Review

Current concepts about treatment options of plantar fibromatosis: A systematic review of the literature

Mario Fuiano\textsuperscript{a}, Massimiliano Mosca\textsuperscript{a}, Silvio Caravelli\textsuperscript{a,}\textsuperscript{x}, Simone Massimi\textsuperscript{a}, Maria Grazia Benedetti\textsuperscript{a}, Francesco Di Caprio\textsuperscript{b}, Salvatore Mosca\textsuperscript{c}, Stefano Zaffagnini\textsuperscript{a}

\textsuperscript{a} Rizzoli Orthopaedic Institute, Bologna, Italy
\textsuperscript{b} Ospedale Ceccarini, Riccione, Italy
\textsuperscript{c} Università Vita-Salute S. Raffaele, Milano, Italy

\textbf{A R T I C L E   I N F O}

\textbf{Article history:}
Received 7 January 2018
Received in revised form 19 February 2018
Accepted 1 June 2018

\textbf{Keywords:}
Foot
Ledderhose
Plantar fibromatosis
Treatment

\textbf{A B S T R A C T}

\textit{Background:} Plantar fibromatosis, or Ledderhose disease, is a benign and hyperproliferative disease of the plantar aponeurosis. There have been described different therapeutic options regarding plantar fibromatosis, both conservative and surgical. The aim of this review is to systematically analyze conservative and operative treatments of plantar fibromatosis described in literature, evaluating which procedure shows the highest success rate and best functional outcome.

\textit{Methods:} A systematic review of PubMed, Google Scholar and Cochrane reviews computerized database was performed focusing on the different types of treatments for plantar fibromatosis. Research was performed using the keywords “plantar”, “fibromatosis”, “Ledderhose”, “Dupuytren”, “foot” in order to identify all papers regarding the treatment of plantar fibromatosis. In addition, the research was extended to the reference list of the relevant articles. A total of 25 citations were obtained from the research and included.

\textit{Results:} Considering all the studies, 233 patients were included in this systematic review. 5 studies reported conservative treatment of plantar fibromatosis, with a total of 35 patients included. Operative outcomes are reported for 178 patients (92 male, 86 female), with 196 feet treated.

\textit{Conclusions:} Valid conservative methods are presented in literature, with debated results. Some operative options show high recurrence rate; wide excision is recommended in selected cases. Further clinical trials with well-defined and standardized outcome measurements should be necessary in future to better evaluate success rate and complications of the various procedures.

© 2018 European Foot and Ankle Society. Published by Elsevier Ltd. All rights reserved.

\textbf{Contents}

1. Introduction .................................................................................................................................................. 559
2. Materials and methods ............................................................................................................................... 560
   2.1. Studies selection ..................................................................................................................................... 560
   2.2. Evaluation criteria ................................................................................................................................. 560
3. Results ......................................................................................................................................................... 560
4. Discussion .................................................................................................................................................... 562
5. Conclusion ................................................................................................................................................... 564
   Authorship .................................................................................................................................................. 564
   Conflicts of interest .................................................................................................................................. 564
   References ................................................................................................................................................... 564

1. Introduction

Plantar fibromatosis, or Ledderhose disease, is a rare benign and hyperproliferative disease of the plantar aponeurosis, of unknown etiology and included among the extra-abdominal desmoid tumors\textsuperscript{[1]}.  

\url{https://doi.org/10.1016/j.fas.2018.06.001}
1268-7731/© 2018 European Foot and Ankle Society. Published by Elsevier Ltd. All rights reserved.
It was described for the first time by the German physician Georg Ledderhose in 1897 [2,3]. It is composed of differentiated fibroblasts and clinically characterized by nodules, mainly in the medial portion of the plantar fascia [4].

It can occur at all ages, but especially in fourth and fifth decades, with prevalence in male and bilateral involvement in 20–50% of the cases. Often it is associated with palmar fibromatosis (Dupuytren’s disease) and La Peyronie’s disease [5].

Etiology is debated, but plantar fibromatosis has been described to be most frequent in patients with diabetes, low body weight, epilepsy and alcoholism. It could also be familiar [6].

