

Indications for Surgery and Functional Status of Patients with Congenital Hand Anomalies Persisting beyond Childhood

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ABSTRACT

Background. Reconstruction for congenital hand anomalies, if indicated, is generally recommended during early childhood to optimize function before formal education starts and minimize the psychosocial impact. There have been limited reports on these conditions persisting beyond the recommended age for surgery.

Objective. The research aimed to explore the common reasons why patients consult for late surgery and determine the baseline function of the patients prior to reconstructive surgery using the Filipino Disability of Arm Shoulder and Hand (Fil-DASH).

Methods. An observational, chart review of cases from 2013-2021 was done. The reasons and indications were extracted from the history and Fil-DASH scores were recorded with available data. The results were presented using summary statistics and tabulation of qualitative data.

Results. Twenty-five hands in 20 patients underwent reconstruction of congenital anomalies persisting beyond childhood. The most common indication for surgery was concerns regarding work opportunities. Hope for a better function, cosmesis, social pressure and hygiene were the other reasons mentioned. Most of the patients did not complain of significant problems in doing activities of daily living. This was supported by their Fil-DASH scores and employment status prior to surgery.

Conclusion. In this population, the indications to request for reconstruction of congenital anomalies beyond childhood were not necessarily due to loss of function but to increase opportunities for employment, improved appearance and hope for increased function. The results may aid in the understanding of the natural history of congenital hand anomalies and guide the physicians with more specific advice for parents with children presenting with these conditions.

Keywords: congenital hand deformities, patient outcome assessment, FilDASH

INTRODUCTION

Congenital upper limb anomalies represent a spectrum of conditions presenting with different levels of anticipated disability. The different physical presentations, combined with factors related to cognitive and social functions, dictate the future abilities. They also direct the recommendations for the type and timing of surgical reconstructions.¹⁻³ The more common types of congenital hand deformities like syndactyly, constriction band syndrome and polydactyly have generally accepted indications to recommend surgery for functional and aesthetic indications.⁴ While there are conditions that have been shown to improve function after

Initial results presented as poster at the International Federation of Societies for Surgery of the Hand in Berlin, Germany, 17-21 June 2019.

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reconstruction, it has been suggested that some conditions may not benefit from surgery and are often prescribed with observation, prosthesis and guided adaptation.⁵

As to the timing, it is indicated to do earlier surgery for anomalies hindering growth and development of the parts of the limb. There are other sound arguments to perform reconstructions prior to school age or even as early as the first year of life, depending on the type of anomaly.⁶

Conditions that persist into adulthood are rarely discussed in literature and are often on the subject of treatment strategies and outcomes.^{7,8} The authors were not able to find previous publications presenting the objective baseline function and detailing the profile and specific indications for adults with non-operated congenital hand anomalies.

This research aimed to answer 2 basic questions. The primary objective was to tally and categorize the indications for undergoing a “late reconstruction.” The secondary objective was to objectively measure their level of function at the time when they consented for surgery.

METHODS

The study was an observational, review of charts of all patients that underwent reconstruction from 2013-2021. The study has been approved by the University of the Philippines Manila - Research Ethics Review Board last 10 October 2018 with registration number UPMREB-2018-123-01. Data collection was performed by the investigators.

All charts and medical records of patients, older than 12 years old, that underwent reconstructive procedures for congenital anomalies in the institution were collected for review.

The indication for surgery came from entries in the chief complaint and history which often narrates the reasons why early surgery was not considered. Other variables relevant to the objectives were also extracted from hospital records.

The recently translated and validated Disability of Arm Shoulder and Hand for Filipinos (Fil-DASH) allowed for routine quantitative measurement of function in the center for more recently encountered patients.⁹ Preoperative functional status evaluation was taken for those with available data and were evaluated as a subgroup.

RESULTS

Twenty-five hands in 20 patients underwent surgical reconstruction between 2013-2021 for a variety of previously untreated congenital anomalies. Their characteristics are summarized in Table 1.

The most common primary indication to consult for surgery was to gain work opportunities (8/20), as some positions require the use of protective equipment like gloves. Appearance (5/20) was the second most common reason, followed closely by hope for a better function (4/20). Hygiene (2/20) and social pressure (1/20) were the less commonly

mentioned reasons. The complete detailed list of indications is summarized in Table 2.

Since the translation and validation of the Fil-DASH was completed and became routine in pre-reconstruction

Table 1. Summary of Population Characteristics (n=20)

Parameter	Profile
Age	Average 25.21; SD – 6.96; Range 16-40
Sex	Male: 12; Female: 8
Employment	Employed: 14 Awaiting employment after college graduation: 1 Student: 5
Diagnosis	Constriction band: 7 Preaxial polydactyly: 6 Syndactyly: 4 Combined: 3
Involvement	Bilateral: 10 Right or left only: 10
Operated side	Bilateral: 5 Dominant: 10 Non-dominant: 5

Table 2. Specific Reasons for Consulting for Surgery with Corresponding Diagnosis

Category	Specific reasons	Categorical diagnosis
Work Opportunities	Wearing of gloves for work	Pre-axial polydactyly
	Requirement for overseas work	Pre-axial polydactyly
	Overseas employment opportunity	Syndactyly
	More opportunities for employment	Syndactyly
	Better work opportunities	Syndactyly
	Overseas employment requirement	Combined
	Overseas employment opportunity/application	Pre-axial polydactyly
Appearance	Military employment requirement	Combined
	Improve appearance	Constriction band
	Improve appearance	Pre-axial polydactyly
	Improve appearance	Constriction band
	Improve appearance	Constriction band
Better function	Improve appearance and get more opportunities	Syndactyly
	Improve function	Constriction band
	Efficiency at work	Pre-axial polydactyly
	Improve function	Combined
Hygiene	Improve function in school	Constriction band
	Hygiene, possible better efficiency in function	Constriction band
	Recurrent skin infection	Constriction band
Social Pressure	Social effects	Pre-axial polydactyly

