

Decision-making and Management of Ulnar Polydactyly of the Newborn: Outcomes and Satisfaction

Salem Samra^{*}, Debra Bourne[†], Joel Beckett[‡], Michael Matthew[§], J. Grant Thomson[‡]

^{*}Samra Plastic Surgery, Holmdel, NJ, [†]University of Pittsburgh Medical Center, Department of Plastic Surgery, Pittsburgh, USA, [‡]Yale University School of Medicine, Section of Plastic Surgery, New Haven, USA, [§]Dartmouth-Hitchcock Medical Center, Section of Plastic Surgery, Lebanon, USA

Background: Polydactyly is the most common congenital hand deformity. There is currently no consensus among pediatricians or hand surgeons regarding optimal management of ulnar (post-axial) type B polydactyly leading to uncertainty in initial treatment decisions for infants with this type of deformity.

Methods: Parents of newborns with type B ulnar polydactyly were enrolled in a prospective, nonrandomized trial from 2010 to 2012. At enrollment they were offered: 1) no treatment, 2) suture ligation, 3) immediate excision under local anesthesia, and 4) delayed excision at four months of age under general anesthesia. After choosing a treatment option, parents were asked to complete an initial survey on their motivation for choosing a particular option, and then additional surveys at one and three month follow-up visits regarding outcomes and satisfaction.

Results: Fourteen newborns were enrolled in the study. Eleven patients had bilateral polydactyly for a total of 25 hands involved in the study. With statistical significance, parents chose an immediate bedside excision over all options. No surgical complications were noted and satisfaction scores were 9.8 or higher in all groups.

Conclusions: A majority of parents whose children are born with type B post-axial polydactyly prefer to have the deformity addressed immediately with excision under local anesthesia at the bedside. This can be accomplished safely, with a satisfaction score of 9.8 out of 10 and no appreciable residual deformity. This may reduce emotional distress or embarrassment in parents who would otherwise have to deal with the deformity for at least a four-month period.

Keywords: Ulnar, Congenital Hand, Polydactyly

INTRODUCTION

Polydactyly is the most common congenital hand deformity. The incidence and location of polydactyly varies depending on the population studied with the highest incidence in people of African descent at 10.7 of 1000 live births, compared to 2.5 of 1000 live births in Asian

populations and 1.6 of 1000 live births in Caucasians. While any digit can be duplicated, ulnar (post-axial) polydactyly is 10 times more common in people of African descent and twice as common in males compared to females. Differently, radial (pre-axial) polydactyly is more common in Asian populations. Central polydactyly is rare in all populations.¹⁾ This investigation focuses on ulnar or post-axial duplication.

Polydactyly has a wide spectrum of presentation, from a rudimentary nub of skin on the ulnar side of the hand, to a fully formed digit with bones and joints. Temtamy and McKusick categorized ulnar polydactyly into type A and B. Type A duplications describe a well-

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Correspondence to: Salem Samra

Samra Plastic Surgery, 300 Perrine Rd, Suite 333, Old Bridge, NJ 08857

Tel: +(732) 739-2100, Fax: +(732) 739-0815

E-mail: salemsamra@gmail.com

developed supernumerary digit that connects with the bony structure of the hand. In contrast, type B describes a rudimentary appendage loosely attached by a soft tissue bridge.^{2,3)} This investigation specifically focus on type B post-axial polydactyly or a supernumerary digit attached to the ulnar side of the hand by a narrow skin bridge (Fig. 1). While diagnosing this deformity is fairly straightforward, providing a treatment plan has proven less so. In fact there is no clear consensus among pediatricians or hand surgeons regarding the management of an ulnar supernumerary digit.⁴⁾

For centuries, many pediatricians have elected to “tie off” the polydactylous small finger with a suture ligation. More recently hand surgeons have advocated an excision of the digit in the operating room when the child is of suitable age for general anesthesia.⁵⁾ Needless to say, this discrepancy has led to uncertainty in initial treatment decisions by parents and caregivers of patients with this deformity. A survey study of pediatricians showed that 79% would refer patients with the deformity to a hand surgeon, 15% would perform suture ligation, and 5% would excise the digit themselves.⁶⁾

There are currently four options for newborns born with a type B ulnar polydactyly. Option 1 would be to do nothing. It is a benign deformity and having a supernumerary digit would likely not interfere with hand function. However, the hand is the second most visible part of the body after the face⁷⁾ and most parents would reject the idea of not intervening.

