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Original article

Dupuytren's disease: A reality in Black Africa

La maladie de Dupuytren: une réalité en Afrique Noire

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ABSTRACT

Long regarded as a disease exclusively found amongst Northern Europeans, Dupuytren's disease was seldom studied amongst Black Africans. Thus, we sought to study the impact of Dupuytren's disease, its etiological, clinical and evolutionary peculiarities on a segment of the Senegalese population. This study analyzed data derived from clinical observations carried out between January 2006 and December 2018. It involved Senegalese subjects with Dupuytren's disease, the patients' history, profession, habitus, clinical findings, therapeutic modalities and disease staging. The population included 20 men and 6 women averaging 63.5 years of age (range 45–77). None of the patients reported a family of Dupuytren's disease. Twelve patients had diabetes, 11 were smokers and 22 were engaged exclusively in manual labor. The condition was bilateral in 14 cases. Tubiana stages N, I, II, III and IV were found in 31, 15, 9, 5 and 6 rays, respectively. Conservative treatment was done in 11 patients. Surgical treatment was carried out in the other 15 patients: needle fasciotomy ($N = 10$) including two bilateral involvement and open fasciectomy ($N = 7$). Functional outcomes were satisfactory. Lesions were all stable in the short and medium term. Two patients had progressive lesions on a longer-term basis. Dupuytren's disease is real among Afro-descendants from Senegal even though it is seldom studied. Based on the patients' recollection of Dupuytren's disease in their families, heredity is not yet a proven factor. The early forms are more common, and the lesions remain stable for a long time.

Mots clés :
Dupuytren
Rétraction
Aponévrose
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RÉSUMÉ

Longtemps considérée comme une maladie de sujets d'ascendance nord-européenne, la maladie de Dupuytren a été rarement étudiée chez les Africains. Le but de ce travail était d'étudier l'impact de la maladie de Dupuytren, ses particularités étiologiques, cliniques et évolutives sur un segment de la population sénégalaise. Cette étude prospective a été menée de janvier 2006 à décembre 2018 chez les patients reçus pour la maladie de Dupuytren. Les paramètres étudiés étaient les antécédents, la profession, l'habitus, les aspects cliniques, les modalités thérapeutiques et évolutives. Il s'agissait de 20 hommes et 6 femmes âgés en moyenne de 63,5 ans (de 45 à 77 ans). De mémoire, aucun des patients n'a signalé un cas similaire de la maladie de Dupuytren dans sa famille. Douze patients étaient diabétiques, 11 fumeurs, 22 avaient une activité exclusivement manuelle. La localisation était bilatérale dans 14 cas. Les stades N, I et II, III et IV de Tubiana ont été trouvés respectivement sur 31, 15, 9, 5 et 6 rayons. Un traitement conservateur a été effectué sur 11 patients. Le traitement a été chirurgical chez les 15 autres par une aponévrotomie à l'aiguille ($n = 10$) dont 2 bilatérales ou une aponévrectomie ($n = 7$). Les résultats fonctionnels à court et moyen termes étaient satisfaisants. Deux patients ont eu des lésions évolutives à long terme. La maladie de Dupuytren est réelle chez les sujets d'ascendance négro-africaine

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même si elle a été rarement observée. Le facteur héréditaire n'a pas encore été prouvé. Les formes de début sont plus communes et les lésions restent longtemps stables.

1. Introduction

Dupuytren's Disease (DD) was named after Baron Guillaume Dupuytren, a famous 19th century French surgeon. As early as 1680, it was first described by Felix Plater followed by Henry Cline Jr in 1818 [1]. DD was clinically described as a "palmar nodule on the line of the ring or small finger". Usually, it affects first the metacarpophalangeal joint and then the proximal interphalangeal joint [1]. Found initially mainly among Scandinavian, English and Irish people, it spread southwards across Europe from the 9th to the 13th century. This propagation is presumed to be from Northern Europe through waves of Viking invasions [2]. DD is rather rare (1% to 2% of total population) although its prevalence amongst people of Northern European descent can reach 30% [3]. Its genetic dimension is a widely acclaimed tracer among White Europeans [3–5]. Cases found in Africa and Asia were considered sporadic and even exceptional. In view of the latest published data in these areas, it should be noted that more and more patients are diagnosed with DD [6,7].

