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ORIGINAL STUDY

Pain in dupuytren disease

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To investigate pain and the effect of treatment in patients with Dupuytren disease, we analyzed the literature on pre- and post-interventional pain and complex regional pain syndrome. The pre-interventional pain intensity score of primary Dupuytren ranged from 0.3/10 to 2.0/10. One year after surgery or needle fasciotomy, no significant change of pain could be found. Collagenase therapy significantly reduced the mean pain intensity score from 1.3/10 [SD 2.2] to 0.5/10 [1.5] (p<0.01) after one year. The prevalence of complex regional pain syndrome after fasciectomy ranged from 0% to 12.8%, after needle fasciotomy from 0% to 6.3%, and after collagenase therapy from 0% to 3.0%. We conclude that for most Dupuytren patients, pain is not an issue. A minority seems to suffer pain and collagenase treatment appears to reduce this complaint significantly. However, complex regional pain syndrome is a known complication of Dupuytren treatment, with a low incidence after minimal invasive treatment.

Keywords : Dupuytren ; pain ; CRPS ; collagenase ; surgery ; needle aponeurotomy ; amputation ; splinting.

INTRODUCTION

Dupuytren disease (DD) is very common in Northern Europe with a 21% prevalence at the age of 65 *(50)*. Nowadays, no curative treatment is known. The indication for a symptomatic therapy is largely based on the degree of flexion contracture and functional impairment. As a consequence, the literature focuses on goniometric outcomes. Long-term pain is less studied.

If DD causes pre-interventional pain, patients hope that this pain may improve with a symptomatic treatment for Dupuytren contractures. However, chronic pain and and complex regional pain syndrome (CRPS) can be a complication of Dupuytren treatment itself.

The aim of this literature review was to analyze reports on long-term pain and complex regional pain syndrome before and after treatment of DD. This information is important for patients who consider treatment for Dupuytren contracture.

MATERIALS AND METHODS

A search was performed on Pubmed, Web of Science, Cochrane library and Embase starting from the inception of the databases until November 2018 with the search terms Dupuytren contracture, pain, CRPS and synonyms, outcome, one year results and all the different treatment options. We

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considered articles written in English, German, Dutch or French, and excluded other languages, case reports, retracted articles and studies with other factors, e.g. simultaneous carpal tunnel syndrome and Dupuytren's disease. We included articles that reported 1) a pain intensity score or prevalence of pain of patients with DD before treatment and/or 2) prevalence of pain/CRPS or pain intensity scores of patients with DD at least 3 weeks after treatment. We converted the pain intensity scores to a scale from zero to ten.

RESULTS

We included 89 articles. The pre-interventional pain intensity score ranged from 0/10 to 3.3/10 *(24,47,48,59,60,69,76,77)* for all DD patients, for primary DD patients these score ranged from 0.3/10 to 2.0/10 *(24,47)*. Hurst L.C. et al. *(44)* observed pain as the first sign of DD in 2.3% of the cases. Spies C.K. et al. *(76,77)* reported that 13% of the patients with primary and recurrent disease had pain, pre-interventionally, and 44% of the patients with recurrent DD.

In table I, the reported pain intensity scores after surgery and needle fasciotomy are listed. Overall, no significant differences were found between the groups. The pain intensity scores one year after surgery ranged from 0/10 to 3.3/10.

One year after collagenase (CCH) therapy, Odinsson A. et al. *(59)* found a significant decrease of the mean pain intensity score from 1.3/10 [SD 22] pre-interventionally to 0.5/10 [15] ($p<0.01$). Scherman A. et al. *(71)* reported a mean pain intensity score of 0.4/10 and 5.3% of the patients complained of pain one year after CCH. Other authors *(5,15,22,23)* reported that their patients were pain free after four weeks to one year. Bear B.J. et al. *(11)* observed 1 patient with persistent pain that resolved within 6 months. Vollbach F.H. et al. *(86)*, mentioned that 71% of the patients experienced temporary pain when grasping objects at 1 month. Overall, little chronic pain was reported after CCH treatment.