Option 2 is to perform a suture ligation of the digit. Using a silk tie, the blood supply to the digit is strangulated. This leads to eventual necrosis and auto-am-

putation. The benefits of this option include avoidance of surgery or anesthesia, immediate implementation in the newborn nursery (although the digit itself may take weeks to slough off), and ease to perform. The disadvantages include the pain that it causes the child (often there is a digital nerve in the skin bridge being tied off), the fact that most patients (up to 40%) end up with a residual stump after the digit auto-amputates,⁵⁾ and the significant risk of neuroma formation in the amputated stump.⁵⁾

Option 3 is to perform an immediate excision of the polydactylous digit in the newborn nursery or office under local anesthesia. The benefits of this approach include immediate treatment of the deformity, and a limitation of the painful effect to the transient discomfort of the injection of local anesthetic. The disadvantages include the risks of a surgical procedure: namely bleeding, infection, or injury to the non-polydactylous small finger, as well as incomplete dissection and high ligation of the accessory digital nerve. With limited lighting, support staff, instrumentation and a non-cooperative patient, a bedside excision can prove to be a stressful experience for both surgeon and parent.

Option 4 is an excision of the supernumerary digit in the operating room under general anesthesia. The advantage of this approach is that it optimizes the surgical outcome given the resources available in the operating room coupled with having a still, anesthetized patient to operate on. It is easier to dissect out the accessory digital nerve and perform a high ligation as advocated by Gosain⁵⁾ and the skin tailoring can be more easily performed



Fig. 1. Type B post-axial polydactyly in an infant prior to treatment.



Fig. 2. Post-operative appearance of infant with type B post-axial polydactyly following amputation of supernumerary digit in the operating room under general anesthesia at 4 months.

to avoid dog-ears or a residual stump (Fig. 2). The disadvantage of this approach is obvious. The risks of general anesthesia and a surgical procedure are real in addition to the fact that parents must wait at least 4 months with the deformity until the child is old enough to undergo general anesthesia safely.

METHODS

This is a prospective, nonrandomized study. Approval for this study was obtained from the Institutional Review Board at the institution where this research was performed. Parents of children born with type B post-axial ulnar polydactyly in the newborn nursery were enrolled over a two-year period, from 2010 to 2012.

The four treatment options were presented to parents by the research team in a non-biased manner. That is, patients were informed of the pros and cons of each option, but none of the options was explicitly recommended or discouraged. Once parents selected a treatment option, they were asked to fill out three surveys. The first survey was completed at the time of the initial consultation, the second at one month post-procedure, and the third at three months following the procedure (Appendix).

Survey questions at the initial consult pertained to parents' motivation for choosing their particular treatment option, family history of polydactyly, the level of surgeon influence they perceived during their initial consultation and when making their selection, and their overall comfort with their decision.

The follow-up survey was completed one month following the procedure to assess satisfaction with the baby's outcome, whether or not they thought their child perceived pain with whatever treatment option was selected, if they perceived a residual deformity, and how much emotional distress they experienced with their child's course.

Finally, three months following the procedure, parents were again asked to answer the questions presented in the 1 month survey as well as rate their satisfaction with the resultant scar.

For testing whether any treatment was preferred, we assumed by default that each of the four treatments would be chosen equally and then performed a one-sample proportion test to see whether any of the treatments deviated from that assumption. These tests were adjusted for multiple comparisons with a Bonferroni correction. A Fisher's exact test was used to determine if family history of polydactyly influenced treatment choice or perceived surgeon influence and a t-test was used to assess

the impact of family history on comfort with treatment decision. Significance was set at a p -value ≤ 0.05 .

