

Clinical Presentation and Management Outcome of Dupuytren's Disease

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Author's Contribution

¹Substantial contributions to the conception or design of the work; or the acquisition, ²Drafting the work or revising it critically for important intellectual content, analysis, or interpretation of data for the work, ³Final approval of the study to be published,

Conflict of interest: None

Funding source: None

Article received: 12-10-2025

Article accepted: 11-04-2025

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ABSTRACT

Objective: To document the clinical epidemiologic profile of patients presenting with Dupuytren's disease and determine the outcome of various surgical treatments employed for their correction.

Methodology: This descriptive case series study was carried out over a period of seven years. All patients who presented with Dupuytren's disease were included. The exclusion criteria were patients who did not receive any surgical intervention. Convenience sampling technique was employed.

Results: There were a total of 41 patients. There were 33(80.48%) males whereas 8(19.51%) females. Their age ranged between 31-71 years with a mean age of 51±17.11 years. Ring fingers (n=27; 52.94%) were the most commonly affected fingers. Majority (n=28; 68.29%) had no known associated co-morbidities whereas 13 patients (31.70%) had co-morbidities. The surgical procedures undertaken included dermatofasciectomy followed by full thickness skin grafting (n=13; 31.70%), percutaneous needle fasciotomy (n=12; 29.26%); open limited fasciectomy (n=9; 21.95%), and open fasciotomy (n=7; 17.07%). The share of postoperative complications included deep scar pain 07(17.07%); partial skin tearing 3(7.31%); partial skin graft loss 2(4.87%); and wound infection 2(4.87%). Recurrence at two years follow up was 3(7.31%).

Conclusions: Majority of the patients with Dupuytren's disease were over the age of 50-years. Individualized approach towards choosing the required surgical intervention not only helped to ensure adequate correction of the finger deformities but was also associated with low overall recurrence.

Keywords: Dupuytren Contracture, Dupuytren Contracture Physiopathology, Surgical Procedures.

Introduction

Dupuytren's disease is fibroproliferative disorder that typically follows a progressive and relentless course. Palmar aponeurosis of the hands is particularly affected by the disease process. The disease is heralded by the formation of nodules in the palmar fascia. This then progresses on to the formation of cords and contractures that result in visible deformities and inability to completely extend the affected fingers.

The disease follows a relentless but variable course. A small percentage of the patients present with Dupuytren's diathesis. This subset of patients presents severe disease, and clinical manifestations typically start at an early age. They may have an associated involvement of the plantar fascia and other body areas as well. Generally, the Dupuytren's disease carries

significant repercussions for the patient as the normal hand function is affected and the quality of life is seriously jeopardized.¹⁻³ Thorough understanding of the morbid anatomy of the palmar fascia and the associated neurovascular structures of the fingers is imperative for ensuring safe surgery for addressing the contractures that result from the progressive Dupuytren's disease.⁴⁻⁷ Both genetic and environmental factors have been incriminated for the causation of Dupuytren's disease. The leading among these factors includes diabetes mellitus, smoking, alcohol intake, liver disease, use of antiepileptics and repeated hand trauma.¹⁻³ Familial trend of the disease has also been reported in some populations of the globe.⁸⁻¹¹

The surgical treatment of the Dupuytren's

disease is tailored according to the extent of the disease and the degree of affliction of the fingers. The goals of management are to ensure adequate release of the contractures and to allow unrestricted optimal extension of the fingers.⁸⁻¹⁰

This study was undertaken to document the clinical and epidemiologic profile of patients presenting with Dupuytren's disease and to document the various forms of treatment options employed in their management.

Methodology

This descriptive case series study was carried out by three plastic surgeons belonging to three centers over a period of seven years. These centers included the National Institute of Rehabilitation Medicine (NIRM), Islamabad, Burns and Plastic surgery Centre, Peshawar and Rawalians Burn and Reconstructive surgery Department, Holy Family Hospital, Rawalpindi. Written informed consent was taken from the patients for inclusion in the study. The study followed the principles of the Helsinki's declaration of 1975, as revised in 2013. Anonymity of the included patients was ensured. Convenience sampling technique was employed. All patients who presented with Dupuytren's disease were included. The exclusion criteria were patients who did not receive any surgical management.