Amputation and arthrodesis of fourth or fifth digit are sometimes chosen as salvage procedure in severe and/or recurrent Dupuytren disease. Degreef I *(25)* et al. reported a median pain intensity score of 1.75/10, 1 to 6 years after amputation and 50% of the patients experienced pain. Jensen C.M. et al. *(45)* observed neuroma in 22% and phantom pain in 17% 4 years after finger amputation. Honecker S. et al. *(42)* presented shortening arthrodesis by resection of the proximal interphalangeal joint as alternative

Table I. — Surgery and needle fasciotomy

	Study type	n (patients)	Treatment	Pre-interventional pain intensity score : Median/mean (0-10) [SD]	Post-interventional pain intensity score : Median/mean (0-10) [SD]	Time after treatment
Ganeval A. et al. *(36)*	Prospective cohort	20	Digital needle fasciotomy	-/-	1/1.8	10 m (range 3-24 m)
Spies C.K. et al. *(77)*	Retrospective cohort	15	Percutaneous needle fasciotomy	2/15 (13.3%)	-/-	40 m
Scherman P. et al. *(71)*	RCT	45	Percutaneous needle fasciotomy + sometimes night splint	-/-	0/0.1	12 m
Kemler M.A. *(48)*	Pilot study (RCT)	26	Limited fasciectomy + primary closure	-/0 [0]	-/0.7 [1.3]	12 m
		28	Limited fasciectomy + primary closure + splinting	-/0 [0]	-/1.3 [2.0]	
Spies C.K. et al. *(76)*	Retrospective cohort	18	Limited fasciectomy	0/1.94	0 /1	94 m (range: 70-114 m)
Pearl R.A. et al. *(60)*	Prospective cohort study	51	Not specified	3.3 /-	3.3 /-	8 m
Rodrigues J. et al. *(69)*	Cross-sectional study	750	Fasciectomy or dermofasciectomy or percutaneous needle fasciotomy	2.5 /-	0 /-	1 y
					0 /-	5 y
				Note : pain in arm, shoulder and hand		

Table II. — Post-interventional pain

	% of patients with pain	post-interventional time
Limited fasciectomy + primary closure *(1,53,54,76,86,91)*	0-27%	4 m-20 y
Limited fasciectomy + open palm technique *(53)*	0%	
Fasciectomy + external fixation *(12)*	7.7%	
Amputation *(25)*	50%	9 m-81 m
Arthrodesis *(42,58)*	25%-83.2%	
Combinations of surgical techniques *(41)*	4%	>3 w
Percutaneous needle fasciotomy *(36,71,77)*	0-55%	3 m-24 m
CCH *(5,11,15,22,23,71)*	0-5%	2 w-12 m
Topical injections of hydrocortisone acetate *(89)*	9%	2 m-24 m

Table III. — CRPS

Treatment	CRPS (%)
Fasciectomy	
Limited fasciectomy + open palm technique *(33,34,37,53,70,73,74)*	0%-9.3%
Limited fasciectomy + primary skin closure *(16,21,37,47,53,55,65,66,76,80,82,91,92)*	0%-9.2%
Complete fasciectomy + primary skin closure *(85)*	1.8%
Segmental aponeurectomy *(20)*	0%
Fasciectomy + external fixation *(18)*	38.5%
Fasciectomy (type not further specified or combination) + primary skin closure *(4,14,19,26,28,46,49,83,87,88)*	0%-12.8%
Dermatofasciectomy/fasciectomy + skin graft/flap *(2,74,83,87)*	0%-5%
Combinations of surgical techniques *(29,30,31,39,40,67,81)*	0%-12.8%
Fasciotomy	
Percutaneous needle fasciotomy *(9,13,35,36,56,64,65,66,75,79)*	0%-6.3%
Percutaneous aponeurotomy and lipofilling *(43,47)*	4.4%-5%
Subcutaneous fasciotomy *(68)*	0%
CCH *(6,7,8,10,23,27,51,57,59,61,62,63,75,79,84)*	0%-3.0%
Sex	
women + all types of surgery *(3,32,33,34,72,78)*	0.9%-50%
men + all types of surgery *(3,32,33,90)*	1.5%-12.5%
Primary versus recurrent	
primary + all types of treatment *(26,38,46,47,65,74,83)*	0-6.3%
recurrent + all types of treatment *(76)*	0%

to amputation of the fifth ray. The median pre-operative pain intensity score dropped from 5/10 and 75% of the patients with pain to a median pain intensity score of zero and 25% of the patients with pain after 9-81 months. Novoa-Parra C.D. et al. *(58)* presented PIP joint arthrodesis with interlocking screw in severe DD recurrence. A hundred percent of the patients reported pre-interventional pain, but the median pain intensity score did not improve significantly, from 4/10 pre-intervention to 1.5/10

one year after the arthrodesis, pain persisted in 83.3%. In general, arthrodesis lowered the pain but the scores remained high.

