (ligation versus surgical excision), complications after the initial procedure, long-term outcomes, and additional required procedures. Postoperative outcomes were separated into short and long-term. Short-term complications were recorded for the immediate 30-day postprocedure period. Long-term outcomes were queried on the survey and included the presence of a raised scar or bump, and pain or sensitivity on the side of the polydactyly excision.

Survey Design and Distribution

A six-question survey asking about long-term outcomes associated with either polydactyly suture ligation or surgical excision was designed on REDcap—Research Electronic Data Capture, hosted at Children's National Hospital of Washington, DC.^{12,13} REDCap is a secure, webbased software platform designed to support data capture for research studies, providing (1) an intuitive interface for validated data capture; (2) audit trails for tracking data manipulation and export procedures; (3) automated export procedures for seamless data downloads to common statistical packages; and (4) procedures for data integration and interoperability with external sources. The survey instrument was not linked to identifiable information; all responses were anonymous.

This survey was distributed to all eligible patients and parents between January 2021 and March 2022. The survey was distributed to selected participants via email; however, to ensure access to the questionnaire and to clarify any questions of the participants, we conducted follow-up phone calls. All responses were recorded through a telephone discussion with a research member. (See appendix, Supplemental Digital Content 1, which displays the survey

Takeaways

Question: What are the long-term outcomes of suture ligation versus surgical excision for type B postaxial polydactyly?

Findings: A survey at our pediatric institution (2010–2016) found that suture ligation resulted in more raised or sensitive scars (51.5%) than surgical excision (35.4%). However, no significant difference in postoperative sensitivity or short-term outcomes was noted.

Meaning: Suture ligation for polydactyly may lead to more noticeable scarring than surgical excision.

distributed to evaluate the long-term outcomes of ligation versus surgical excision for type-B postaxial polydactyly. http://links.lww.com/PRSGO/D37.) Questions were formulated to identify any potentially undesirable long-term outcomes, including the presence of a residual bump or raised scar, and pain/sensitivity at the site of the removed accessory digit. The hypertrophic scar was defined as a thickened, raised scar that is confined to the region where the procedure or ligation occurred. However, since residual bumps and raised scars could be potentially confused, we decided to analyze these variables together. Sensitivity was described as a burning, shooting, or electric shock pain that can occur at the site of the procedure due to exposure to stimuli. If any of these outcomes were present, the degree of parental concern or patient symptoms was queried and graded using five descriptors as follows: (1) always, (2) often, (3) sometimes, (4) rarely, and (5) never. Surveys with incomplete or contradictory responses were excluded.

Statistical Analysis

Statistical analysis was performed using Stata software, version 14.2 MP (Stata Corporation, College Station, Tex.). Fisher exact or chi-square tests were used to analyze categorical data. Mann-Whitney or t tests were conducted to study continuous data. The frequency of early complications, longer term outcomes, and descriptive statistics of the sample's clinical and demographic characteristics were recorded. Values were expressed as mean \pm SD (SD) or median with interquartile range (IQR) for the continuous variables and frequencies with percentages for the categorical variables. A P value less than 0.05 was considered statistically significant. Gender, race, and laterality of reconstruction were incorporated in a multivariate logistic regression model to evaluate any association between the different treatment modalities and the incidence of complications (nubbin, pain or sensitivity, and hypertrophic scar).

Technique

Suture ligation was usually performed at the outpatient clinic or at the newborn nursery. For patients managed by ligation in our center, this option was not offered to parents if the base of the accessory digit was excessively wide. One of our surgeons (M.J.B.) refused to offer ligation to any patient. For those patients in whom ligation was considered feasible, parents were offered the choice

	Total (N = 158)	Ligation (N = 57)	Excision (N = 101)			
Patient Characteristics	Median [IQR] or n (%)					
Age at procedure (d)	49.0 [21.0-97]	13.0 [0.0–38.0]	63.0 [33.3-113.0]	<0.001		
Female, n (%)	73 (46.2)	26 (48.1)	47 (45.2)	0.724		
Race, n (%)						
African American	106 (67.1)	36 (66.7)	70 (67.3)	0.053		
Non-African American	17 (10.8)	2 (3.7)	15 (14.4)			
White	14 (8.9)	2 (3.7)	12 (11.88)			
Hispanic	2 (1.3)	0	2 (1.98)			
Asian	1 (0.6)	0	1 (0.99)			
Unknown	35 (22.2)	16 (29.6)	19 (18.3)			
Redo*	21 (13.29)	18 (31.58)	3 (2.97)	0.001		
Short-term complications†	4 (1.6)	1 (1.2)	3 (1.7)	0.745		

P values less than 0.05 indicate statistical significance.