RESULTS

The parents of 14 newborns with type B post-axial ulnar polydactyly were enrolled in the study. Eleven newborns had bilateral involvement, and therefore 25 hands were treated. Twelve of the 14 newborns had parents who identified themselves as black or African-American. Parent ages ranged from 15–34 years old (average 25 years of age).

Ten newborns had a positive family history of the deformity. Parents with a history of polydactyly personally or in close family members responded similarly to parents who did not report a positive family history. There was no significant difference in parents of children with a family history of polydactyly compared to parents without a family history in treatment choice ($p = 0.706$); nor was there a difference in the level of comfort with their treatment decision ($p = 0.613$), or the perception of surgeon influence on their choice ($p = 1.000$).

In 10 of the 14 cases, the parents chose to have the supernumerary digit excised immediately at the bedside in the newborn nursery. Two cases requested a suture ligation, and two cases opted for delayed excision in the operating room (Table 1). Parents preferred immediate excision under local anesthesia with statistical significance ($p < 0.001$).

Among the cases where the parents selected suture ligation, fear of a complication from a surgical procedure, fear of general anesthesia and concern for pain were cited as motivational factors. No surgeon influence at initial consultation was noted and parents were comfortable with their decision. One of the two sets of parents followed up at three months and reported 10 out of 10 satisfaction score with their child's outcome and described no residual deformity or unpleasant scarring.

Among the ten cases where parents selected immediate bedside excision under local anesthesia, the main motivating factors cited for their selection were concern for general anesthesia and avoiding potential pain in

Table 1. Number of subjects who chose each treatment option

Treatment	Number of Subjects
No Treatment	0
Suture Ligation	2
Immediate Excision	10
Delayed Excision	2

Table 2. Results from surveys completed by parents of infants with type B post-axial polydactyly

	Suture ligature	Excision under local anesthetic	Excision under general anesthesia
Motivation	Fear of surgery, Avoid pain	Avoid general anesthesia, Avoid pain, Embarrassment	Avoid pain
Surgeon influence	None	None to Moderate	Moderate
Comfort with decision (1–10)	10	9.5	9.5
Satisfaction with outcome (1–10)	10	9.8	10
Perceived pain in child (1–10)	5	2.1	0
Emotional distress (1–10)	0	0	6
Residual deformity (1–10)	0	0	0
Satisfaction with scar (1–10)	10	9.7	10

Results shown for each treatment option including suture ligature, excision under local anesthetic, and excision under general anesthesia. Motivation to choose a treatment option was based on multiple-choice questions. Other data points were based on ten point Likert scales with 1 representing the lowest and 10 representing the highest possible score. The mean value for each group is presented. Motivation, surgeon influence, and comfort with the decision were obtained at the initial survey. Satisfaction with outcome, perceived pain, emotional distress, residual deformity and satisfaction with scar scores were obtained from the 3 month follow-up survey.

their child (6 of 10 cases), while three cases cited embarrassment taking their child home with the deformity. In this group surgeon influence was described as moderate by 6 out of 10 cases, and the average “comfort with your decision” score was 9.5 (out of 10).

At one month follow-up satisfaction score averaged 9.9 among parents but 50% thought their baby did experience some pain with the procedure. None described any emotional distress with their child’s course. At three-month follow-up, satisfaction scores remained high (9.8) and there were no cases of residual deformity. Scar quality was rated an average of 9.7 out of 10.

Between the two babies who had a delayed excision in the operating room after the baby reached an age of at least four months, both sets of parents described their motivation to be avoiding potential pain in their child. They both described moderate surgeon influence and felt mostly comfortable with their decision (9.5). At one month post-operative follow-up satisfaction score averaged 9.5 and both sets of parents reported their child had 0 out of 10 pain with the procedure. Both sets of parents did describe emotional distress (average score of 6 out of 10) with having to wait four months before the surgery could take place. At the three month post-operative follow-up visit, the satisfaction score was 10, there was no appreciable residual deformity and scar quality score was 10 (Table 2).