Three phases in the progression of the disease have been distinguished: proliferative, involutorial and residual [1]. The first stage is characterized by cellular proliferation, the second phase by nodule formation and the third by tissue contraction [7].

Clinically patients present one or more subcutaneous nodules, most frequently in the medial portion of the plantar fascia (Fig. 1) that can be also asymptomatic, but in most cases it causes pain and swelling in the foot. In the late stage can also be observed contractures of the plantar fascia and retractions [8].

Diagnosis of plantar fibromatosis is usually clinical. Ultrasound and MRI (Fig. 2) are required for excluding differential diagnosis and to define dimension and invasion of nearest soft tissues [9].

There have been described different therapeutic options regarding plantar fibromatosis, both conservative and surgical [10].

Most used conservative treatments are rest, ice, orthoses, stretching of the plantar fascia, physical therapies, local injections of steroids [11]. In some cases, conservative treatment do not solve the symptoms and it is necessary to proceed to the operative removal of the lesion.

Plantar fibromatosis often tends to recur after operative removal. A complete excision of the lesion without leaving pathological tissue is necessary to minimize the risk of recurrence [11].

There is no consensus in literature about the width of resection. Besides a wider excision leads more frequently to complications like dehiscence of operative wound, infections, skin necrosis, painful neuroma [12].

The aim of this review is to systematically analyze conservative and operative treatments of plantar fibromatosis described in literature, evaluating which procedure shows the highest success rate and best functional outcome.

2. Materials and methods

2.1. Studies selection

A systematic review of PubMed, Google Scholar and Cochrane reviews computerized database was performed focusing on the different types of treatments for plantar fibromatosis.

Research was performed using the keywords “plantar”, “fibromatosis”, “Ledderhose”, “Dupuytren”, “foot” in order to identify all papers regarding the treatment of plantar fibromatosis.

In addition, the research was extended to the reference list of the relevant articles.

A total of 25 citations were obtained from the research and included.

Two independent researchers performed separately a more accurately review considering articles with the following inclusion criteria: clinical studies regarding the treatment of plantar fibromatosis, articles written in English.

Exclusion criteria were: articles written in other languages, articles not reporting clinical trials, articles regarding fibromatosis disease but without plantar localization, literature reviews.

Finally, 16 studies fulfilled the criteria and were analyzed. Selected studies are summarized in Table 1. All studies included in this review except 5 (3 prospective case series, 2 case reports), were retrospective case series.

There were no randomized controlled trials or prospective controlled studies.

6 different treatment groups were identified: local steroid injections, extracorporeal shockwave, local collagenase of C. histolyticum injection, radiotherapy, surgery, surgery + radiotherapy.

2.2. Evaluation criteria

Evaluation criteria used by the authors to evaluate and describe outcomes are different.

In the studies regarding a conservative treatment, like therapeutical local injection or shockwave therapy, success was evaluated with VAS (visual analogue scale), satisfaction of the patient, consistency of the lesion.

In the studies regarding the operative approach, success was evaluated by the absence of recurrence and satisfaction of the patient. Only 1 study regarding operative approach reported AOFAS score.

10 studies reported general or local complications of the treatment, 6 studies have not reported complications.

3. Results

Considering all the studies, 233 patients were included in this systematic review (Table 2).
Fig. 2. Plantar fibromatosis. MRI shows a single small nodular thickening of the plantar fascia (arrows). (A) T1W and (B) STIR sagittal plane of the foot, low-intermediate signal mass growing from plantar fascia. (C) Post-gadolinium T1W axial image of the midfoot, Ledderhose’s nodules often, although not invariably, enhance with intravenous paramagnetic contrast fluid.

Table 1
Selected studies (RT: radiotherapy).