*Pvalues were obtained from Mann-Whitney U test for continuous data and chi-square/Fisher exact test for binary and categorical data.

†Number of digits are used as the unit of analysis.

of ligation or surgical excision, and all were informed, based on prior literature, that ligation may be more likely to result in a residual bump or raised scar or increase the chance for increased scar sensitivity. Thus, in all instances of ligation, parents elected this option despite these potential disadvantages. Ligation was performed using a 3-0 Vicryl suture secured as close to the base of the accessory digit as possible. Suture ligature was considered successful if the pallor of the polydactylous digit occurred.

Surgical excision was performed under local or general anesthesia, depending on the age of the patient. A digital block was performed in all cases on the ulnar aspect of the digit with either 0.5 or 1% lidocaine with 1:100,000 epinephrine, proximal to the base of the supernumerary digit. The hand was prepared with povidone-iodine or chlorhexidine and draped with sterile towels. Iris scissors or scalpel were used to remove/incise the stalk. After excision, the wound was closed with absorbable sutures, keeping the neurovascular bundle out of the closure. Dermabond or antibiotic ointment was placed on the wound and wrapped with a sterile dressing. All patients were discharged on the same day of the procedure.

RESULTS

A total of 158 responses met the inclusion criteria for a response rate of 53% (158 of 298 patients). Overall, there were 258 digits excised; 67.4% were initially managed surgically (n = 174) and 32.6% underwent suture ligation (n = 84). Median age at the time of procedure across both cohorts was 49 (IQR 21–97) days. The majority of patients were African American (67.1%) and male (54.8%). Patients treated with surgical excision were significantly older at the time of the procedure [median age 63.0 (33.3–113.0) versus 13.0 (0.0–38.0) days, P < 0.001] and were more likely to be an ethnicity other than African American (14.4% versus 3.7%, P < 0.053) patients. The baseline demographic characteristics of the study population are summarized in Table 1.

The total rate of short-term complication rate was 1.6%, composed of four cases (ligation 1.2% versus surgical

excision 1.7%, P=0.745; Table 1). Three cases had a surgicalsite infection, treated with an oral antibiotic, and one case had minor bleeding that spontaneously resolved. Twentyone cases (ligation n = 18 versus surgical n = 3, P < 0.05) underwent re-operation due to the persistence of a remnant that parents found unacceptable (n = 15), remnant with scar sensitivity (n = 5), or scar sensitivity (n = 1; Table 1). The majority of these patients had a ligation performed elsewhere, usually in the newborn nursery (57.1%), and the remaining had the digits ligated at our center (42.9%). To evaluate the long-term outcomes, ligations that were later converted to excisions were counted as surgical excision, which ultimately resulted in 66 ligated digits (25.6%) and 192 (74.4%) digits treated with surgical excision.

Median age at the time of survey was 8 (IQR 5.4-10.2) years. Patients who underwent surgical excision were significantly older at the time of the survey compared with the suture ligation group [median age 8.2 years (6.2-10.2) versus 5.3 years (4.7-10.2), P < 0.05]. Overall, the rate of having either scar sensitivity, or a raised scar or residual bump, was 39.5% (ligation 51.5%) versus surgical excision 35.4%, P < 0.05). Because this study relied on parent-reported outcomes and the distinction between a residual bump or nubbin and raised or hypertrophic scar was considered too arbitrary, these outcomes were treated as a single condition for the purpose of the analysis; the rate of persistent bump or raised scar at the treatment site was 38.4% (Table 2). For patients who underwent surgical excision, we analyzed the long-term outcomes comparing those who had a primary procedure against those with a secondary (revision) excision. This analysis showed that although the rates of raised scar formation or persistent bump were comparable, there were significantly higher rates of pain and sensitivity in patients who underwent a secondary procedure (Table 3). Univariate logistic regression analysis demonstrated that the odds of persistent bump or raised scar in the excision group was 52% lower than in the ligation group [odds ratio (OR): 0.48; 95% confidence interval (CI), 0.27–0.86; *P* = 0.013; Table 4]. This trend remained significant after adjusting age at