In Africa, it is not clear whether the many apparently sporadic cases have a genetic basis because the disease is much less known and has rarely been studied [6,8–13], particularly among Black Africans. Despite DD being easy to diagnose, its impact is widely underestimated in sub-Saharan Africa. Indeed, based on the largest sample to date (48 cases in South Africa), Sefeane and Mwangi emphasized the need to broaden DD knowledge and statistics on people of African descent [6]. They did not find any conclusive evidence on DD prevalence among Black Africans. Several explanations were put forward as why DD is under-reported in Africa. First, patients have long endured the chronic and insidious evolution of DD, which explains the delays in consultation, especially among elderly people who may no longer be working [6]. Furthermore, lack of knowledge about this disease in sub-Saharan Africa has contributed to confusion about cutaneous sequelae and tendinous retractions. Treatment is a function of DD stage. The treatment can be conservative or surgical; a strong potential for recurrence within 5 years defines DD evolution [14].

The purpose of our work was to study the impact of DD in our Senegalese population, along with its etiological, clinical and evolutionary peculiarities.

2. Patients and method

The study was carried out in a hand surgery unit of a public University Hospital, the only one of its kind in Senegal offering specialized hand surgery consultations until 2016. Senegal has 16 million inhabitants. Patients with particular hand pathologies are referred in this hospital. The Orthopedic Department receives a yearly average of more than 41,000 patients, mostly with upper limb conditions (32%) of non-traumatic origins.

This study lasted 13 years from January 2006 to December 2018. It included all patients with DD at the Orthopedic Department whether they underwent surgical treatment or not and were followed for at least 1 year. The main criterion for inclusion in the survey was that patients had to be exclusively of Black African descent. During this period, we operated on six Cape Verdean and four European DD patients. These patients were not included in the study. Furthermore, selected parameters under study were age, sex, personal and family history, profession, habitus, clinical manifestations, therapeutic modalities and evolution.

Clinical examination involved looking at the affected side (right or left hand), lesion site (digital and/or palmar), number of rays impacted, presence of nodules, umbilication and digital retraction. Severity of retraction was evaluated according to Tubiana staging system for DD [15] and its associated forms. Therapeutic modalities were pooled into three treatment methods: no treatment, needle fasciotomy and fasciectomy. Outcomes were studied on a short-term (less than 6 months), medium-term (between 6 months and 2 years) and long-term basis (greater than 2 years) depending on whether the lesions progressed or recurred.

3. Results

Overall, this study involved 26 patients. Four DD patients were treated from 2006 to 2012 and 22 additional patients were treated between 2013 and 2017: 2013 (3 cases); 2014 (1 case); 2015 (7 cases); 2016 (5 cases); 2017 (6 cases). They were 20 men and 6 women (sex ratio 3.33). Age averaged 63.5 years (range: 45-77 years), with 10 patients 40 to 59 years old, 14 patients 60 to 74 years old and 2 patients 75 to 90 years old. Twenty-two of the patients were exclusively manual laborers (farmer: 5; office worker: 7; housewife: 5; breeder: 2; teacher: 5; policeman: 2). Twelve had type 2 diabetes. Eleven were active smokers. Alcohol consumption was reported by eight patients. None of the patients reported a similar case in their family and they had no history of White European admixture for at least two generations. DD was bilateral in 14 patients (Fig. 1). Among the remaining 13 patients with unilateral lesions, the dominant side was prevalent in 7 cases. Twenty-six patients had 66 rays (R) affected in the right or left hand. The 4th ray (R4) was involved in 32 cases. Simultaneous ray involvement occurred in 65% of hands and concerned three pairs of rays (3rd and 4th rays, 4th and 5th rays and 3rd and 5th rays). Fig. 2 summarizes which rays were impacted by DD. All stages of the Tubiana system [15] were found (Fig. 3). Stage N was the most prevalent in 31 rays. Stages III and IV were found in 5 and 6 rays. Adhesions to the deep plane were limited to advanced forms (Fig. 1, Fig. 4). The table test was positive in 16 hands. Epilepsy,

