



Dupuytren's Disease or Nodular Fasciitis in a Black African Child. The First Clinical and Histological Observation?

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How to cite this paper: Kibadi Kapay, A., Meiers, I., Kabeya Katonkola, J. and Moutet, F. (2022) Dupuytren's Disease or Nodular Fasciitis in a Black African Child. The First Clinical and Histological Observation? *Open Access Library Journal*, 9: e9205.
<https://doi.org/10.4236/oalib.1109205>

Received: August 11, 2022

Accepted: September 4, 2022

Published: September 7, 2022

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Abstract

We report the case of a 10-year-old child, African of black skin and free of any Caucasian genetic load, victim of a traumatism of the palm of the left hand. It was a fall to the palm of the hand, without wound or hematoma. This is followed a few months later by the appearance at the site of the trauma of a firm nodule, painful when locking the socket. Surgery was performed a year after the trauma, at the age of 11, a fibrous tumor was discovered, nodular mass, affecting the middle superficial palmar aponeurosis at the expense of the pretending bands. Surgical treatment had consisted of excision of the nodular fibrous tumor with the immediate aponeurotic environment. Histopathological analysis of the surgical specimen and immunohistochemical examination had confirmed Dupuytren's contracture (superficial palmar fibromatosis). We also mention in the differential diagnosis, the diagnostic of nodular fasciitis which is rare, but the most probable diagnosis in the absence of retraction and in the context of a trauma with a similar histology. This study is probably the first case report which showed that the Dupuytren's disease or nodular fasciitis exists in black African children, free of any Caucasian genetic load.

Subject Areas

Surgery & Surgical Specialties

Keywords

Dupuytren's Disease, Nodular Fasciitis, Hand Trauma, African Black Child, Histological Confirmation

1. Introduction

Dupuytren's disease, also known as the Dupuytren's contracture, is a chronic, benign, progressive, fibro-proliferative condition that results in abnormal scar-like tissue developed from the palmar aponeurosis of the hand [1]. It usually affects men over the age of 40 [1]. It is very rare to find it in the pediatric age [2]. The studies conducted on the Dupuytren's disease and concerning black-patients are limited and for there, were carried out in small samples [3] [4]. The majority of these studies are primarily conducted in African adults. The authors don't find in the literature the studies with reporting the histological confirmation for the Dupuytren's disease in a black African child. Many treatment options for Dupuytren's disease currently exist [5] [6] [7].

This study aims to draw the attention of clinicians to this rare observation on black skin in pediatric hand surgery and to the differential diagnosis. The originality of this study lies in the rarity of such studies with the first report of such an observation.

2. Case Report

A 10-year-old child, African with black skin and free of any Caucasian genetic load, suffered a trauma to the palm of his left hand during a child's game. It was a fall to the palm of the hand, without wound or hematoma. This is followed a few months later by the appearance at the site of the trauma of a firm nodule, painful when locking the socket. The hand x-ray was normal. Ultrasound showed a 16 × 10 mm tissue lesion on the palmar surface without bone involvement, with the appearance of an inflammatory granuloma on the palmar surface of the hand versus an old hematoma given the history of trauma. Surgery performed one year after the trauma, at the age of eleven, discovered a fibrous tumour, nodular mass, involving the middle superficial palmar aponeurosis at the expense of the pretendinous bands (**Figure 1**).



Figure 1. Fibrous tumour, nodular mass, involving the middle superficial palmar aponeurosis at the expense of the pretendinous bands.

Surgical treatment had consisted of excision of the nodular fibrous tumor with the immediate aponeurotic environment. The results of the surgery were very satisfactory: disappearance of the swelling and pain when locking the socket.

The result of the anatomopathological analysis of the excision piece sent to the Luc Olivier Laboratory in Belgium showed a poorly delimited multinodular lesion, with alternating hyper- and hypo-cellular, hyalinized areas (**Figure 2**).

The tumor was made up of spindle-shaped cells with elongated and vesicular nuclei with a small central nucleolus (**Figure 3**).

Histopathological observations appeared to Dupuytren's contracture (palmar fibromatosis) with no dysplasia or malignancy. The result of the immunohistochemical examination confirmed the histopathological appearance with a Dupuytren's contracture (superficial fibromatosis).