The patients underwent initial evaluation with complete history and physical examination. Baseline investigations were performed to assess the general health and fitness for local anesthesia. The demographic profile of the patients; data regarding whether one or both hands were affected by the contractures and deformities; duration of the disease; corrective surgical procedures undertaken, immediate postoperative complications encountered and recurrence at 2-years follow up were all recorded.

The patients underwent the surgeries under local anesthesia. The extent of surgery was tailored according to the degree of affliction of the hand with Dupuytren's disease. The goals of the surgical management included ensuring adequate release of the contractures and to ensure unrestricted extension of the fingers.

Adequate contracture release was performed

by excising the diseased tissue, and the wounds were either closed using Z-plasty technique or resurfaced with full thickness skin grafts (FTSGs). Padded absorbent dressings were applied and the first dressings were changed after 7-days. The FTSGs were harvested from the retro-auricular regions.

The primary outcome measures included correction of the deformities and any postoperative complications encountered over three months. The secondary outcome measure was recurrence at two years follow up.

Figures 1 through 6 show some illustrative cases of the patients included in the study.

The data were analysed through Statistical package for social sciences (SPSS) version 22. Various descriptive statistics were used to calculate frequencies, percentages, means and standard deviation. The numerical data such as age of the patient was expressed as Mean $\pm$ Standard deviation whereas the categorical data such as the fingers affected were expressed as frequency and percentages.

Results

There was a total of 41 patients in the study. There were 33(80.48%) males whereas 8(19.51%) females. Their age ranged between 31-71 years with a mean age of 51 ± 17.11 years.

The duration of disease was 3-11 years. The mean duration of disease of 4 ± 2.56 years. The presentation of disease for surgical intervention was unilateral among 33(80.48%) patients whereas bilateral among 8(19.51%) patients.

Frequency wise, ring fingers (n=27; 52.94%) were the most commonly affected fingers. It was followed by little fingers (n=17; 33.33%), middle fingers (n=5; 9.80%) and index fingers (n=2; 2.92%).

Majority (n=28; 68.29%) had no known associated co-morbidities whereas 13 patients (31.70%) had co-morbidities. The associated co-morbidities observed included diabetes mellitus 7(17.07%); hypertension 2(4.87%); smoking 2(4.87%); use of antiepileptic agents 1(2.43%); and advanced liver disease 1(2.43%).

The surgical procedures undertaken among the patients included dermatofasciectomy followed by full thickness skin grafting (n=13; 31.70%), percutaneous needle fasciotomy (n=12; 29.26%); open limited fasciectomy (n=9; 21.95%), and open fasciotomy (n=7; 17.07%).

Table 1 presents a brief summary of the clinical presentation and outcome observed among the included patients.

Table I: Summary of the clinical presentation and outcome data among the included patients. (n=41)	
Clinical Features	N (%)
Gender:	
Male	33(80.48%)
Female	8(19.51%)
Side affected (Hand):	
Right side	21(63.63%)
Left side	12 (36.36%)
Bilateral	8(19.51%)
Fingers affected:	
Ring finger	27(52.94%)
Little finger	17 (33.33%)
Middle finger	5(9.80%)
Index finger	2(3.92%)
Associated conditions:	
Diabetes mellitus	7(17.07%)
Hypertension	2(4.87%)
Smoking	2(4.87%)
Use of antiepileptic agents	1(2.43%)
Advanced liver disease	1(2.43%)
Age of the patients: 31-71 years	Mean 51±17.11 years.
Duration of the disease: 3-11 years	Mean 4±2.56 years.
Surgical procedures undertaken:	
Dermatofasciectomy followed by full thickness skin grafting	(n=13;31.70%)
Percutaneous needle fasciotomy	(n=12;29.26%)
Open limited fasciectomy	(n=9;21.95%)
Open fasciotomy	(n=7;17.07%)
Postoperative complications observed:	
Deep scar pain	07(17.07%)
Partial skin tearing	3(7.31%)
Partial skin graft loss	2(4.87%)
Wound infection	2(4.87%)
Recurrence at two years	3(7.31%)

Figure 2: (A-F). These clinical photographs show the left hand of male aged 71-years who had Dupuytren’s disease for nine years. He had developed contracture of the little finger. He underwent surgical excision of the cord as well as limited fasciectomy of the adjacent involved fascia. The wound was closed with Z-plasties and partially directly. The patient had good recovery at 3-months follow up.