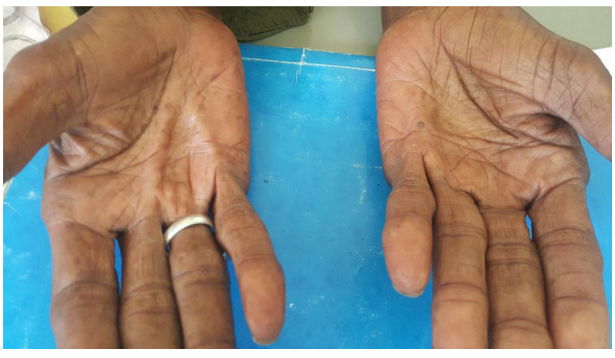

Fig. 1. Picture showing bilateral involvement with stage II on the right hand (3rd ray and 5th ray) and left hand (5th ray).

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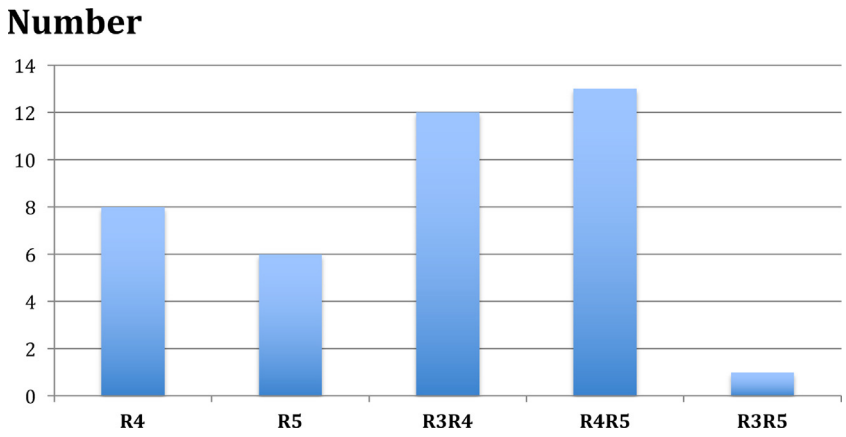

Fig. 2. Distribution of Dupuytren's disease by ray (R) R4: 4th ray; R5: 5th ray; *R3R4*: 3rd ray and 4th ray involved; *R4R5*: 4th ray and 5th ray involved; *R3R5*: 3rd ray and 5th ray involved.

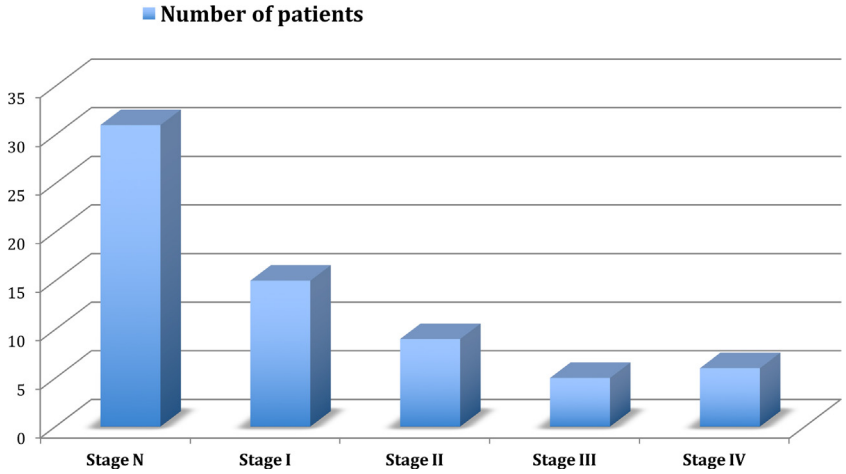

Fig. 3. Severity of Dupuytren's disease on Tubiana's disease staging system. N: nodule.

tuberculosis, gout and hyperuricemia were evaluated in 18 patients but not found.