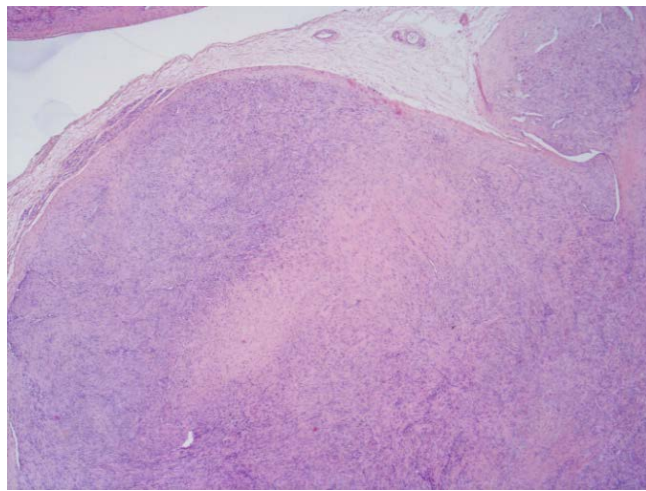


Figure 2. Palmar fibromatosis—multinodular, poorly demarcated lesion with alternating hyper- and hypo-cellular, hyalinized (4X) areas.

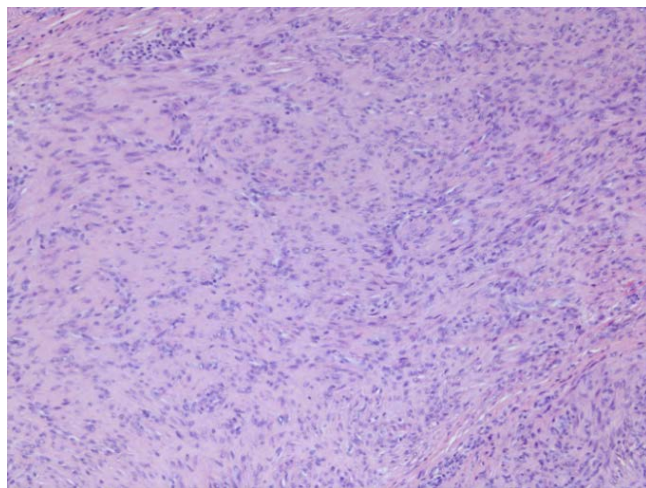


Figure 3. Fibroblasts are uniform, with normochromatic nuclei, small, elongated, sometimes vesicular, with a small central nucleolus (20x).

3. Discussion

3.1. First Case of Dupuytren's Disease in a Black African Child?

Dupuytren's disease has been reported on black patients: Mennen [6] reported only 5 cases of Dupuytren's disease in black South Africans, free of any Caucasian genetic load. Coulibaly *et al.* [7] reported a population of black Senegalese people included 20 men and 6 women averaging 63.5 years of age (range 45 - 77). In the United States of America, Saboeiro *et al.* [8] showed that the prevalence of the Dupuytren's disease in black patients was 130 per 100,000 inhabitants versus 734 for whites. These various African studies [6] [7] only concerned black African adult people.

The notion of an initial trauma in the palm of the hand was reported in our young patient. A single trauma can trigger the Dupuytren's disease, as was the case in our patient. These authors [9] therefore underline the influence of microtraumas. There is a relationship between the initial trauma and the development of the Dupuytren's disease in our patient, an African child with black skin, thus meeting the six criteria of Elliot and Ragoowansi [9] below: 1) existence of a true trauma to the palm of the left hand without pre-existing Dupuytren's disease lesion; 2) trauma to the left hand on the same side where the disease developed; 3) the patient was not within the age criteria predisposing him to the Dupuytren's disease or diathesis, our patient being 10 years old at the time of the trauma; 4) the disease appeared within a year of the trauma and he was operated on a year later, at age eleven; 5) appearance of a single nodule in the palm of the traumatized left hand; 6) the disease remains isolated to the part affected by the trauma, that is to say the palm of the left hand.