Table 3. Summary of Fil-DASH Scores of Recent Patients with Diagnosis and Indications (n=10)

Age/Sex	Diagnosis	Indication	Fil-DASH Scores
24/F	Preaxial Polydactyly (Wassel IV)	Work opportunities	0.86
23/M	Constriction band with syndactyly	Hygiene and possible work efficiency	11.67
32/M	Constriction band with syndactyly	Recurrent skin infection	40.52
19/M	Syndactyly RF SF	More opportunities for employment	1.72
24/F	Pre-axial Polydactyly (Wassel II) both thumbs	Improve appearance	0.00
19/M	Syndactyly both hands	Improve appearance and get more opportunities	25.86
27/F	Syndactyly, both hands	Improve appearance	15.74
40/F	Pre-axial polydactyly (Wassel VII)	Overseas employment opportunity/application	1.70
24/F	Postaxial polydactyly and syndactyly, both hands	Overseas employment requirement	5.00
27/M	Symbrachydactyly	Military employment requirement	5.00

evaluation, ten patients were evaluated preoperatively for baseline function. Fil-DASH is interpreted similarly to the original DASH with a numerical value for the patient-reported disability. A value of 0 is no disability and 100 means total disability. All, except two, claim to have no significant disability. This is supported by their Fil-DASH scores averaging 10.807 (SD:12.58). Fourteen have been previously employed prior to surgery while 6 were students or recently graduated at the time of consultation. The Fil-DASH scores with corresponding diagnosis and indications are detailed in Table 3.

DISCUSSION

The variety of congenital hand cases presenting in adulthood closely parallels the types seen in children in the same institution. There seems to be no tendency for certain cases to be better tolerated, without intervention, to adulthood. Constriction band being the most common is a pattern seen similarly in other countries. Aside from this, the proportions are similar to epidemiologic studies in literature, including up to 50% presenting with bilateral affectation.¹⁰

The expectations following upper limb reconstructions are often assumed to be mostly related to improving function and appearance.⁴ While congenital anomalies vary in presen-

tation, most patients in this population did not necessarily consult because of the need to overcome physical disability. All have been able to function independently with previous employment or were able to attend regular school.

Securing more opportunities for employment seems to be the most logical reason to request for a reconstruction after having lived to adulthood with physical differences in the hand. The request also stems from requirements, more than personal preference and are likely to impact individuals with more and/or fused fingers.

Potential social and emotional gains from reconstructions have been highlighted in more recent publications. In older children with a variety of congenital hand differences, aside from reporting difficulty in hand function, many are bothered by appearance and the conditions' impact on social interactions.^{11,12} Long-term psychosocial effects from polydactyly in adults, have also been described in case reports.¹³

While the reasons were clustered as they were stated in the records, patients looking to improve appearance may also have overlapping psychosocial concerns. The need to have a more "normal looking" hand points to more than just the cosmetic advantage but also relates to not standing out and being able to comply with norms. In this population, the gains may also extend to being able to comply with uniformity and standards for safety especially in certain occupations. Hygiene concerns were rarely encountered but were naturally related to the small sinuses in constriction band deformities.

It is noteworthy that the need to improve function was a reason from more than a fifth of the patients even though all patients were either employed or able to attend school prior to consultation. This may be a wish to improve some aspects of function like efficiency and ease of doing things. While the 4 patients who hoped for "improved function" did not have Fil-DASH scores available, the ten with available measurements, had 2 presenting with scores above 16. This is the highest threshold for what may be considered normal in previous normative studies in the age group in European countries.^{14,15} That appears to reflect the same proportion of patients who may have perceived significant limitations in function even if those were not stated as the primary complaint. The age range of this population also involves mostly young adults who may be more focused on better employment than anything else.

In a series of 10 pre-axial polydactyly in adults, Cetik et al.⁷ found that the main reason for admission was the "social consequences of cosmetic problems from the anomaly". While it is not widely discussed in publications, the presence of extra digits may be associated with good luck or is given a certain level of importance in certain cultures.^{16,17} This is anecdotally a common belief in the Philippines. This may have some impact on why appearance is not the most common complaint in this group. The relatively high number of cases with constriction bands also brings to question other factors that may have led to the delay and/or deterrence to consult early. This may be a subject for future investigations.

The results presented may aid in the understanding of the natural history of the common congenital anomalies if left untreated to adulthood. This may guide the physicians with more specific advice for parents on somewhat optimistic expectations, but at the same time mention opportunities that may be missed if modifiable conditions are not promptly addressed.

CONCLUSION

In summary, for congenital anomalies persisting beyond childhood presenting for reconstruction in a tertiary center, most have relatively satisfactory function. The most common reason for self-presentation for reconstruction is the possible gain in employment opportunities, followed by appearance.

The investigators acknowledge the limited variety of conditions represented in the series and may represent the more functional spectrum of conditions. The data collected did not always present specific details to understand the indications which are inherent to the study design. A qualitative exploration of the subject may be recommended for future studies.

Acknowledgments

We acknowledge the help of Ms. Cheenee Mabelle Calantoc of the University of the Philippines Manila - Research Grants Administration Office for her technical support in preparing the manuscript for submission.

Statement of Authorship

Both authors contributed in the conceptualization of work, acquisition and analysis of data, drafting and revising, and final approval of the version to be published.

Author Disclosure

The authors do not have any potential conflicts of interest with respect to this manuscript.

Funding Source

The authors received no financial support for the preparation, research, authorship, and/or publication of this manuscript.

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