DISCUSSION

With various treatment modalities available and no clear consensus regarding treatment, parents of newborns with type B post-axial ulnar polydactyly face a

somewhat difficult decision. While most would seek an intervention for their child, parents are unable to accurately gauge how much pain a procedure could entail for their baby, and there is a deficiency of data regarding outcomes comparing treatment options. Furthermore, among health care providers there are varying accounts of problems with surgical procedures (hemorrhage and neuroma among others) but no evidence-based guidelines from which to make recommendations.

This study presents helpful data regarding parents’ preferences and motivational factors when presented with the spectrum of treatment modalities in an unbiased manner. Additionally, we offer short-term follow-up (one and three months) of treated newborns to assess outcomes from their parents’ perspective.

This information is useful for clinicians advising parents of newborns with type B ulnar polydactyly as to what their peers prefer when facing treatment decisions. Furthermore it sheds light on possible risks and benefits of treatment options and how this affects the parents’ decision-making process. We found that immediate excision at the bedside can be safely performed in the newborn period. No complications were noted in the ten children who underwent the procedure and no residual deformities were appreciable. These findings have ramifications for not only patient education but also for future cost analysis. A procedure that can be performed successfully and safely in an office setting under local anesthesia may reduce health care expenditure dollars, especially considering the cost of operating room time and pediatric general anesthesia.

We found that parents of children with polydactyly experience distress when intervention is delayed. Only

two of the 14 cases opted to wait for a procedure in the operating room, and both sets of parents described discomfort with introducing their new baby to friends and family due to the supernumerary digit. If a local procedure can be done safely and immediately, with low risk of neuroma and residual nub, this may be the ideal treatment option. We would recommend that an experienced hand surgeon perform the procedure given the delicate nature of the structures involved in the surgery and the potential for bleeding and neuroma.⁵⁾

Limitations of this study must be acknowledged. This is a relatively small sample size with follow-up of only three months. Results were based on surveys and therefore subjective. The validity and reliability of the questionnaires were not evaluated. No objective evaluation of neuroma formation and scarring was performed. This study investigates ulnar type B polydactyly. Future studies are needed to address the treatment for other types of polydactyly. Additionally, a cost analysis comparing treatment options would be valuable. A prospective, randomized trial with a large cohort that includes objective examination of the scar and perception of pain over the long-term (at least to an age when the child can communicate) would be helpful in further elucidating the optimal treatment of type B ulnar polydactyly.

In conclusion, immediate excision of the supernumerary digit in ulnar type B polydactyly in the nursery or office setting under local anesthesia can be safely performed in the newborn period. This treatment is preferred by the majority of parents in our study and results in high levels of satisfaction.

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CONFLICT OF INTEREST

The authors have no conflict of interest to disclose.

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APPENDIX

Polydactyly Study Survey – Initial Consult

Thank you for your participation in our study. For multiple choice questions please circle the response that you feel most accurately answers the question.

- What treatment option did you choose for your child?
 - No operation (do not want surgical correction of polydactyly)
 - Suture ligature (the extra digit was tied off)
 - Immediate excision under local anesthesia
 - Delayed excision at age 4 months under general anesthesia (in the operating room)
- What motivated you to choose the above treatment option? (Can circle more than one response)
 - Comfort with the deformity
 - Fear of complications of intervention
 - Concern regarding potential pain for child
 - Concern regarding potential risks of general anesthesia
 - Influenced to choose by medical professional
 - Embarrassment with others perceiving child's deformity
- Do you have any family or personal history of polydactyly (extra digits)?
 - Yes
 - No
- How much did your hand surgeon influence your treatment option decision?
 - No influence
 - Moderately influenced me
 - Decision based entirely on surgeon's opinion
- How old are you? _____
- What ethnic or racial group do you identify yourself as?

- Did you discuss your treatment option decision with your spouse or other family members?
 - Yes
 - No
- On a scale of 1 to 10, how comfortable are you with your decision?

1	2	3	4	5	6	7	8	9	10
Not comfortable			Somewhat comfortable				Very Comfortable		

Polydactyly Study Survey – One Month Follow-up Visit

Thank you for participating in our study. For multiple choice questions please circle the response that you feel most accurately answers the question.

