

Original Research Article

Profile of congenital digital anomalies in children seen in two tertiary health facilities in Southern Nigeria

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ABSTRACT

Background: Anomalies of the digits are among the common congenital limb anomalies seen worldwide. Whereas the majority of these digital malformations are relatively minor and require simple treatment, some of them are complex and often associated with other anomalies and systemic disorders. Reports on the occurrence of these anomalies and their treatment are scanty in our region. The study sought to evaluate the burden of these digital anomalies among paediatric patients seen in our centres and outcome of treatment.

Methods: Study was carried out as a prospective study in two tertiary health facilities in Southern Nigeria between June 2014 and May 2019. Data obtained and analyzed were the patients' gender, age, family history, type of digital anomaly, the limb/s involved, other limb anomalies, systemic disorders, treatment given, outcome, and duration of follow up.

Results: A total of 113 children with anomalies of the fingers and or toes were seen during the study period, but 104 were analyzed. They comprised, 63 (60.6%) were males and 41 (39.4%) females giving a male female ratio of 1.5:1. Polydactyly was by far the commonest digital anomaly and the upper limbs were mostly affected. Associated anomalies contributed significantly to morbidity and mortality in those cases.

Conclusions: Digital anomalies are fairly common surgical problems in our region. Though majority of cases are mild and are easily treated, the more complex ones still pose a challenge to treatment in our practice. There is need for more interest and training among surgeons in our region to improve the surgical care and outcome of treatment of these digital anomalies in our region.

Keywords: Congenital, Digital anomalies, Foot, Hand, Outcome, Pattern, Treatment

INTRODUCTION

Anomalies of the digits are among the common congenital limb anomalies seen worldwide. They can occur as isolated entities or in association with other anomalies or syndromes.¹⁻⁴ The appearance of the digits contributes significantly to an individual's self-esteem and social confidence as well as functional capabilities. Career choices and professions requiring dexterity with the hands can be affected profoundly by the state of the digits of the hands. Similar challenges are seen with

digital anomalies of the foot in the use of some types of foot wears for sports or jobs. Whereas the majority of these digital malformations are relatively minor and require simple treatment, some of them are complex and often associated with other anomalies and systemic disorders. The aetiology is most often unknown but a number of genetic factors have been linked to these anomalies.^{6,7} Reported cases of occurrence in generations of a family support the role of genetic factors.^{8,9} Treatment in these complex cases can be challenging with resultant less than optimum functional or cosmetic

appearance of the hand or foot. Fortunately, the more complex cases tend to be rare.¹⁰ Reports on the occurrence of these anomalies and their treatments are scanty in our region. The study sought to evaluate the pattern of these digital anomalies among paediatric patients seen in our centres and their treatment.

METHODS

Study was carried out prospectively in two tertiary health facilities in Southern Nigeria from June 2014 to May 2019.

Inclusion criteria

Patients with congenital anomalies, malformations involving the fingers and toes of paediatric patients 12 years and below seen at the paediatric surgery, burns and plastic surgery outpatient clinics labour ward and post natal clinics of the two tertiary health facilities within the study period.

Exclusion criteria

Age beyond 12 years, acquired digit deformities and refusal to participate.

A proforma was designed to capture the following data: the patients’ gender, age, family history, type of digital anomaly, the limb/s involved, other limb anomalies, systemic disorders, treatment given, outcome, and duration of follow up. Data was subsequently collated and subjected to simple statistical analysis using the Microsoft excel spread sheet.

RESULTS

A total of 113 children with anomalies of the fingers and or toes were seen during the study period. Nine were excluded because of age above 12 years, and inability to establish whether malformation was congenital or acquired. Of the 104 patients analyzed, 63 (60.6%) were males and 41 (39.4%) females giving a male female ratio of 1.5:1. The age range at presentation was 0-12 years with a median at 3 months. There was history of digital anomaly in first degree relatives in 17 (16.3%) of the patients. The distribution of the anomalies according to the limbs affected is shown in Table 1.

The total number of affected limbs in the 104 patients was 204 and the types of digital anomalies encountered are shown in Figure 1. Polydactyly was by far most common anomaly of the digits encountered and was post axial in 74 (72.5%) and pre-axial in 28 (27.5%) cases. The hands were affected in 67 (65.7%) cases while the feet were affected in 33 (32.3%) cases.

Various degrees of digital loss, from partial to complete absence of some or all digits were encountered in 15 patients. Some complex combinations of digital mis

arrangement, deformation, hypoplasia, syndactyly and or polydactyly, was encountered in 13 patients. There were associated limb anomalies in 17 (16.3%) patients as shown in Table 2.

Table 1: Limbs involved in digital anomalies.

Limbs affected	Number of patients (n=104)	Percentage
Right upper limb only	6	5.8
Left upper limb only	11	10.6
Both upper limbs only	50	48.1
Right lower limb only	2	1.9
Left lower limb only	3	2.9
Both lower limbs only	23	22.1
All four limbs	9	8.7

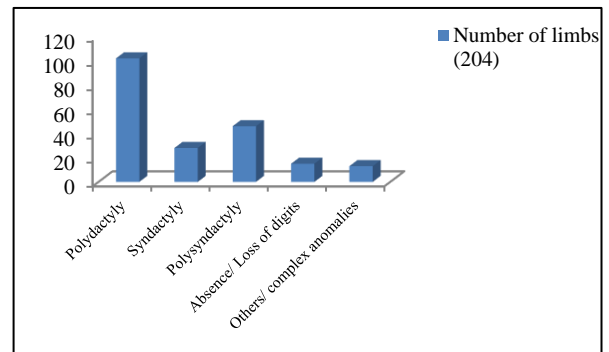


Figure 1: Types of digital anomalies.