<table>
<thead>
<tr>
<th>Author</th>
<th>Approach</th>
<th>Date</th>
<th>Type of study</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sammarco et al. [8]</td>
<td>Surgery</td>
<td>2000</td>
<td>Retrospective</td>
</tr>
<tr>
<td>Dürr et al. [17]</td>
<td>Surgery</td>
<td>1999</td>
<td>Retrospective</td>
</tr>
<tr>
<td>Kadir [29]</td>
<td>Surgery</td>
<td>2017</td>
<td>Retrospective</td>
</tr>
<tr>
<td>Wäpner et al. [12]</td>
<td>Surgery</td>
<td>1995</td>
<td>Retrospective</td>
</tr>
<tr>
<td>Alusio et al. [26]</td>
<td>Surgery</td>
<td>1996</td>
<td>Retrospective</td>
</tr>
<tr>
<td>Ushijima et al. [19]</td>
<td>Surgery</td>
<td>1984</td>
<td>Retrospective</td>
</tr>
<tr>
<td>Haedicke et al. [22]</td>
<td>Surgery</td>
<td>1988</td>
<td>Prospective case series</td>
</tr>
<tr>
<td>Fetsch et al. [20]</td>
<td>Surgery</td>
<td>2005</td>
<td>Retrospective</td>
</tr>
<tr>
<td>Rao et al. [23]</td>
<td>Surgery</td>
<td>1988</td>
<td>Prospective case series</td>
</tr>
<tr>
<td>de Bree et al. [18]</td>
<td>Surgery + RT</td>
<td>2004</td>
<td>Retrospective</td>
</tr>
<tr>
<td>van der Veer et al. [21]</td>
<td>Surgery + RT</td>
<td>2008</td>
<td>Retrospective</td>
</tr>
<tr>
<td>Heyd et al. [14]</td>
<td>Radiotherapy</td>
<td>2010</td>
<td>Retrospective</td>
</tr>
<tr>
<td>Grenfell et al. [10]</td>
<td>Radiotherapy</td>
<td>2014</td>
<td>Retrospective</td>
</tr>
<tr>
<td>Knobloch et al. [15]</td>
<td>Extracorporeal shockwave</td>
<td>2012</td>
<td>Prospective case series</td>
</tr>
<tr>
<td>Hammoudeh et al. [16]</td>
<td>Collagenase injection</td>
<td>2014</td>
<td>Case report</td>
</tr>
</tbody>
</table>

Table 2
Population characteristics (NR: not reported).

