

# Pterygium Inversum Unguis: Report of an Extensive Case With Good Therapeutic Response to Hydroxypropyl Chitosan and Review of the Literature

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## ABSTRACT

Pterygium inversum unguis is a rare but not exceptional dermatological condition, with few descriptions in literature. It occurs more frequently in females and may be associated with several clinical conditions. About 50% of cases are concurrent with collagen diseases such as systemic lupus erythematosus and scleroderma. Severe cases are accompanied by moderate morbidity caused by discomfort when the patient has to perform minor tasks. The treatment has been considered complex, regardless of its underlying cause, with poor response to the topical therapies such as keratolytics and corticosteroids. This paper reports a case of pterygium inversum unguis with a good therapeutic response to hydroxypropyl chitosan and includes a review of the literature.

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## CASE REPORT

A 32-year-old woman presented with pain, discomfort, and bleeding when she clipped her nails. She had no other symptoms or skin lesions. The condition had been present for 4 years, and she had refused any treatment previously. On examination, a forward extension of the hyponychium was adherent to the ventral aspect of the nail plate. The condition involved all nails on both hands and spared the toenails (Figure 1). There was no relevant family history.

A treatment recently used for psoriatic nail dystrophy<sup>1</sup> was initiated with once-daily application of a water-soluble nail lacquer containing hydroxypropyl chitosan, horsetail extract (*Equisetum arvense*), and methylsulfonylmethane. After 2 months of the daily treatment, examination revealed considerable improvement in the nail changes (Figure 2). After 4 months of treatment, there was an overall improvement in the condition, but with some persistence of the lesion on the third fingernail.

## DISCUSSION

Pterygium inversum unguis (PIU) is a disorder consisting of a forward extension of the hyponychium (a subungual structure distal to the nail bed) anchoring to the undersurface of the nail plate and thus obliterating the distal nail groove. It may affect a single finger or multiple fingers, as shown in this case. PIU may be congenital and/or familial, or acquired.

PIU was first described in 1973 by Caputo and Prandi,<sup>2</sup> in a female patient who presented with ventral pterygium in mul-

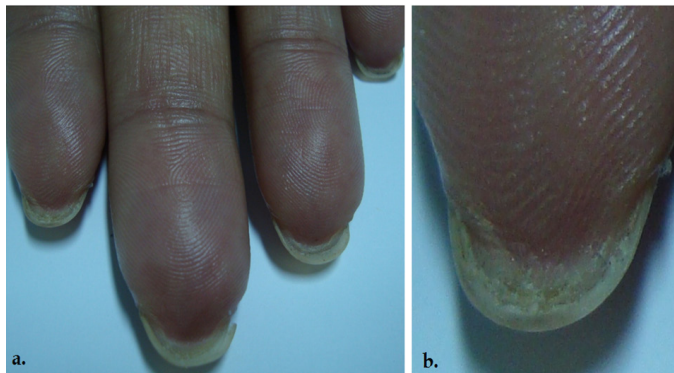
tiple fingers of the hands, with no apparent cause, similar to the case described in our paper. Since then, some cases have been described as related to systemic diseases, principally including collagen diseases,<sup>3,4</sup> allergic dermatitis,<sup>5,6</sup> neurofibromatosis, hemiparesis (due to stroke),<sup>7</sup> subungual exostosis,<sup>8</sup> and leprosy.<sup>9</sup> A congenital type was first described by Odom et al.<sup>10</sup> The causes of PIU (*ventral pterygium*) are summarized in Table 1.<sup>2,4-6,8-9,11-18</sup>

Women are more frequently affected than men, with ages ranging between 20 and 70 years. The most common complaints are pain and bleeding occurring on clipping the nails.<sup>19</sup> In about 50% of cases there is a history of associated collagen disease. In addition, in some cases, there is a family history of the condition.<sup>12,13,16,18,20,21</sup> Sometimes the condition is idiopathic.<sup>14</sup>