3.2. Histological Evidence of Dupuytren's Disease

In our patient, the analysis of the slides presents irrefutable proof of the Dupuytren's disease. We observed a multinodular lesion, poorly delimited with alternating hyper- and hypo-cellular areas, hyalinized (4X) and uniform fibroblasts, with normochromatic, small, elongated, sometimes vesicular nuclei, with a small central nucleolus (20X). Histological confirmation of the Dupuytren's disease is therefore essential given the differential diagnosis of the disease.

Differential diagnosis for Dupuytren's disease in children [10] [11] include nodular fasciitis, extra-abdominal fibromatosis, calcifying aponeurotic fibroma, infantile fibrosarcoma, fibroma of the tendon sheath, localised nodular tenosynovitis (giant cell tumour of the tendon sheath), fibrous hamartoma of infancy and infantile myofibromatosis. Calcifying fibroma affects mainly children and adolescents and presents as a relatively small mass most often located in the palm. Histologically, more significant difference is the invariable presence of calcified foci surrounded by plumper cells.

Goetzee & Williams reported the first histologically proven case of the Dupuytren's disease in a 14-year-old boy with flexion contractures of the ring finger and little finger with similar plantar fascia disease [12]. Bebbington and Savage

reported three histologically proven cases in children under 9 years of age, only two of which were infants [13]. Cheryl *et al.* [14] studied 108 cases of fibromatosis in children over a 25-year period, among which there were only three cases of Dupuytren's disease. The Dupuytren's disease is therefore rare in infants and children.

3.3. Nodular Fasciitis in a Black African Child?

We cannot provide to give solid evidence that the lump removed from the child's hand could effectively be a Dupuytren disease: the anatomical location of the nodule is very proximal and ulnar. The site seems to be more linked to the fascia of the hypothenar muscles in the absence of a retraction, one cannot conclude that the lump is a clinical Dupuytren. Although compatible with Dupuytren, the histology is nonspecific and there is unfortunately no specific marker for this pathology. We can thus not conclude only based on histology—a couple of differential diagnosis of post traumatic lump on young population are evoked, but we can't forget the nodular fasciitis which is rare but the most probable diagnosis in the absence of retraction and in the context of a trauma with a similar histology.

Indeed, factors such as the age of the patient, the rapid appearance of a painful subcutaneous posttraumatic nodule at the level of the hypothenar eminence, as well as the macroscopic aspect of the nodule in continuity with the muscular fascia, visible on the peroperative photograph (**Figure 1**), are all these elements which can plead also in favor of nodular fasciitis.

We do not find in the literature the studies with reporting the clinical and histological observation of nodular fasciitis in the palmar surface of hand in a black African child. This would therefore be the first report.

3.4. Therapeutic Modalities

In our patient, the treatment was essentially surgical, consisting of excision of the nodular fibrous tumor with its immediate aponeurotic environment. Surgery remains the standard treatment for the Dupuytren's disease. Currently, there is no proven preventive treatment or effective medical treatment to cure the Dupuytren's disease in children. We can nevertheless recommend injections of corticosteroids in the painful nodule as in adults. Furthermore, no adequate study has been performed on the relationship between age and the effects of injection of steroids, collagenase and enzymes in the pediatric population [2] [5] [6] [7]. The safety and effectiveness of these conservative measures have not been established [2] [5] [6] [7]. Limited fasciectomy and limited dermofasciectomy are the techniques mostly used.

4. Conclusion

Trauma to the palm of the hand in a black African child may be the basis for the development of the Dupuytren's disease or nodular fasciitis. Anatomopatholog-

ical and immunohistochemical examination is necessary for confirmation and differential diagnosis of the disease. Surgery, if necessary, involves excision of the nodular fibrous mass with its immediate aponeurotic environment. It emerges from this study, probably the first report of such a case, that the Dupuytren's disease or nodular fasciitis, although rare, exists in black African children free of any Caucasian genetic load. The report shows also the importance and the difficulties of differential diagnosis of nodules and fibrotic bands in children's hands for the surgeons or pathologists.

Statement of Patient Consent

Written informed consent was obtained from the guardian of the patient for publication of this case report and accompanying images.

Conflicts of Interest

The authors declare no conflicts of interest.

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