<table>
<thead>
<tr>
<th>Author</th>
<th>No. patients</th>
<th>No. patients to FU</th>
<th>No. feet</th>
<th>No. feet to FU</th>
<th>M</th>
<th>F</th>
<th>Age (yrs)</th>
<th>Bilateral</th>
<th>Single/multiple</th>
<th>Familiar</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sammarco et al. [30]</td>
<td>18</td>
<td>16</td>
<td>23</td>
<td>21</td>
<td>9</td>
<td>9</td>
<td>49 (11–67)</td>
<td>12</td>
<td>NR</td>
<td>1</td>
</tr>
<tr>
<td>Dürr et al. [7]</td>
<td>11</td>
<td>11</td>
<td>13</td>
<td>13</td>
<td>5</td>
<td>6</td>
<td>33 (9–53)</td>
<td>6</td>
<td>NR</td>
<td>1</td>
</tr>
<tr>
<td>de Bree et al. [5]</td>
<td>9</td>
<td>9</td>
<td>11</td>
<td>11</td>
<td>3</td>
<td>6</td>
<td>26 (8–46)</td>
<td>2</td>
<td>NR</td>
<td>2</td>
</tr>
<tr>
<td>Kadir et al. [17]</td>
<td>18</td>
<td>18</td>
<td>18</td>
<td>18</td>
<td>12</td>
<td>6</td>
<td>41.3 (20–57)</td>
<td>1</td>
<td>15/3</td>
<td>3</td>
</tr>
<tr>
<td>Wäpner et al. [34]</td>
<td>10</td>
<td>10</td>
<td>10</td>
<td>10</td>
<td>4</td>
<td>6</td>
<td>46</td>
<td>2</td>
<td>NR</td>
<td>NR</td>
</tr>
<tr>
<td>Alusio et al. [2]</td>
<td>30</td>
<td>30</td>
<td>33</td>
<td>33</td>
<td>9</td>
<td>21</td>
<td>38 (5–69)</td>
<td>4</td>
<td>23/10</td>
<td>4</td>
</tr>
<tr>
<td>van der Veer et al. [32]</td>
<td>27</td>
<td>27</td>
<td>33</td>
<td>33</td>
<td>21</td>
<td>6</td>
<td>42 (18–72)</td>
<td>13</td>
<td>36/4</td>
<td>9</td>
</tr>
<tr>
<td>Ushijima et al. [31]</td>
<td>12</td>
<td>12</td>
<td>12</td>
<td>12</td>
<td>5</td>
<td>7</td>
<td>40 (20–69)</td>
<td>2</td>
<td>NR</td>
<td>NR</td>
</tr>
<tr>
<td>Haedicke et al. [13]</td>
<td>4</td>
<td>4</td>
<td>4</td>
<td>4</td>
<td>3</td>
<td>3</td>
<td>45.75</td>
<td>2</td>
<td>3/1</td>
<td>0</td>
</tr>
<tr>
<td>Rao et al. [28]</td>
<td>3</td>
<td>3</td>
<td>3</td>
<td>3</td>
<td>2</td>
<td>1</td>
<td>7 (5–8)</td>
<td>0</td>
<td>3/0</td>
<td>1</td>
</tr>
<tr>
<td>Fetsch et al. [9]</td>
<td>56</td>
<td>38</td>
<td>57</td>
<td>38</td>
<td>19</td>
<td>37</td>
<td>8.6 (2–12)</td>
<td>1</td>
<td>NR</td>
<td>11</td>
</tr>
<tr>
<td>Pentland et al. [25]</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td>0</td>
<td>52</td>
<td>0</td>
<td>1/0</td>
<td>0</td>
</tr>
<tr>
<td>Heyd et al. [15]</td>
<td>24</td>
<td>24</td>
<td>33</td>
<td>33</td>
<td>12</td>
<td>12</td>
<td>52 (28–83)</td>
<td>9</td>
<td>NR</td>
<td>NR</td>
</tr>
<tr>
<td>Grenfell et al. [12]</td>
<td>3</td>
<td>3</td>
<td>5</td>
<td>5</td>
<td>3</td>
<td>0</td>
<td>48.7 (44–52)</td>
<td>2</td>
<td>4/1</td>
<td>2</td>
</tr>
<tr>
<td>Knobloch et al. [18]</td>
<td>6</td>
<td>6</td>
<td>6</td>
<td>6</td>
<td>5</td>
<td>1</td>
<td>58 ± 4</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
</tr>
<tr>
<td>Hammoudeh et al. [16]</td>
<td>1</td>
<td>1</td>
<td>2</td>
<td>2</td>
<td>1</td>
<td>0</td>
<td>72</td>
<td>1</td>
<td>2/0</td>
<td>NR</td>
</tr>
</tbody>
</table>

5 studies [10,13–16] reported conservative treatment of plantar fibromatosis, with a total of 35 patients included.

Operative outcomes are reported for 178 patients (92 male, 86 female), with 196 feet treated.

57 patients had bilateral involvement, and 38 patients had also a Dupuytren’s contracture. 34 cases had a familial history of fibromatosis (of the hand, foot or penis).

87 feet (82%) treated were affected by a single nodule, instead in 19 cases (18%) were present multiple nodules; 7 studies [12,14,15,17–20] do not report these findings.

1 study [19] do not report the exact operative technique used and, for 5 studies [19–23] of the surgery group, complications after treatment are not referred.

Due to the low number of patients of each study and the heterogeneity of the data reported, it was not possible to standardize demographic and outcome of the whole population.

For this reason, it was not possible to divide the population in subgroups.

Operative indication, for both primary lesions and recurrences, was intractable pain and not responding to conservative treatments.
Table 3
Surgical recurrences and further treatments (RT: radiotherapy).