Figure 1(A-F): These clinical photographs show the common presentations of patients with Dupuytren’s disease. Figure A: A male patient aged 51 with contractures of both little fingers. The metacarpophalangeal joints (MCPJs) were involved whereas the proximal interphalangeal joints (PIPJs) were spared. Figure B: This photograph shows the hands of male aged 42-years. He had developed Dupuytren’s disease over the last 5-years. There was disease involvement of both palms; however, he developed disabling contracture of the left ring finger. Figure C: This photograph shows the left hand of female patient aged 52-years. She had the disease over the last nine years. She had severe disabling contractures of the little and ring fingers of her left hand. The MCPJs as well as the PIPJs were involved. The other hand was also involved but had less severe disease requiring no surgery. Figure D: This photograph shows the hands of male aged 45-yeears who had developed Dupuytren’s disease over the last seven years. He had contractures of the little finger on the right hand whereas contracture of the ring finger on the left side. Figure E: This photograph shows the right hand of male 31-years who had developed Dupuytren’s disease over the last three years. He had developed severe contracture of the ring finger. He also had knuckle pads on his skin overlying the PIPJs. The other hand was also involved by the disease but there was no contracture formation. Figure F: This photograph shows the left hand of male aged 63-years who had developed Dupuytren’s disease over the last eleven years. He presented with formation of cords and contracture affecting the ring finger.

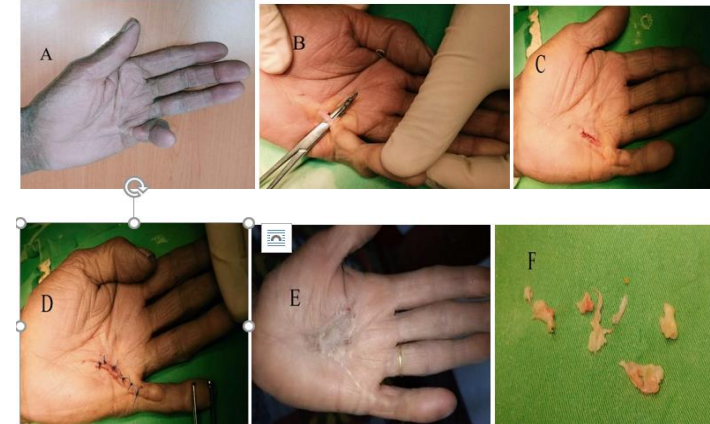




Figure 3: (A-C): These clinical photographs show the left hand of female aged 52-years. She had developed Dupuytren's contracture of the ring finger over the last four years. She underwent excision of the cord with limited fasciectomy. She had good recovery at three months follow up

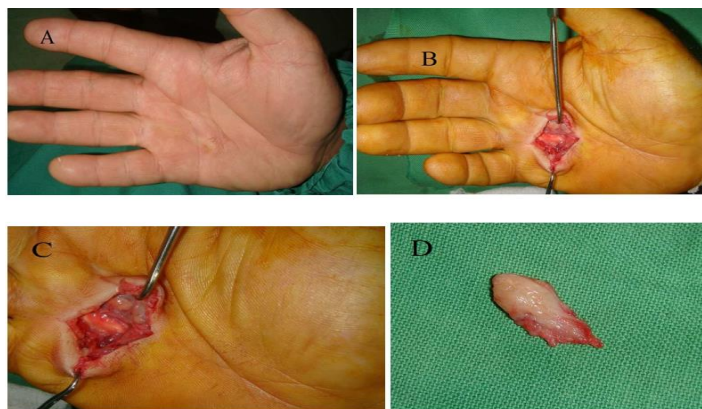


Figure 4: (A-D): These clinical photographs show the right hand of male aged 49-years. He was a laborer by profession. He had developed Dupuytren's disease over the last seven years. He underwent limited fasciectomy and had good recover at three months follow up