Conservative treatment (i.e., therapeutic abstention) was carried out on 11 patients, 4 of whom had bilateral lesions. Among the other 15 patients, surgical treatment was performed including 10 needle fasciotomies (Fig. 4) of which 2 were bilateral, and 7 fasciectomies (Fig. 5) with full extension of the rays. Complete closure was achieved in 5 patients. The open palm technique was applied to 2 patients. Cast immobilization was used in 9 patients for an average of 18 days (ranging from 7 to 21 days). Involved fingers were immobilized in extension, followed by a night posture splint for 1 month in 4 patients. For the others, self-rehabilitation was started one day after the surgery. The mean uncomplicated healing time was 15.7 days (range: 13 to 23 days). One patient had a cutaneous necrosis complication. Healing was achieved after 8 weeks.

In terms of outcomes, all patients were followed at least for 1 year, with follow-up time averaging 2.32 years (range, 14 months to 6 years). In the short and medium term, the lesions were all stable, without digito-palmar retraction. In the long term, 4 out of 16 patients were lost to follow-up. Among reviewed patients, two got worse: stage N to stage 1 and stage 1 to stage 2. One of the two patients was being monitored whilst needle fasciotomy was applied to the second patient. No recurrence was observed in the operated patients. No digito-palmar retraction occurred among other patients for up to 4 years (Fig.4c).

4. Discussion

One may infer from the data presented above that DD is indeed a reality in Senegal, a country mostly if not wholly populated with Black Afrodescendants. Over 13 years, we diagnosed 26 cases, i.e., 2 cases per year. This represents 0.0048% of all upper limb non-traumatic pathologies in our Orthopedic Unit. Recognizing that there is a major flaw in DD detection and care, all DD surveys point to the fact that DD is very rarely observed in sub-Saharan Africa. However, this overwhelming fact does not mean that DD never existed in continental and diaspora Africa. Indeed, Zaworski and Mann [16] described the first two DD cases in 1979 involving Afrodescendants in Miami, Florida. Subsequently, small case series of up to 8 cases each were published before 2018 [8–12,17–19]. The cumulative number of DD cases in Afrodescendant patients is 121 according to studies carried out by Sefeane and Mwangi in 2018 [6] and our study. In sub-Saharan Africa, the DD global prevalence rate is unknown because a sizeable number of potential patients do not consult with medical specialists; many go first to a bonesetter. As DD is an insidious disease that does not cause major functional discomfort at the beginning, it goes widely unreported in most cases especially in distant or underprivileged areas. Another adverse factor may be the scarcity of African hand surgeons who have experience diagnosing fascia as opposed to burn contracture, a very common pathology in Africa. However, it may be noted that a specialized workforce has been growing