<table>
<thead>
<tr>
<th>Author</th>
<th>Approach</th>
<th>Primary cases treated</th>
<th>Recurrences</th>
<th>Recurrence rate%</th>
<th>Recurrences treated</th>
<th>Recurrences</th>
<th>Recurrence rate%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sammarco [8]</td>
<td>Subtotal fasciectomy</td>
<td>18</td>
<td>2</td>
<td>11%</td>
<td>5</td>
<td>0</td>
<td>0%</td>
</tr>
<tr>
<td>Dürr [17]</td>
<td>Local excision</td>
<td>3</td>
<td>3</td>
<td>100%</td>
<td>4</td>
<td>3</td>
<td>75%</td>
</tr>
<tr>
<td>Wide excision</td>
<td>8</td>
<td>6</td>
<td>75%</td>
<td>1</td>
<td>1</td>
<td>100%</td>
<td></td>
</tr>
<tr>
<td>Plantar fasciectomy</td>
<td>2</td>
<td>0</td>
<td>0%</td>
<td>3</td>
<td>2</td>
<td>67%</td>
<td></td>
</tr>
<tr>
<td>de Bree [18]</td>
<td>Marginal excision</td>
<td>6</td>
<td>6</td>
<td>100%</td>
<td>3</td>
<td>3</td>
<td>100%</td>
</tr>
<tr>
<td>Wide local excision</td>
<td>3</td>
<td>2</td>
<td>66.60%</td>
<td>3</td>
<td>2</td>
<td>66%</td>
<td></td>
</tr>
<tr>
<td>Plantar fasciectomy</td>
<td>1</td>
<td>1</td>
<td>100%</td>
<td>1</td>
<td>0</td>
<td>0%</td>
<td></td>
</tr>
<tr>
<td>Below-knee amputation</td>
<td>1</td>
<td>1</td>
<td>100%</td>
<td>1</td>
<td>0</td>
<td>0%</td>
<td></td>
</tr>
<tr>
<td>Surgery + RT</td>
<td>6</td>
<td>1</td>
<td>17%</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Kadir [29]</td>
<td>Partial fasciectomy</td>
<td>16</td>
<td>1</td>
<td>6%</td>
<td>3</td>
<td>0</td>
<td>0%</td>
</tr>
<tr>
<td>Wapner [12]</td>
<td>Wide excision</td>
<td>5</td>
<td>1</td>
<td>20%</td>
<td>3</td>
<td>0</td>
<td>0%</td>
</tr>
<tr>
<td>Staged excision</td>
<td></td>
<td></td>
<td></td>
<td>4</td>
<td>0</td>
<td>0%</td>
<td></td>
</tr>
<tr>
<td>Aluisio [26]</td>
<td>Local excision</td>
<td>10</td>
<td>4</td>
<td>40%</td>
<td>1</td>
<td>1</td>
<td>100%</td>
</tr>
<tr>
<td>Wide excision</td>
<td>3</td>
<td>1</td>
<td>33.30%</td>
<td>2</td>
<td>2</td>
<td>100%</td>
<td></td>
</tr>
<tr>
<td>Subtotal fasciectomy</td>
<td>4</td>
<td>2</td>
<td>50%</td>
<td>13</td>
<td>3</td>
<td>23%</td>
<td></td>
</tr>
<tr>
<td>van der Veer [21]</td>
<td>Local excision</td>
<td>5</td>
<td>5</td>
<td>100%</td>
<td>4</td>
<td>3</td>
<td>75%</td>
</tr>
<tr>
<td>Partial fasciectomy</td>
<td>18</td>
<td>11</td>
<td>61%</td>
<td>5</td>
<td>2</td>
<td>40%</td>
<td></td>
</tr>
<tr>
<td>Plantar fasciectomy</td>
<td>4</td>
<td>1</td>
<td>25%</td>
<td>4</td>
<td>2</td>
<td>50%</td>
<td></td>
</tr>
<tr>
<td>Surgery + RT</td>
<td>5</td>
<td>2</td>
<td>40%</td>
<td>2</td>
<td>2</td>
<td>100%</td>
<td></td>
</tr>
<tr>
<td>Surgery without RT</td>
<td>22</td>
<td>15</td>
<td>68%</td>
<td>11</td>
<td>6</td>
<td>54.50%</td>
<td></td>
</tr>
<tr>
<td>Ushijima [19]</td>
<td>Unspecified excision</td>
<td>12</td>
<td>3</td>
<td>25%</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Haedicke [22]</td>
<td>Wide excision</td>
<td>2</td>
<td>0</td>
<td>0%</td>
<td>2</td>
<td>0</td>
<td>0%</td>
</tr>
<tr>
<td>Rao [23]</td>
<td>Local excision</td>
<td>3</td>
<td>3</td>
<td>100%</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Dermofasciectomy</td>
<td></td>
<td></td>
<td></td>
<td>3</td>
<td>1</td>
<td>33.30%</td>
<td></td>
</tr>
<tr>
<td>Fetsch [20]</td>
<td>Local excision</td>
<td>38</td>
<td>32</td>
<td>84.20%</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Operative approach and extension of the excision was reported in 10 on 11 studies of the surgery group. 13 cases (reported in 2 studies [18,21]) underwent radiotherapy after operative treatment.

