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Study of Macrodactyly in the Plastic and Reconstructive Surgery Department of Aristide Le Dantec Hospital in Dakar: About 22 Cases

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Abstract

Background: Macrodactyly is a congenital enlargement of the digital volume involving both the soft parts and the skeleton. The therapeutic prognosis, despite conservative treatment, is disappointing, sometimes leading to the choice of a radical procedure such as amputation. The aim of our study was to evaluate the epidemiological, clinical and therapeutic aspects of macrodactyly in our department.

Patients and Methods: We collected the records of 22 patients with macrodactyly, received between 2007 and 2022 in the department of plastic, reconstructive and aesthetic surgery of the National university hospital Aristide Le Dantec in Dakar. The parameters studied were age, sex, location of the disease, number of fingers or toes affected, and surgical management.

Results: The mean age of the patients at the first consultation was 6.9 years. The sex ratio was 1. The mean time to consultation was 78 months. The average number of affected fingers or toes was 1.53. We found as many macrodactylies on the right as on the left. Toes were more frequently affected than fingers with 71.8% of macrodactyly. In terms of treatment, 63.6% of the patients seen in consultation were treated. A reduction plasty alone was performed in 18.2% of patients, a plasty associated with an epiphysiodesis in 18.2% of patients and an amputation in 22.7% of patients. Post-operative care was simple in all patients. With an average follow-up of 12 months, a majority of patients were satisfied with a good recovery of the functionality of the affected limb.

Conclusion: Macrodactyly is one of the most difficult congenital limb defects to manage. The complex treatment and uncertain results often force the practitioner to opt for radical treatment.

Keywords: Macrodactyly; Finger; Toe; Surgery

Introduction

Macrodactyly is a congenital enlargement of the digits involving both the soft tissues and the skeleton. It is also known as lipomatous macro dystrophy or local gigantism. The exact aetiology is poorly understood, however vascular abnormalities and genetic factors (PIK3CA mutation) have been implicated. There are two main treatment modalities: conservative treatment consisting of reduction plasty with or without epiphysiodesis and radical treatment consisting of amputation of the involved digital radii. The aim of our study was to evaluate the clinical and therapeutic epidemiology of macrodactyly in our department.

Patient and Methods

This study was conducted in the department of plastic, reconstructive and aesthetic surgery of the National university hospital Aristide Le Dantec in Dakar.

Between 2007 and 2022, twenty-two patients with macrodactyly were seen in the department. The parameters studied were the following

Epidemiological factors

- Age and sex
- Time to consultation
- Notion of parental consanguinity
- History of macrodactyly or other malformation in the family; clinical aspects.
- Number of affected rays
- Laterality
- Notion of other associated digital malformation (syndactyly, polydactyly); therapeutic aspects
- Surgical procedure
- Results and complications

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Results

Epidemiological factors

The mean age of our population at first consultation was 6.9 years with extremes of 20 days and 35 years. In our cohort, 81% of the patients (n=18) were under 16 years of age at the first consultation. There was a sex ratio of 1 with as many women as men. The average consultation time was 6.5 years (extremes 20 days and 33 years).

A notion of parental consanguinity was found in 13.6% of the patients (n=3) and no notion of macrodactyly or other limb malformation was found in the patients' families.

Clinical aspects

On average, there was an involvement of 1.53 radii (extremes 1 and 3). The involvement was unidigital in 59% of patients (n=13).

Concerning laterality, there was as much involvement on the right as on the left.

The lower limbs were more frequently affected than the upper limbs, with 71.8% of cases.

The most affected radii were the 1st and 3rd radii of the lower limbs. Figure 1 shows the distribution according to the affected ray.



Therapeutic aspects

In total, 63.6% (n=14) of the patients seen in consultation were treated. Two patients had decided to refrain from treatment, five patients had been lost to follow-up and one patient had preferred to be treated abroad.

In terms of treatment modalities:

- 4 patients had had a reduction plasty alone
- 4 patients had a reduction plasty associated with an epiphysiodese
- 1 patient had epiphysiodesis alone
- 5 patients underwent ray amputation (Figure 2)



Figure 2: Macrodactyly of the 2nd ray of the right lower limb; a and b: Before surgical treatment by ray amputation, c: After amputation.

Post-operative care was simple in all patients. No complications such as infection, bleeding or suture loosening were noted.

With an average follow-up time of 12 months, the majority of patients were satisfied with the recovery of the affected limb's functionality and, above all, with the ease of wearing shoes.