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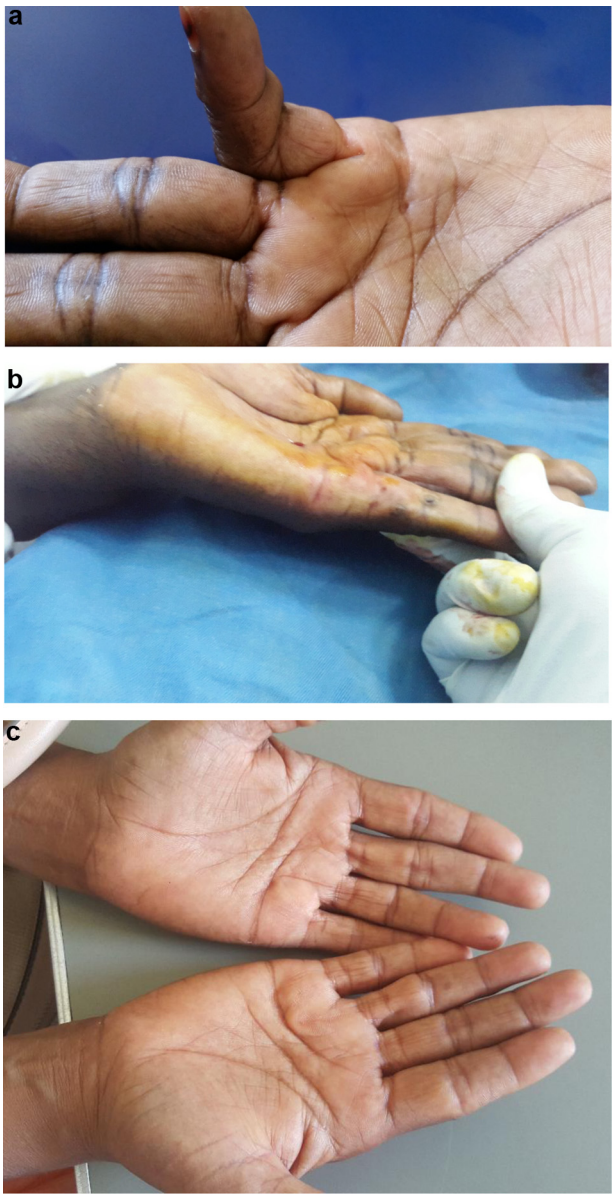

Fig. 4. Images showing outcome for stage III disease in the 5th ray of the left hand: before needle fasciotomy (A); after needle fasciotomy (B); 4 years later (C).

Fig. 5. Another stage III on the 5th ray: intraoperative view showing fibrous cord (arrow) before fasciectomy (A); full extension obtained after fasciectomy and complete closure (B).

locally and regionally over the years through a better mastery of hand surgery academia, teaching and referral.

Etiologically, we did not find a history of DD in our patients' families. They are Afrodescendants with no similar pathology in their family history, and no history of mixity with White Europeans for at least two generations. These antecedents were also not found in the other Black African populations interviewed elsewhere [6,8–11,13,16–21] except for one case [12]. Yet this genetic factor linked to DD has been studied in several studies [4,22,23]. In most cases, DD is transmitted from generation to generation within families and represents the most common connective tissue disorder [22]. The mode of transmission is not well understood and appears to be dominantly autosomal with variable penetrance and most often autosomal recessive matrilineal suggesting mitochondrial transmission [5,23]. Our finding led us to hypothesize that, in populations where sporadic cases are reported, genetics play only a small part in the occurrence of DD given its weak relationship with the demographic rate and the equally low prevalence of DD. According to Hindocha et al. [3], this situation suggests that there may be spontaneous genetic mutations causing the disease and that environmental factors may also play a role in the development of DD. However, the relevance of genetic traces of DD should be examined in Black African populations in a larger study because it is difficult to carry out a local study with sufficient statistical power. Indeed, it is still difficult to prove the genetic factor clinically because of the following obstacles:

- populations with limited life expectancy;
- absence of complaints from patients reporting DD at the treatment sites and from the offspring of DD patients;
- the low level of management of health services and their inability for the moment, in most cases, to create state-of-the-art patient databases.

Such a breakthrough could shed new light on the specific determinants of hereditary transmission.

The others etiological factors we noted are diabetes, smoking, alcohol consumption and manual labor in 12, 11, 8, 22 cases out of 26, respectively. The manual labor factor has been discussed most often. In European studies, diabetes was found in 3% to 33% of persons with DD [5,24] and tobacco use in between 16% and 21% [24]. It has been suggested that DD is the result of local hypoxia and chronic ischemia [5]. Although the mechanism has not been clearly established, these authors believe that alcohol influences the local circulation of the palm and produces adipose tissue lesions causing a fibrotic response and changes in prostaglandin production.

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As for diabetes, the association rate is between 3% and 33% with an average of 20% [5]. However, it should also be noted that this condition is very common at this advanced age even outside the occurrence of DD. The other factors included epilepsy, tuberculosis, gout and hyperuricemia [3,6,24,25]. These elements were not examined in the early stages of this study. These parameters were not found in the subsequent 18 patients. One DD case associated with sickle cell trait was found in Haeseker's study [19]. Regardless of the factors involved, its etiology remains unknown [2,3,24,26].