Considering the whole population surgically treated, 136 recurrences were reported; 101 recurrences (74.3%) occurred in primary cases, 35 (25.7%) in patients already operatively treated (Table 3). Local excision showed the highest rate of failure, subtotal fasciectomy and wide excision showed to lower rate. Only 1 study [8] reported the variations of the radiological values of the foot after operative excision.

4. Discussion

Plantar fibromatosis is a benign condition, but characterized by local aggressiveness and high recurrence rate [8,22,24,25].

The disease generally starts with an asymptomatic nodule in plantar fascia with a slow progression in size [26]. Only occasionally the lesion grows rapidly and can be confused with a low-grade fibrosarcoma on histopathology [27]. After a variable time, the lesion may progress in size and become more painful.

The lesion is typically located at the longitudinal medial arch of the plantar fascia of the foot. Lesions in the distal aspect of the great toe have also been described [28]. The lesion can be locally aggressive extending in fat and fibrous tissue, can be adherent to the overlying skin and muscles but it does not invade adjacent nerves or blood vessels [22].

It is important to properly differentiate this lesion from other diseases like lipoma, fibrosarcoma, cyst. Histological exam of the lesion surgically excised is recommended for the definitive diagnosis of plantar fibromatosis.

Conservative treatment should always be the first approach. In this review 5 studies [10,13–16] regarding conservative treatment were included: one [13] about local injection of steroids, one [16] about local injection of collagenase of Clostridium hystolyticum, one about extracorporeal shockwave [15] and 2 [10,14] about local radiotherapy (as single treatment, without surgery associated).

Only the 2 studies [10,14] concerning radiotherapy treatment considered the complete remission from the disease as outcome. The other 3 studies [13,15,16] described treatments with no possibility of healing from the disease and they have considered as outcome the satisfaction of the patient, the pain and the modifications of the consistency and size of the nodules.

Pentland and Anderson [13] reported one case of Ledderhose disease treated with 5 monthly injections of triamcinolone acetonide and lidocaine. At the 9 months followup it has been found an improvement of the symptoms, a smaller lesion with no complications associated.

It is well known the risk of dermal atrophy after local injections of steroids and patients should be informed of this complication.

Hammoudeh [16] reported one case treated with 3 injections of collagenase of Clostridium hystolyticum, a treatment known to be effective in Dupuytren's contracture and Peyronie's disease. Patient has reported no clinical improvement at the end of the treatment.

For the authors the poor outcome is due to the anatomical properties unique to this disease in the plantar region (which typically includes a nodule instead of a cord or plaque). However, additional studies are necessary in order to evaluate the effectiveness of this treatment regarding plantar fibromatosis.
Knobloch and Vogt [15] have considered the treatment with high-energy extracorporeal shockwave in 6 patients with plantar fibromatosis. Extracorporeal shockwave at 2000 impulses at 3 Hz of frequency were applied in 2 therapeutic sessions. All patients have reported an improvement of the pain (evaluated with VAS scale) and have noted a softness of the nodules.

