

# Incidence of Dupuytren's disease in Africans: A report of 48 new cases and a literature review

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## Abstract

**Background:** Dupuytren's disease is rare in the black population. It was initially believed it occurred only in males of north European origin and since the initial confirmed report of a male of African descent with the disease only a few other Africans have been reported with the condition in the literature. We wanted to conduct a literature search for all reports of patients of African descent with the disease and to report on our own numbers in our institution.

**Methods:** We searched our theatre records for all surgeries for Dupuytren's disease from 1997 to 2015 and identified all those that are of pure African descent. A comprehensive literature search in Pub Med, Google Scholar and Clinical Key was then done to identify all reported cases to date.

**Results:** From our records a total of 48 patients were identified. Of them, 43 were male and five were female with an average age of 58.7 years (29 to 75). From the literature, only 462 patients were ever reported with the condition to date. One paper which constituted the majority of these cases was treated with caution as there were questions of reliability about the true ethnicity of the patients, leaving only 50 cases ever reported to date prior to our 48.

**Conclusion:** The true incidence of Dupuytren's disease in Africans cannot be accurately determined but it is an extremely rare condition. Population studies should be encouraged so as to come up with an accurate incidence and disease burden.

**Key words:** Dupuytren's disease, contracture, black population, Africans

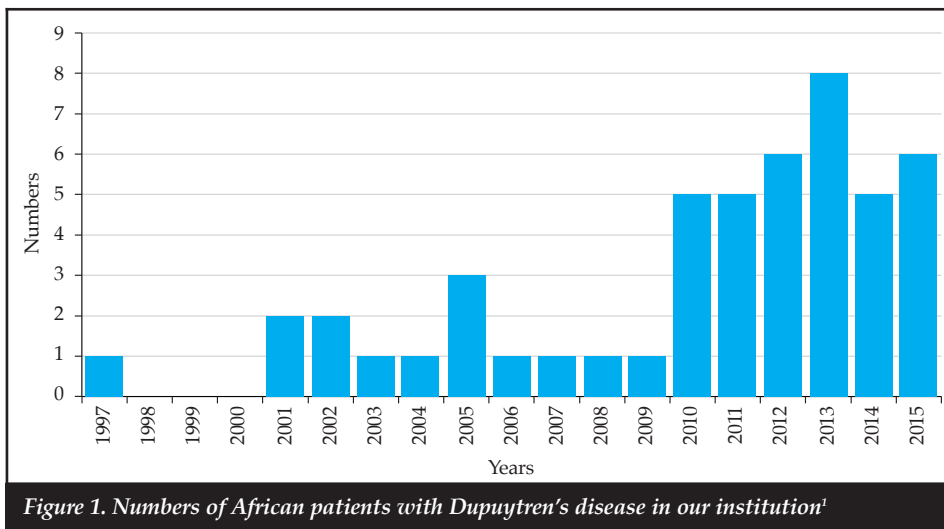
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## Introduction

When Mennen and Grabe<sup>1</sup> accurately reported on the discovery of a patient of pure African descent with Dupuytren's disease in 1979, little was known of the disease ever affecting other ethnic groups, especially Africans. This condition was first described by the French surgeon Guillaume Dupuytren in 1834 and it has largely been believed to occur in males of Northern European descent, and that migratory patterns of society distributed this condition to other parts of the world.<sup>2</sup> Since the first initial report of a black patient with the disease,<sup>3</sup> a few more cases

have been reported in black African patients<sup>4-20</sup> and other ethnic groups.<sup>21,22</sup>

We have seen an increasing number of patients of pure African descent that have undergone surgery at our institution in recent years. We conducted an audit of our theatre records to identify black African patients with Dupuytren's contracture who underwent surgery in a referral-based patient population. We also undertook a literature search of all reported cases of the condition in patients of African descent to try and determine the true incidence and prevalence in this population group.



## Methods

We searched our hand unit database for all patients who underwent Dupuytren's disease contracture surgery between 1997 and 2015. We then determined the number of these patients who were African by way of surnames and their location (black townships), which is a useful method in South Africa. Our hospital is situated in Soweto which is a black township with an estimated population of about 6 million. We then compared this number to the number of total surgeries performed in the unit during the same period and worked out our local prevalence. The number of these patients were compared year on year and charted to determine any possible trends (*Figure 1*).

The two authors independently undertook a literature search of all reports of patients of African descent having been diagnosed with Dupuytren's disease using the search words Dupuytren's, disease, contracture, blacks, Africans in PubMed, Google Scholar and Clinical Key in order to find the numbers of patients that were ever reported. The search included all kinds of studies (prospective, retrospective, case reports, letters and communications). Included were all studies or reports of patients of African descent with the disease, and excluded were those studies that reported on non-African patients. We selected all relevant abstracts and among those selected, the most relevant were identified and the full articles and reports thereof then fully reviewed. The authors then looked at any possible bias (about the true ethnicity of these patients) in these articles and extensively reviewed those that could be potentially controversial or misleading.