Clinically, advanced age of onset is widespread and supports results suggested elsewhere [1,3]. DD usually occurs in the fifth, sixth and seventh decades and the prevalence increases with age. No cases of early onset were found in patients in the African studies [6,8], unlike the European ones [1]. In these early cases, the disease usually leads to more pronounced contractures [26].

We noticed a clear male predominance. The reported sex ratio ranges from 1.5 to 9 [14,18,20,25,27]. Male predominance in the DD literature in Northern countries is generally explained by the existence of a genetic factor in up to 20% of the population [3,14,27–29]. The expression of the disease gene would be almost complete in men over the age of 75 but is of much lower penetrance in women, unless it arises from both parents [1].

Bilateral involvement was the most common feature and both hands were affected to varying degrees as observed in European populations [17]. Saboeiro et al. [20] think differently and hold that lesions are mostly unilateral among Black African people.

All disease stages were encountered in our population. The most prevailing forms in our series were less advanced. Digitopalmar fibrous cord adhesions (stages II, III IV) relative to the deep plane were limited compared to lesions found in Europeans operated in our Orthopedic Department during the same period. It was observed that sporadic cases tended to begin late and be less severe than familial cases [22].

No early onset lesions were observed, nor any associated forms such as Ledderhose or Lapeyronie's diseases [20]. Only one case out of the 121 found in sub-Saharan Africa was reported with an associated form [9], whereas the incidence of these associated forms is high, reaching 21% in White European studies [24,26].

Involvement of the 1st and 2nd rays was not observed. This site is estimated at 7% in the study by Loos et al. [24]. Simultaneous involvement of rays was most frequent, with the 3rd, 4th and 5th rays being more often involved than the 1st and 2nd rays. The association between the 3rd and 4th rays, on the one hand, and the 4th and 5th rays, on the other hand, was dominant, as was observed in a European study [2,3,14,29]. In our study, this association was mostly found in farmers, teachers and housewives. The frequent bending position of the last 2 fingers and the side force could be responsible for palmar fascia retraction.

Our treatments were similar with the literature [21] and based on DD stages. Given the limited degree of impairment, conservative treatment was adopted in 23 hands. Although risks are associated in cases of neurovascular pedicles, needle fasciotomy gave us much satisfaction in the shorter and longer term. Regarding the 7 fasciectomies that were performed, the medium-term results were good with healing and restoration of affected rays' joint range of motion. It should be noted that four patients were lost to follow-up more than 4 years later.

This study has limitations linked to the patients' advanced age (averaging 63.5 years) and a long follow-up duration explaining the large number of patients lost to follow-up. Other categories of risk factors relevant to DD such as hyperuricemia, tuberculosis and epilepsy were not examined in our early cases. Methodologically, a high-powered statistical study was not possible because of the small sample of patients for the reasons mentioned in the discussion.

5. Conclusion

DD is real among Black Africans even though its occurrences are seldom found on a significant scale. Relying on patients' recollection of DD history at the family level, heredity factors were not found to be relevant. In this study, the only etiological factors were diabetes, smoking, alcoholism and manual labor. Early forms were the most frequent and only isolated forms were found. The treatments we adopted were not different from the treatments reported elsewhere in the DD literature; treatment was a function of disease stage. Recurrences were not observed up to 4 years. In the near future, a higher number of hand surgery specialists may provide better DD identification, diagnosis and treatment to the point where a more robust statistical analysis will be possible. The continuation of our follow-up will allow us to eventually validate these findings. The introduction of immunohistochemistry studies and molecular biology may warrant further objective investigations on the genes linked to DD occurrence amongst sub-Saharan Africans and will improve our understanding DD pathophysiology.

Ethics approval and consent to participate

This study was approved by the Ethics Committee Board of Le Dantec University Hospital, Dakar, Senegal.

Consent for publication

All authors agree to consent for publication.

Disclosure of interest

The authors declare that they have no competing interest.

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