Heyd et al. [14] and Grenfell et al. [10] considered the treatment of plantar fibromatos is with local radiotherapy.

Heyd et al. [14] used two different dose fractionation scheduled. 28 cases were treated with five weekly fractions of 3.0 Gy, repeated after a period of 6 weeks to a total dose of 30.0 Gy. 5 cases received two single fractions of 4.0 Gy on consecutive days, repeated at intervals of 4 weeks to cumulative dose of 24–32 Gy. Authors have found a complete remission from the disease in 11 sites treated, a partial remission in 18 cases and 4 cases where the lesion was stable. Complete remission from pain was achieved in 13/19 patients.

Grenfell and Borg [10] have treated 3 patients with a dose of 15 Gy in five fractions at 3 Gy per fraction with a second analogue treatment after a 6 weeks break. At followup, 5 sites responded symptomatically, with 3 sites reduced in size.

Radiotherapy showed good outcome, but concerns exist about side effects, above all regarding the skin. Heyd et al. [14] found at the followup 6 cases of erythema or hyperpigmentation and 3 cases of moderate soft tissue fibrosis and increased dryness of the skin.

The main indications for operative treatment of plantar fibromatosis are: painful plantar mass not responding to conservative treatments, growing mass, difficulty in fitting footwear, altered activity level [8].

Important finding of this review is that local excision was associated with the highest rate of recurrence, both in primary and in recurrence cases.

Since not all the studies reported the complications, it is not possible to compare the rate and the severity of the complications of the various techniques. It could be possible that local excision, for the necessity of a smaller approach and less extensive excision, has a lower risk of complications in comparison to the other technique.

Based on these results, local or marginal excision are not recommended in the treatment of this disease, both in primary than in recurrence cases.

An exception could be lesions still very small and locally not aggressive, in patients with a low risk of recurrence (no familiarity, not bilateral, no Dupuytren’s or Peyronie’s disease associated). These cases could benefit from a less invasive surgery.

Partial fasciectomy and wide excision were found to have the lowest recurrence rate, both in primary than in recurrence group. This outcome suggests the need of a wide margin excision for reducing the risk of relapse.

The wider excision of the subtotal fasciectomy leads to a higher risk of delayed skin complications.

Sammarco and Mangone [8] found 11 delayed skin healing on 23 cases treated. 4 of these cases required a skin graft. On the other hand Kadir and Chandrasekar [29] showed, as complications 19 cases treated, only 2 cases of scar pain and one case of hypersensitive scar.

Considering the low rate of recurrences of the studies, these are the most recommended approaches to both primary and recurrent lesions.

Recurrent lesions are generally more aggressive and multiple [30], and should be treated more aggressively.

Total plantar fasciectomy was used both in primary than in recurrence cases. It has reported [17,21,23] low recurrence rate in both groups, but only few cases (7 primary, 8 recurrence) were treated with these techniques. For this reason, it is not possible to compare plantar fasciectomy with the other techniques.

Plantar fasciectomy is more invasive than subtotal fasciectomy and has more risk of wound healing problems and necessity of skin graft.

Incomplete excision of the lesion represents the most important factors for recurrence prediction. de Bree et al. [18] reported that all microscopically incomplete excisions were followed by recurrence. Other studies, instead, showed that incomplete excision is not automatically linked to higher recurrence rates [13,31,32].

One explanation to the high recurrence rates could be the unencapsulated nature of the lesion, leading to an incomplete microscopically removal of the lesion [26].

The approach to the plantar fascia in order to excise the lesion requires an accurate dissection to avoid injuries to neurovascular structures and respect margin of surrounding healthy tissue [8]. Several authors reported local complications due to the approach to the plantar fascia. Wapner et al. [12] reported 2 cases of medial plantar neuroma; Alusio et al. [26] reported 2 cases of lateral plantar nerve laceration and one case of medial plantar nerve entrapment.