Table 2: Associated limb anomalies.

Associated limb anomaly	Patients (n=17)
Congenital constriction rings	3
Hypoplastic calcaneum	5
Malformed tibia/ulnar	7
Hypoplastic limb	2

Sixteen patients were cases of multiple congenital anomalies giving a total of 28 associated anomalies in the 16 patients (Table 3). Of these, 4 cases of specific syndromes were identified- Down’s syndrome (trisomy 21) (2), McKusick- Kaufman syndrome (post axial polydactyly, heart defects and genital anomalies) (1), Oral-facial-digital syndrome (anomalies of mouth, face and digits) (1). Of the 204 cases seen, 157 received one form of treatment or the other (Table 4). Forty seven cases were not treated either because anomaly was so complex that treatment could not assure improvement (10 patients (4.9%)), patient had more serious condition requiring treatment (24 patients (11.8%)), or parents did not consider anomaly serious enough to warrant intervention (13 patients (6.4%)). There were 8 mortalities in this series, but none was related to the digital anomalies or their treatment.

Table 3: Other system disorders and syndromes associated with digital anomalies.

Anomalies/syndromes	Cases (n=28)
Craniosynostosis	1
Tracheoesophageal fistula or esophageal atresia	2
Congenital heart disease	7
Arthrogryposis multiplex	2
Duodenal atresia	3
Microtia and microphthalmia	2
Anorectal malformation	4
Undescended testis	5
Hypospadias	2

Table 4: Treatment offered.

Anomaly	Treatment	Cases (n=204)	%
Polydactyly	Simple suture ligation	68	33.3
	Simple excision/ amputation	23	11.3
Polysyndactyly	Digit amputation and/or osteotomy and flap movement	34	16.7
Syndactyly	Separation and/or osteotomy and flap movement	20	9.8
Complex anomalies	Staged procedures	12	5.9
Others	Watchful waiting	47	23

DISCUSSION

Anomalies of the fingers and the toes are among the common congenital anomalies seen among children in Southern Nigeria. This study points to a slight preponderance among male children and the familial occurrence of this condition, though there are no local reports of its incidence in our general population. Polydactyly is the commonest form encountered, particularly the type B post axial variant. This is in keeping with the findings by Holmes et al.¹¹ This present report is however in contrast with another report by Masada et al who reported more pre-axial polydactyly of the hand and post axial polydactyly of the foot. That report also showed a significantly higher incidence (17.8%) of cases with involvement of all four limbs compared with incidence of 8.7% in present study.¹² This indicates that involvement of all four limbs is relatively uncommon in our region and is similar to report by Hosalker et al.¹³ The majority of them required simple suture ligation or excision. Indeed many of them

had been tied and waiting to fall off or excised altogether by the birth attendants or referring clinician before presentation to us. The outcomes of these cases on follow up were generally excellent. Few of these cases of polydactyly however were part of groups of multiple anomalies, in which the digital malformation were only kept in view while the other more serious anomalies were addressed. Polysyndactyly and syndactyly were among the other frequently encountered digital anomalies which were amenable to treatment in the setting. The interdigitating triangular flap technique was deployed in most cases of syndactyly. The interdigitating rectangular flap technique recently described was not used as we were not conversant with it.¹⁴ On the other hand, desyndactylization, a technique described by Dowdy et al was deployed for some of our cases of syndactyly with successful correction.¹⁵ There was no case of macrodactyly in present series as had been reported in other studies. This highlights the rarity of this type of digital anomaly in our region.¹⁶⁻¹⁸ The more complex cases which tended to be combinations of anomalies were more intricate and often required multi-disciplinary approach involving paediatric and plastic and reconstructive surgeons. Microsurgical toe-to-hand transfers as well as the non-vascular phalangeal transfers which have been reported with excellent results in cases of absent digits were not feasible in our practice due to lack of requisite equipment and manpower.^{19,20} These cases of congenital absence or amputation of the digits were all isolated and did not involve more than one limb in any given patient. However there has been a previous report of a case in which there was involvement of all the limbs.²¹ Whereas improvements in cosmetic outlook and function were achieved with staging of reconstruction in some of those complex cases, some were left alone as we were not convinced that our treatment would improve the outlook. The most challenging cases of digital anomaly encountered in this study were those who had significant associated limb anomalies. From this study it was noted that such associated limb anomaly should be considered in planning whether or not to intervene and what treatment to offer in these cases. There was no mortality in this series due to the digital anomalies or their treatment. However other anomalies in association are the major determinants and contributors to mortality in these cases. This study reveals that there is need for more training and more efforts to enable us treat and improve on the results of treatment of the more complex cases of digital anomalies as is the case in other regions of the world.²²

CONCLUSION

Digital anomalies are commonly encountered in our practice in Southern Nigeria. Though majority are mild and simple to treat with excellent outcome, some more complex cases are more tasking to treat, and require multi disciplinary approach. Despite relative improvements in surgical care in our region, many of the more complex forms are still left untreated with all the negative impacts

on the psychosocial and functional integrity of the patients. There is need for more interest and training in the specific area of reconstructive surgery for children with anomalies of the digits.

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Conflict of interest: None declared

Ethical approval: The study was approved by the Institutional Ethics Committee

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