Zachariae L. et al. *(89)*, reported in 1955 that the percentage of patients who experienced pain in DD dropped from 36% to 9% 2 months to 2 years after injecting hydrocortisone acetate.

von Campe A. et al. *(17)* observed patients that underwent surgery for persistent pain due to DD. The mean pain intensity score dropped from 4.9/10 to 0/10 25 months after fasciectomy, however, 2 of the 10 patients developed CRPS.

Table II summarizes the percentages of patients with post-interventional pain. The percentages were the lowest after collagenase, the highest after amputation and arthrodesis.

Table III summarizes the percentages of patients with CRPS. The studies that compared the prevalence of CRPS between the sexes *(3,32,33,90)* all reported a higher percentage of CRPS in women, with the exception of Anwar M.U. et al. *(3)*, who reported a percentage of CRPS of 2% for men, versus 0.8% for women. The percentage of CRPS in women was significantly higher in the study by Sennwald G.R. et al. *(72)* ($p < 0.02$).

Zemel N.P. et al. *(90)* reported on long term outcome of Dupuytren surgery in women in 1987. He performed a simultaneous carpal tunnel release in 12 women without the presence of carpal tunnel syndrome but that received an extensive fasciectomy. CRPS developed in 58% (7 of 12) of these hands, versus 8% (5 of 61 hands) of the hands without carpal tunnel release. This was statistically significant. Furthermore, 46% of the hands that underwent a more extensive fasciectomy, developed CRPS, versus 10% of the hands who received a limited fasciectomy. This was also significant.

DISCUSSION

Pre-interventional pain intensity scores of primary Dupuytren ranged from 0.3 to 2.0/10. One year after surgery or needle fasciotomy, no significant change in pain reporting could be found. However, collagenase therapy significantly reduced the pain after 1 year. The prevalence of CRPS after fasciectomy ranged from 0% to 12.8%, after needle fasciotomy from 0% to 6.3%, and after collagenase therapy from 0% to 3.0%.

This review has several weaknesses, i.e. little statistical evidence in the studies, the diversity of the patient groups, and the fact that, throughout the years, no clear definitions of CRPS and lots of synonyms were used. Moreover, we only focused on pain and not on other outcomes. This review's strength is however its specific focus on pain in DD, which is rarely highlighted in treatment outcome reports.

The treatment of primary pain in DD is challenging. The role of surgery is unclear. Recently, von Campe A. et al. *(17)* performed fasciectomy for painful DD and suggested that the indication for surgery in DD should even be extended to the presence of painful nodules for over 1 year. They suggested a histological inspection on a large scale to investigate whether histological changes are linked to pain in DD. However, surgery for DD may also induce pain.

Another promising option for persistent pain in DD seemed to be CCH therapy. As stated in the results, Odinsson A. et al. *(59)* reported significantly less pain after 1 year and Costas B. et al. *(24)* found that CCH therapy improved nodular pain. However, it is possible that less severe cases of Dupuytren were treated by CCH therapy. This might bias our results compared to the surgery groups.

Furthermore, Jensen C.M. et al. *(45)* reported amputation as a treatment of pain due to nerve lesions of previous operations. In general, amputation of the fourth and especially the fifth finger is frequently complicated with neuromata after more proximal amputations. Therefore, amputation should be limited to a selected group of patients. Honecker S. et al. *(42)* presented arthrodesis as an alternative to amputation, with the advantage that nerves are preserved, and consequently, less neuroma's and pain are present. However, in this review, we did not find a difference between the prevalence of pain after amputation versus after arthrodesis.

A common cause of pain after Dupuytren treatment is CRPS with an incidence of 0 to 12.8% after fasciectomy, but it is difficult to predict. Possibly preoperative pain could indicate a higher susceptibility. We did not find any reports that stated

that patients who suffered from pre-interventional pain had a higher chance of developing CRPS. However, von Campe A. et al. *(17)* reported an incidence of 20% in patients undergoing fasciectomy for pain, which is higher than the overall incidence after surgery. The role of simultaneous carpal tunnel decompression in unclear since Zemel et al. *(90)* found significantly more instances of CRPS and Lilly S.I. et al. *(52)* reported that only 3% of the simultaneous group developed CRPS.

CONCLUSION

DD is a highly prevalent disease, but pain is rather rare and mostly mild if left untreated. The effect of surgery on pain is unclear, although CCH was reported to significantly decrease pain. Preoperative pain may indicate a higher risk for CRPS after treatment, which is more likely if in more invasive surgery. We suggest that future studies on treatment outcome in DD include pain in their results.

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