## Results

From our theatre records we were able to document 70 patients who underwent 73 surgeries for Dupuytren's disease from 1999 to 2015 by different surgeons in our hand unit. There were 65 males and five females with an average age of 58.7 years (29 to 75). Out of this group, 48 were identified as pure Africans as per our criteria; one had bilateral disease; and fasciectomy was done in different settings (*Figures 2 and 3*).



*Figure 2. Left hand of an African patient with bilateral Dupuytren's disease*



*Figure 3. Right hand of the same patient as in Figure 2 during the second operation*

In this period a total of 22 468 patients were operated for various conditions in our operating rooms. **This would then mean that 0.2% of these patients were black patients who presented with Dupuytren's disease and underwent surgery.** We also found that there has been an increasing number of African patients year on year since 1999 (*Figure 1*).

From the literature search in PubMed, Google Scholar and Clinical Key using our key words, there were initially 873 articles to consider. Sixty-six of these articles were deemed relevant and their abstracts were reviewed. Sixteen of these abstracts were selected as relevant and their full articles were comprehensively reviewed and analysed. From these 16 articles dating from 1974 to present, 462 patients of African descent have been reported with Dupuytren's disease worldwide. Adding our 48 cases to this total would bring the total to 510 African patients ever reported with the disease worldwide to date.

## Discussion

Hindocha *et al.*<sup>23</sup> described the prevalence of Dupuytren's disease in northern Europe as between 18 and 39%, North America 4 to 26%, Oceania 23% and Japan 19% in an epidemiology evaluation of the disease in 49 papers from across the globe.<sup>23</sup> Africa and the rest of Asia had only sporadic cases in this report. In a Japanese study, Egawa *et al.*<sup>21</sup> suggested that the incidence of Dupuytren's disease in Japan differs little from that in northern Europe. In their series of 1 154 individuals over 60 years of age, 19.7% of the men and 9% of the women were found to have the disease. There have not been any specific epidemiological studies of the incidence and prevalence of the disease in Africans, and indeed most papers reviewed in our report were case reports.

The question of Dupuytren's disease occurring in black patients of pure African descent was settled by Mennen and Grabe by way of genotyping in 1979. Since then other authors have gone to great lengths to try to prove the bona fides of the ethnicity of these patients.<sup>11,13,14,19,20</sup> It has been asserted that the disease is rare in non-white races but there are no population studies to support this. Most population studies are from northern Europe and Australia where the disease is prevalent.

A ten-year retrospective study of the Department of Veteran Affairs records showed that there are similarities in the characteristics of the disease in black and white races.<sup>18</sup> In both groups the disease has a late onset and affects predominantly the ulna digits. Unlike the disease in whites, it is rarely bilateral in blacks and it is rarely reported. Mitra and Goldstein<sup>12</sup> reported on eight patients in whom the disease is often bilateral and associated with manual labour. The differential prevalence among the racial groups, geographic distribution pattern and familial predilection suggests a genetic component to the pathogenesis of the disease.<sup>18</sup> It has been suggested that you may inherit the genetic predisposition but certain environmental factors are also required to induce the genetic expression of the disease.

The largest series of people of African descent with Dupuytren's disease that has ever been reported was from the American Department of Veteran Affairs of

412 patients.<sup>18</sup> In scrutinising this article, a lot of potential conflicting factors about the purity of the ethnicity is found; in addition, the report was not a clinical report but a data base search. In the 2000 paper describing the database search at the US Department of Veteran Affairs, 412 black patients were identified who presented to various centres with Dupuytren's contractures. Of those, full details of only 136 were obtained. From the search, all these patients were self-declared as black, and considering American history and society, it might be problematic to assess the bona fides and legitimacy of these claims. In their own admission, they also declare that there were no standardised criteria to make the diagnosis and all the doctors that made the diagnosis were not hand surgeons. This is contrary to all the other reports in the literature where the diagnosis was clinical and made by hand surgery units. In the light of these remarks, we believe that these 412 cases cannot accurately be relied upon as being people of pure African descent with Dupuytren's disease. If these 412 patients are not considered, the final reliable number of reported cases of Dupuytren's disease in Africans in the literature is 50 to date. If we include ours, the total is then only 98.

## Conclusion

Considering the literature, it is evident that the disease is extremely rare in people of African descent though the presentation and associations do not differ much among different population groups. The literature also suggests that the true incidence and prevalence of Dupuytren's disease in Africans cannot be accurately determined. From our own records, the number of new cases is steadily increasing in recent years. The limitations of this study are that our hospital is the largest in the southern hemisphere and a major referral centre in Southern Africa hence not a true reflection of the population. Secondly, theatre records give only the name, age, sex, diagnosis and its laterality and surgery done. There are no details on racial descent, disease presentation or risk factors, and often some details are missing or not correctly entered. Finally, purity of race can only be determined genetically and there is a need for population studies that sample the general population to identify subtle disease not just those with severe disease that present to hospital.

## Compliance with ethics guidelines

The authors declare that this is an original work undertaken by them and there are no potential conflicts of interest with respect to the research, authorship and declaration of conflicting interests. The authors received no financial support for the research, authorship and publication of this article.

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