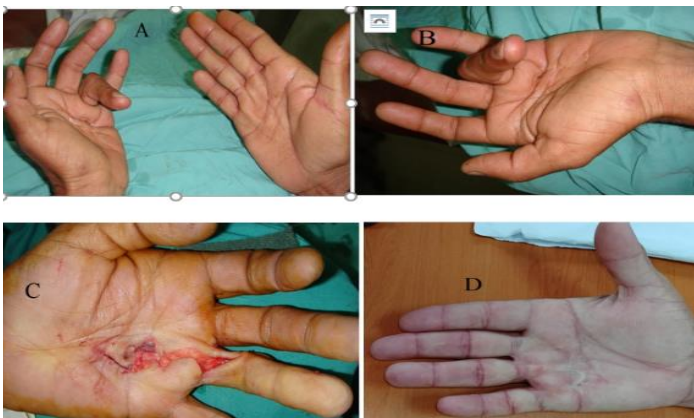


Figure 5: (A-D): These clinical photographs show the hands of male aged 49-years. He had developed Dupuytren's disease over the last six years. The disabling contracture of the left ring finger was corrected with limited fasciectomy performed through Z-plasty skin incisions. He had good recovery at three months follow up.



Figure 6: (A-F): These clinical photographs show the right hand of male aged 51-years. He had developed Dupuytren's disease over the last 10-years. He underwent dermatofasciectomy with Z-plasty skin incision. The resultant wound was resurfaced with Full thickness skin graft which was harvested from the retro-auricular area. He had good recovery at three months follow up.

The share of complications included deep scar pain 07(17.07%); partial skin tearing 3(7.31%); partial skin graft loss 2(4.87%); and wound infection 2(4.87%). Recurrence at two years follow up was 3(7.31%). There was no mortality in the series.

Discussion

Pathophysiologically, the Dupuytren's disease typically goes through three stages: The first stage is called the proliferative stage which is characterized by hypercellularity, presence of immature fibroblasts, formation of large myofibroblasts and nodules. The second phase is called involutonal stage. This is characterized by formation of a network of dense myofibroblast and increasing ratio of type III to type I collagen. The third stage is known as the residual stage. At this stage, collagen rich dense tissue is left and the myofibroblast gradually disappear.¹⁻³

The reported incidence of Dupuytren's disease varies widely across different regions of the globe. It varies from 0.6%-31.6% in the general population.^{1,11,12} Lee KH et al¹³ from Korea reported a mean annual incidence of 1.09 (1.11)/100,000 population/year. Yeh CC from Taiwan¹⁴ reported the incidence to range between 0.39-0.63/105 for men whereas 0.14-0.44/105 for women. There is also considerable variation in the reported rates of

surgical interventions instituted for treating the Dupuytren's disease.

In our series, majority of our patients were males. The published literature has also reported similar findings. Most of the studies have reported men to be 4-5 times more affected than women with the disease ^{1-3,13-15} In our series, the mean age of the patients was 51 years. Most of the reported cases are in the age range of 5th to 7th decades of life. Lee KH et al ¹³ reported the mean age of patients to be 53.2 years. Yeh CC from Taiwan ¹⁴ observed that the mean age of patients with Dupuytren's disease was significantly higher in men (60.7 ± 18.4 years) than in women (53.7 ± 15.5). They observed that the incidence was lowest among individuals below the age of 40 years. They reported the highest incidence among men aged 70-79-years whereas women aged 50-59-years.

In our study, ring finger was most commonly affected followed by the little finger. The published literature has reported that the order of descending frequency of affliction is ring, small, middle and index fingers. ⁷

In our series, diabetes mellitus (DM) was observed among 17% patients. Several published studies have reported an association between DM and Dupuytren's disease ^{1,16,17}, however; other studies have not confirmed any such association. ^{1,18} Sporadic cases of Dupuytren's disease are more common whereas cases of autosomal dominant with variable penetrance may also occur sometimes.