	Total (N = 258)	Ligation (N = 66)	Excision (N = 192)		
Long-term Outcomes	Median [IQR] or n (%)				
Age at survey (y)	8.0 [5.4–10.2]	5.3 [4.7–10.2]	8.2 [6.2–10.2]	0.028	
Long-term outcomes*	102 (39.5)	34 (51.5)	68 (35.4)	0.021	
Bump or noticeable scar at site	99 (38.4)	33 (50.0)	66 (34.4)	0.024	
Pain/sensitivity	30 (11.6)	8 (12.1)	22 (11.5)	0.965	

Table 2. Comparison of Prevalence of Long-term Outcomes between the Two Groups

*We analyzed the long-term outcomes as separate independent variables, as some patients had both complications.

Pvalues less than 0.05 indicate statistical significance.

Table 3. Comparison of Prevalence of Potentially Adverse Outcomes between Primary versus Secondary Surgical Excision

majority of patients (>50%) reported it being rarely bothersome (Table 6).

DISCUSSION

	Primary (N = 171)	Secondary (N = 21)		
Outcomes	n (%)		P	
Nubbin/scar	60 (35.1)	6 (28.6)	0.553	
Pain/sensitivity	16 (9.4)	6 (28.6)	0.009	

Pvalues less than 0.05 indicate statistical significance.

procedure, gender, race, and whether the procedures were performed at birth (nursery) or later by a surgeon (Table 4). Rates of postoperative pain or sensitivity and hypertrophic scarring were similar between the treatment groups (Tables 2–4). Lastly, among the patients who underwent surgical excision, the prevalence of a residual bump/raised scar or pain was similar among the patients who underwent excision with scissors and scalpel (Table 5).

Patients who described the presence of a bump/ noticeable scar and/or pain/sensitivity at the site of the previously removed digit were asked additional questions to identify the degree of disturbance of each finding. Among the group of patients with a bump/scar, over half the patients reported never being bothered by it (51.3%). This percentage was consistent between both the excision group and the ligation group (53.6% versus 50.0%). Among the patients with persistent pain or sensitivity, the

Management of postaxial type B polydactyly invariably involves removal of the nonfunctional extra digit.^{1,3,8} Although the method and age of definitive treatment vary, the goals are similar-complete removal of the redundant tissue, minimize residual scarring and limit potential morbidity associated with the procedure. Considerations such as the digit's size, presence/absence of the bone, and diameter of the digit's base will influence the decision to undergo either of the treatment modalities discussed in this study and should be carefully taken into consideration before deciding the treatment approach.^{7,11,14} For instance, previous authors have indicated that incomplete amputation with ligation could be more likely when the base of the polydactylous digit is greater than 2mm in diameter.^{11,15} Moreover, ligation is an inappropriate treatment for any polydactylous digit that has a bony connection to the adjacent digit.^{10,14} In our study, only patients who would present with a relatively narrow soft tissue stalk were offered the option of ligation. For such accessory digits, both suture ligation and surgical excision are viable treatment options, and the evidence to support one method over the other is unconvincing due to the methodologic limitations of most studies.^{15,16} One recent systematic review and meta-analysis compiling the patient-reported