- What treatment option did you choose for your child?
 - No operation (do not want surgical correction of polydactyly)
 - Suture ligature (the extra digit was tied off)
 - Immediate excision under local anesthesia
 - Delayed excision at age 4 months under general anesthesia (in the operating room)
- On a scale of 1 to 10, how satisfied are you with your child's treatment outcome?

1	2	3	4	5	6	7	8	9	10
Not Satisfied			Somewhat Satisfied				Very Satisfied		
- On a scale of 1 to 10, how satisfied are you with your decision?

1	2	3	4	5	6	7	8	9	10
Not Satisfied			Somewhat Satisfied				Very Satisfied		
- Would you make the same decision again?
 - Yes
 - No
- If the answer to question 4 is no, which treatment option would you choose now?
 - No operation (do not want surgical correction of polydactyly)
 - Suture ligature (the extra digit was tied off)
 - Immediate excision under local anesthesia
 - Delayed excision at age 4 months under general anesthesia (in the operating room)
- Do you think your child had pain with the treatment option you chose?
 - Yes
 - No
- If the answer to question 6 is yes, on a scale of 1-10 how much pain do you think your child experienced?

1	2	3	4	5	6	7	8	9	10
No Pain			Some Pain				Extreme Pain		

8. Does your child have a residual deformity after undergoing the treatment option you chose?
a. Yes b. No
9. If the answer to question 8 is yes, do you want the residual deformity addressed with another procedure in the future?
a. Yes b. No
10. Did you experience any embarrassment or emotional distress with your child's deformity until the point in time the extra digit was removed, or do you at all if you've chosen not to have the digit removed?
a. Yes b. No
11. If the answer to question 10 is yes, on a scale of 1-10 how much embarrassment or emotional distress did/ do you experience?

1 2 3 4 5 6 7 8 9 10
 No Embarrassment/Distress Some Embarrassment/Distress Extreme Embarrassment/Distress

Polydactyly Study Survey – Three Month Follow-up Visit

Thank you for participating in our study. For multiple choice questions please circle the response that you feel most accurately answers the question.

1. What treatment option did you choose for your child?
 a. No operation (do not want surgical correction of polydactyly)
 b. Suture ligature (the extra digit was tied off)
 c. Immediate excision under local anesthesia
 d. Delayed excision at age 4 months under general anesthesia (in the operating room)
2. On a scale of 1 to 10, how satisfied are you with your child's treatment outcome?
- 1 2 3 4 5 6 7 8 9 10
 Not Satisfied Somewhat Satisfied Very Satisfied
3. On a scale of 1 to 10, how satisfied are you with your decision?
- 1 2 3 4 5 6 7 8 9 10
 Not Satisfied Somewhat Satisfied Very Satisfied
4. Would you make the same decision again?
 a. Yes b. No
5. If the answer to question 4 is no, which treatment option would you choose now?
 a. No operation (do not want surgical correction of polydactyly)
 b. Suture ligature (the extra digit was tied off)
 c. Immediate excision under local anesthesia
 d. Delayed excision at age 4 months under general anesthesia (in the operating room)
6. Do you think your child had pain with the treatment option you chose?
 a. Yes b. No
7. If the answer to question 6 is yes, on a scale of 1-10 how much pain do you think your child experienced?

1 2 3 4 5 6 7 8 9 10
 No Pain Some Pain Extreme Pain

8. Does your child have a residual deformity after undergoing the treatment option you chose?
a. Yes b. No
9. If the answer to question 8 is yes, do you want the residual deformity addressed with another procedure in the future?
a. Yes b. No
10. Did you experience any embarrassment or emotional distress with your child's deformity until the point in time the extra digit was removed, or do you at all if you've chosen not to have the digit removed?
a. Yes b. No
11. If the answer to question 10 is yes, on a scale of 1-10 how much embarrassment or emotional distress did/do you experience?
- 1 2 3 4 5 6 7 8 9 10
No Embarrassment/Distress Some Embarrassment/Distress Extreme Embarrassment/Distress
12. If you chose to have the digit removed, on a scale of 1 to 10, how satisfied are you with the scar?
- 1 2 3 4 5 6 7 8 9 10
Not Satisfied Somewhat Satisfied Very Satisfied