The published studies have reported a variety of conditions that may be associated with Dupuytren's disease. For instance, type I DM, type II DM, advancing age beyond 50, gender, smoking and alcohol consumption, family history and geographic background of the affected individuals.¹⁸⁻²⁰ Lee KH et al ¹³ from Korea reported the Dupuytren's disease to be associated with hypertension among (n=5,076;30.5%) patients, DM among (n=4,437;26.7%) patients, hyperlipidemia among (n=3,396;20.4%) patients, ischemic heart disease among (n=1,309;7.9%) patients, cerebrovascular disease among (n=769;4.6%) patients, and obstructive lung disease among (n=438;2.6%) patients. Among patients with Dupuytren's Diathesis, there may be associated involvement of ectopic sites. For instance, the dartos fascia of penis (i.e., Peyronie's disease),

knuckle pads (i.e., Garrod disease) and plantar fascia (i.e., Ledderhose disease).¹⁷

The available surgical treatments range from minimally invasive procedures such as the percutaneous needle fasciotomy (also known as needle aponeurotomy) and operative open fasciectomy to more invasive procedures such as limited fasciectomy, and dermatofasciectomy with full thickness skin grafting (FTSG). The conservative and non-surgical treatment options include observation and the use of collagenase injections.⁴

In our study, as the majority of patients presented with advanced disease and hence the most frequently employed procedure was dermatofasciectomy and full thickness skin grafting. Young patients with a positive family history of the disease, patients with severe contractures and those with associated co-morbidities such as DM, primary dermo fasciectomy and full thickness skin grafting are instituted as the first line procedures because the percutaneous needle fasciotomy and limited fasciectomy are associated with early recurrence among these high-risk patients.¹⁷⁻²¹

Contrary to our experience, Toppi JT et al ²¹ had majority of early-stage patients and observed that operative open fasciectomy and percutaneous needle fasciotomy procedures provided adequate deformity correction for uncomplicated primary Dupuytren's disease.

The easy execution and reduced associated side effects of percutaneous needle fasciotomy constitutes the first-line management for uncomplicated primary Dupuytren's disease. With contractures, minimally invasive treatments lead to good immediate outcomes as indicated by rapid recovery, early return to everyday life and less cost of the procedures. The minimally invasive interventions are however met with higher recurrence rates over long term periods.^{4,21,22}

Limited fasciectomy is indicated particularly if there are either mild proximal interphalangeal joints (PIPJs) flexion contractures or metacarpophalangeal joints (MCPJs) flexion contractures of $\geq 30^\circ$. Where primary contractures are more severe (i.e., total extension deficit is over 90° and isolated PIPJ contractures are over 60°), minimally invasive treatments are less effective.^{4, 21-23}

In the current study, we met with two cases of partial skin graft loss. Ansari SA et al²⁴ also reported two cases (n=49;4%) of graft failure in their series. In the published literature there is limited evidence regarding graft failure rates in dermo fasciectomy with full thickness skin grafting, some authorities have reported this to be ranging between 1%-8.6%.^{24,25} In the published literature, the reported recurrence rates in Dupuytren's disease vary from 2%-86%. The recurrence depends on a variety of factors. For instance, the definition of recurrence adopted, the period of follow up, and the

procedure instituted. The employment of FTSGs is reported to be associated with lowest recurrence rates.²⁶⁻²⁹

In our study we did not employ the medically managed cases who received Collagenase injections. A plethora of published literature exists regarding the use of Collagenase injections for managing the disease.³⁰⁻³²

Strengths and Limitations of the Study:

The current research has certain strengths as well

as presents certain limitations. Firstly, the strength included the fact that the study is the first of its kind to document the clinical and epidemiological presentation of Dupuytren's disease in the local Pakistani population at three different cities. Secondly,

it documented the various surgical procedures which are undertaken for addressing Dupuytren's disease. The study has certain limitations as follows: The main limitation is that it was an observational study. The results of the study may be interpreted keeping in mind the aforementioned limitations. Future well-designed multicenter studies are recommended to overcome these limitations.

Conclusion

Majority of the patients with Dupuytren's disease were over the age of 50-years. Individualized approach towards choosing the required surgical intervention not only helped to ensure adequate correction of the finger deformities but was also associated with low overall recurrence

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