	Table 4. Unad	justed and Ad	justed Regre	ession Analysi	is Comparing	g Outcomes between	Two Procedures
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	Nubbin/Scar		Pain or Sensitivity						
Variables		Univariate		Multivariable		Univariate		Multivariable	
		OR (95% CI)	Р	Adjusted Odds Ratio (95% CI)	Р	OR (95% CI)	Р	Adjusted Odds Ratio (95% CI)	Р
Treatment	Ligation	Ref	Ref	Ref	Ref	Ref	Ref	Ref	Ref
	Excision	0.48 (0.27-0.86)	0.013	0.48 (0.25-0.92)	0.027	0.90 (0.39-2.24)	0.802	0.69 (0.28-1.86)	0.44
Age at procedures	Per 1 day	1.00 (1.00-1.00)	0.403	1.00 (1.00-1.00)	0.759	1.00 (1.00-1.00)	0.107	1.00 (1.00-1.00)	0.091
Sex	Male	Ref	Ref	Ref	Ref	Ref	Ref	Ref	Ref
	Female	1.18 (0.72-1.96)	0.51	1.14 (0.68-1.92)	0.62	0.85 (0.39-1.82)	0.677	0.81 (0.36-1.76)	0.593
Race	African American	Ref	Ref	Ref	Ref	Ref	Ref	Ref	Ref
	Non-African American	0.77 (0.30-1.88)	0.58	0.93 (0.35-2.32)	0.886	0.63 (0.10-2.33)	0.544	0.59 (0.09-2.28)	0.501
	Unknown	0.71 (0.38-1.30)	0.27	0.63 (0.33-1.17)	0.15	0.59 (0.19-1.50)	0.302	0.56 (0.18-1.45)	0.264
Surgeon or at birth	Nursery	Ref	Ref	Ref	Ref	Ref	Ref	Ref	Ref
	Surgeon	0.54 (0.18-1.55)	0.246	0.80 (0.25-2.55)	0.71	1.93 (0.37-35.68)	0.532	2.06 (0.34-39.97)	0.511
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Pvalues less than 0.05 indicate statistical significance.

	Scissors (N = 60)	Scalpel (N = 132)	
Outcomes	n ((%)	P
Nubbin/scar	24 (40.0)	42 (31.8)	0.269
Pain/sensitivity	7 (11.7)	15 (11.4)	0.951

Table 5. Comparison of Prevalence of Potentially Adverse Outcomes between Scalpel versus Scissors

Table 6. Percentage of How Much the Complications Disturb the Patient

	Overall	Ligation	Excision
Outcomes			
Pain or sensitivity (N = 30)			
Always	0 (0.0)	0 (0.0)	0 (0.0)
Often	4 (13.3)	0 (0.0)	4 (18.2)
Sometimes	10 (33.3)	3 (37.5)	7 (31.8)
Rarely	15 (50.0)	4 (50.0)	11 (50.0)
Never	1 (3.3)	1 (12.5)	0 (0.0)
Nubbin/scar (N = 78)			
Always	1 (1.0)	1 (3.0)	0 (0.0)
Often	8 (8.1)	2 (6.1)	6 (9.1)
Sometimes	20 (20.2)	6 (18.2)	14 (21.2)
Rarely	20 (20.2)	7 (21.2)	13 (19.7)
Never	50 (50.5)	17 (51.5)	33 (50.0)

outcomes after ligation or excision found that potentially undesirable outcomes were six times more likely to occur with ligation procedures compared with surgical removal. These complications included treatment failure, neuroma, nubbin or unaesthetic scarring, and procedure revision. Nevertheless, the authors acknowledged a high risk of bias and low methodological quality in all included studies.¹⁷ Our single-center comparison study of 258 polydactylous digits managed with ligation or surgical excision overcomes the power limitations of previous smaller investigations.

Some authors view ligation as inferior to surgical excision for the treatment of type B postaxial polydactyly, and some reports have suggested there is not enough evidence to support the use of suture ligation for the treatment of supernumerary digits.^{14,15} The proposed advantages of surgical excision over ligation include better long-term cosmesis, lower rates of neuroma formation, and decreased need for revision surgery.⁸ Nevertheless, ligation has many clear advantages. First, it is easy to perform and can be done immediately and by any provider, thus eliminating the need for a time-consuming referral to a specialist. While ligation can be performed in the newborn nursery, allowing for immediate treatment of the patient and reduced parental burden, surgical excision is generally performed by an experienced surgeon due to the delicate nature of the structures involved.^{5,6,8} Second, ligation is far less expensive than surgical excision since it requires no special skill, equipment, or facility. Mills et al compared the cost of surgical clip ligation (\$230) versus surgical excision (\$7000-\$10,000), demonstrating a positive cost balance for procedures that do not require general anesthesia. Lastly, ligation rarely requires an anesthetic, even in

older infants, thus eliminating the minor risks associated with the use of local or general anesthesia.^{5,18,19} Patients in our study underwent ligation at a median of 13 days of age in either the nursery or outpatient clinic.