Since the lesion is benign and radiotherapy may lead in long term to secondary malignancies, Dürre et al. [17] recommended radiotherapy only for nonresponding operative cases [32].

de Bree et al. [18] used the association of surgery and radiotherapy only in recurrent cases, instead van der Veer et al. [21] also in primary cases.

This technique has showed good outcomes. In de Bree case series there was 1 recurrence on 6 cases treated.

Instead van der Veer found 2 recurrences on 5 primary cases (40% recurrence rate, less than 68% of cases treated with surgery alone) and both the recurrent cases relapsed.

de Bree reported 3 cases of dystrophic foot with impaired foot function as side effects of postoperative radiotherapy.

The low number of patients treated with surgery and radiotherapy does not permit to understand if it is an effective treatment compared to the side effects.

Overlying skin should be excised if involved and may result in a skin defect, requiring skin graft. de Bree reported that excision of uninvolved overlying skin does not decrease recurrence rate [18].

Sammarco and Mangone [8] described an intraoperative tumor grading system in four grades, based on unifocal–multifocal disease, possible deep extension to the flexor sheat and adherence to the skin layer. In their case series of 21 feet, patients with a grade III or IV experienced frequently delayed surgical wound healing. Authors did not recommend skin grafting during the initial procedure or within the first weeks postoperatively, since many patients will heal with local wound care alone.

Oster and Miller [33] proposed interposition of marlex mesh after excision of plantar fascia, but only with a low improvement.

Ushijima et al. [19] reported the clinicopathological relationship of 12 cases of plantar fibromatosis. All 3 recurrences reported in this study were in proliferative phase; no recurrence was observed in lesions in involutinal and residual phase. No other article included in this review compared histological findings with clinical outcome and recurrence rate. Further studies should be necessary in order to correlate the phase of the disease with the risk of recurrence.

Only one of the studies included in this review analyzed how change the radiological measurements of the foot after operative procedure.

Sammarco and Mangone [8] measured the changes in longitudinal arch height measurement with weight-bearing foot X-rays in 14 patients (18 feet) who underwent subtotal plantar fasciectomy. Authors found a decrease of the height in the medial longitudinal arch. Other radiological findings were a reduction of calcaneal pitch, medial cuneiform height and navicular height.
Authors did not find a correlation between modification of radiological measurements and residual symptoms of the patients. Lelderhose disease rarely involves pediatric population. Very few studies in literature reported pediatric cases, Jacob and Kumm [34] proposed to distinguish pediatric form of plantar fibromatosis with the definition of “benign anteromedial plantar nodules of childhood”. Pediatric lesions are located in the anteromedial region of the plantar fascia and have no association with lesions of hands or penis. They are asymptomatic and not aggressive, unlike the adult presentation of the disease; as in adults, after excision the lesion often recurred. Treatment in pediatric cases should be conservative [35].

5. Conclusion

Local steroid injection is a valid conservative method, already used in mostly cases. Radiotherapy showed good clinical outcome but the benefits compared to the side effects are not clear. Extracorporeal shockwave showed promising outcome, but further studies on more patients are necessary to evaluate outcomes and possible complications. Injections of collagenase of C. histolyticum are not recommended.

Regarding operative treatments, local excision showed the highest rate of recurrences and should not be recommended as treatment neither in primary nor in recurrent cases. Plantar fibromatosis is locally very aggressive; excision with wide operative margins should be considered even in presence of a small and single nodule.

For this reason, subtotal fasciectomy and wide excision should be recommended for the treatment of both primary and recurrent lesions.

Anyway, every single treatment option should be individualized based on the dimensions of the lesion, if it is a primary or a recurrent lesion, patient’s risk factors of aggressiveness and recurrence such as familiarity, multiple nodules, lesion size and association with Dupuytren’s or Peyronie’s disease.

Further clinical trials with well-defined and standardized outcome measurements should be necessary in future to better evaluate success rate and complications of the various procedures.

Authorship

All authors were fully involved in the study and preparation of the manuscript, the material within has not been and will not be submitted for publication elsewhere. No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.

Conflicts of interest

None.

References