We performed a revision in 2 patients due to a still significant residual mass. The reoperations consisted of:

- A 2nd reduction plasty
- A ray amputation after an epiphysiodesis during the first operation

Discussion

Epidemiological factors

The average age of our patients at the time of the first consultation of 6.9 years is relatively high compared to western studies where this age is around 1 to 3 years: 1.3 years for Golyana [1] in Russia and 3.6 years for Chen [2] in China. This delay could be attributed to the socio-economic conditions of the general Senegalese population. Indeed, 38% of the Senegalese population lives on less than 1.90 USD per day [3], *i.e.* about five times the price of a specialised consultation; in addition, access to the specialist is difficult, as the country has only one plastic surgery department to date.

We found as many men as women affected by macrodactyly. The sex ratio found in the literature was variable. Some studies found as many women as men and others a higher number of women [2] or men. Ultimately, there does not appear to be a correlation between macrodactyly and gender.

The notion of an associated digital malformation was often found, particularly syndactyly.

Syndromic associations, particularly with klippel trenaunay and proteus syndromes, have been reported in the literature. However, our study only found isolated macrodactyly.

It would be tempting to correlate macrodactyly with consanguineous marriage, as consanguinity is often linked to many congenital malformations. However, not all congenital anomalies are hereditary. The rate of consanguinity in our population was much lower than the rate of consanguinity in the general Senegalese population, which was 48% [4].

Clinical aspects

We found a majority of uni-digital attacks, mainly on the lower limbs, in contrast to the literature, where attacks on

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several rays of the lower limbs predominate. This discrepancy between our results and those of the literature could be explained by the small size of our cohort compared with Asian studies which had 7 to 8 times larger cohorts [5]. This discrepancy could also be explained by a selection bias in the aforementioned studies, as patients with upper limb damage are more likely to consult because of the functional impairment but also because of the greater aesthetic damage than in the lower limbs.

Lower limb damage also affects patients with difficulties in wearing closed shoes or the inability to participate in certain sports or play activities with other children at school.

As for the affected side, as for the sex, there was a great variability in the literature suggesting that there is no correlation between laterality and macrodactyly.

Therapeutic aspects

Management is essentially surgical and results can be inconsistent. Due to the variety of presentations, there is no stereotyped therapeutic approach and the modalities of treatment are left to the discretion of the surgeon.

More than half (63.6%) of the patients in our study had received surgical treatment.

Patients were advised to refrain from treatment and to be monitored after careful assessment of the benefit-risk balance. These included patients in whom macrodactyly had little effect on limb functionality.

The fact that 6 patients were lost to follow-up after the first consultation could be attributed to the cost of surgery, patient apprehension about surgery, but also to the relief of discovering that macrodactyly is relatively harmless and not life threatening.

Different therapeutic approaches exist for the management of macrodactyly, grouped into 2 main groups: Conservative and radical treatment. In the conservative treatment we find epiphysiodesis which allows us to act in a preventive way on the angulation and the length of the affected ray. We also have softtissue reduction of the ray by degreasing. Radical treatment consists essentially of amputation of the affected ray.

Radical treatment is rarely recommended in the first instance. However, it may be necessary when the size of the affected ray impairs the functionality of the limb and affects the patient's independence and comfort. The choice of treatment also depends on whether it is static or progressive. The choice of the type of treatment therefore requires a careful assessment of the benefit-risk balance by the surgeon after consultation with the patient or his/her parents. The choice of the type of treatment may also depend on social reasons. Indeed, the cost of iterative plastic surgery can be daunting for patients with limited financial resources and the follow-up difficult for patients living outside the capital.

Our study found 9 cases where conservative treatment was chosen against 5 cases where amputation was necessary. Two patients who were more affected by aesthetic than functional problems were advised to refrain from treatment and to be monitored after evaluation of the benefit-risk balance. These results are similar to those reported in the literature, where the majority of epiphysiodesis and reduction plasty procedures are also performed. Indeed, Chen [2], Cerrato [6] and Ishiga [7] found a majority of patients in whom conservative treatment was preferred. However, we found two series with a majority of radical treatment, including one with 100% ray amputation [8,9]. It should be noted, however, that these two studies only observed macrodactyly of the foot and that radical treatment may have been preferred in these cases in the context of making it easier for the patient to wear shoes.

Evolution

No early or late complications were found in our series.

However, some complications have been reported in the literature, such as infection, necrosis, loosening of sutures, hypertrophic or retractile scars.

At the end of the healing process, there was an improvement in the function and appearance of the affected limbs, with an improvement in the wearing of shoes and walking with an average follow-up of 12 months.

A single degreasing plasty may not be sufficient to reduce the volume of the ray. A second plasty had to be performed in one of our patients due to dissatisfaction with the results, as well as a ray amputation.

Conclusion

The management of macrodactyly is complex and the results of conservative treatment are sometimes mixed. Radical treatment is a feasible option after a comprehensive analysis of the clinical and socioeconomic context and informed consent of the parents.

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Competing and Conflicting Interests

We have no conflict of interest

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