Our study found that the risk of short-term complications was extremely low (1.6%) and comparable among the two groups. However, the revision rates of patients who underwent ligation was significantly higher. Long-term outcomes in our study focused on inquiring about two main aspects: the presence of residual bump or a raised scar and the presence of abnormal sensitivity or pain. Scar appearance can be subjective, and the distinction between a residual bump or nubbin and hypertrophic scarring can be challenging. Heterogeneity and variability in the literature regarding this endpoint-scar-is not surprising, as previous authors have highlighted the subjective nature of this parameter, especially when evaluated by patients, parents, or healthcare providers.²⁰ Our analysis grouped together factors related to the appearance of the scar to avoid these issues. Parents in our study reported a 38.4% rate of postoperative bump or raised scar, and this was significantly higher for patients who underwent ligation at 50%. This is comparable to the findings of Watson and Hennrikus, who observed noticeable or raised scars in 43% of these patients following suture ligation.¹¹ Surprisingly, this finding was also observed in 34.4% of our patients who had undergone surgical excision, a rate much higher than reported in some other studies.⁹ Although this was evenly distributed among patients who underwent primary and secondary surgical excision, the use of a straight iris scissor to perform the excision led to slightly higher rates of residual bump or nub than using a scalpel. We have no explanation why these two techniques, both of which cut the polydactylous digit flush to the remaining skin, would have such a disparate outcome or why over a third of patients were reported to have residual bumps or raised scars after excision. We hypothesize that our use of parent-reported impressions of scar appearance, as opposed to that of a clinician, may lend toward a more inclusive reporting of this outcome since any visible or palpable scar may be perceived as a residual bump. Regardless, surgical excision still yielded significantly lower rates of bump formation when compared with suture ligation. This finding is similar to that of previous cohort studies.^{16,21} Although the presence of a raised bump has been touted as an undesirable outcome or complication of ligation, all patients treated at our center with ligation chose this treatment despite being informed of this possibility, and comparatively few reported concerns or elected to undergo revision surgery.

One of the most important comparisons in the present study relates to scar sensitivity or neuroma formation. Previously published studies have demonstrated a tendency towards higher rates of pain and sensitivity symptoms in patients who underwent suture ligation of the extra digits.^{5,8} In fact, some authors have attributed this discomfort to the presence of neuroma in painful vestigial amputation stumps that resulted in the ligation procedure.⁶ In a prospective nonrandomized study, Samra et al reported that despite 1-month follow-up satisfaction scores averaging 9.9 of 10 after suture ligation, 50% of parents thought patients still experienced some degree of pain related to the procedure.⁸ Mills et al. also reported that raised scars were painful to a certain degree in 7% of cases after surgical clip ligation.⁵ Rayan and Frey reported a 16.19% rate of tenderness after ligation.⁴ Even though our series shows a higher rate of pain and sensitivity for the suture ligation group compared with patients who underwent surgical excision, the difference was not statistically significant (18% versus 15%, P = 0.647). Nonetheless, it is important to mention those patients who underwent secondary excision for various reasons, as they experience significantly higher rates of pain/sensitivity compared with those patients who underwent primary digit excision.

This study is not without limitations that require acknowledgment. Patient-reported outcomes are subjective in nature and, as with the majority of survey studies, questions could be interpreted differently by each participant; hence, variability in the responses may have occurred. Furthermore, recall bias must be mentioned as a possible limitation due to the nature of this study. Also, as parents are used as a proxy for patient-reported outcomes in some cases, under-or-overreport of information can occur. The questionnaire used in the survey, as well as the five descriptors used to evaluate frequency of bothersome related to long-term complications are not validated. Due to the retrospective nature of this study and the unblinded design of this report, selection and performance bias can also affect the results.

CONCLUSIONS

Treatment of type B postaxial polydactyl by ligation or excision provides comparable outcomes in terms of early postoperative complications and long-term pain or scar sensitivity. Suture ligation is more likely to result in the presence of a raised scar or bump at the treatment site, yet most parents were not concerned by this finding. Given the time and cost differences between these comparable alternatives, the decision about which treatment is preferred for any individual patient should be vested in the family.

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DISCLOSURE

The authors have no financial interest to declare in relation to the content of this article.

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