Hand nails are more often affected than foot nails with a predominance of spontaneous cases. Because of its rarity, the appropriate mechanism of origin is speculative,<sup>7,19</sup> but blood alterations and ischemia are the most acceptable hypotheses.<sup>7</sup> A similar mechanism has been considered in patients with leprosy.<sup>9</sup> Peripheral vascular and sensorineural alterations were suggested as genesis of morbidity in a case report associated with stroke.<sup>7</sup>

On physical examination of these patients, it is possible to verify that there is a thick layer of keratin between the hyponychium and the ventral face of the nail plate.<sup>14</sup> This is histologically corroborated with an intense hyperkeratosis tightly fixed to the nail

**FIGURE 1.** Pterygium inversum unguis. **a)** Initial presentation of the diagnosis, with involvement of all fingers on both hands. **b)** Detail of pterygium inversum on the second finger of the left hand.



**FIGURE 2.** Pterygium inversum unguis. Patient results after 4 months of treatment. There is an improvement in the general profile with a discrete persistence on the third finger.



**TABLE 1.**

**Causes of Pterygium Inversum Unguis (Ventral Pterygium)**

Acrylate allergy <sup>6</sup>
Causalgia of the median nerve <sup>11</sup>
Congenital and/or familial <sup>12,13</sup>
Formaldehyde-containing hardeners <sup>5</sup>
Idiopathic <sup>14</sup>
Lenticular atrophy of the palmar creases <sup>15</sup>
Leprosy <sup>9</sup>
Systemic lupus erythematosus <sup>2</sup>
Neurofibromatosis <sup>16</sup>
Paresis <sup>17</sup>
Scarring in the vicinity of the distal nail groove <sup>4,18</sup>
Subungual exostosis <sup>8</sup>
Systemic sclerosis <sup>4</sup>

plate, associated or not with acanthosis.<sup>7</sup> Oiso et al<sup>22</sup> suggested the pathogenesis may be related to a deficiency in the regulation of the production of the horny layer of the nail isthmus region.

The treatment has been considered complex, regardless of the underlying cause of PIU, with poor response to topical therapies such as keratolytics and corticosteroids.<sup>23</sup> The best therapeutic suggestion is to remove the causative agent identified.<sup>6,23</sup> Our hypothesis is that in PIU there is no parallelism between the nail plate and nail bed growth. However, the cause of this discrepancy of growth is uncertain, as few cases have been reported and there are many causes associated with it.

Therefore, because of the risk of association with systemic diseases and the good therapeutic response achieved in this case, we suggest the use of a flowchart for screening patients with other disorders, treatment of associated diseases, follow-up, and establishment of therapeutic proof (Figure 3).

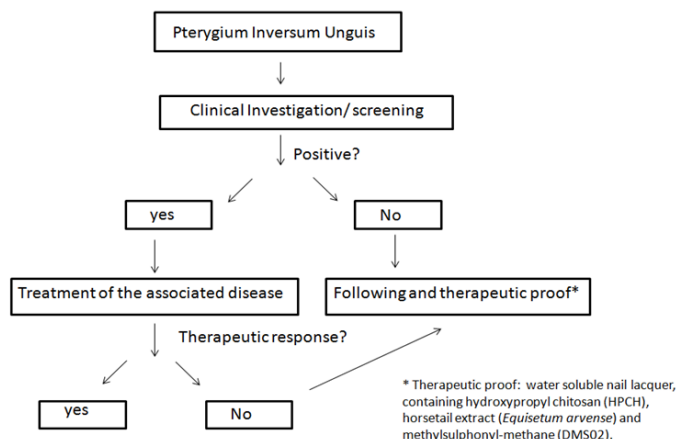
"The knowledge of possible PIU etiologies and therapeutic methods is crucial for the dermatologist in order to appropriately investigate comorbidities and immediately relieve the pain and discomfort manifested by the patient."

Because of the risk of progression of systemic diseases, the patients must be followed, including, when necessary, enlisting the assistance of other medical specialties. Our patient did not present with any clinical and/or laboratory signs of collagen disease or any other systemic diseases. This finding reinforces the hypothesis of an idiopathic condition.

Our case has a clinical importance for its impressive and excellent response to the treatment established. The knowledge of possible PIU etiologies and therapeutic methods is crucial for the dermatologist in order to appropriately investigate comorbidities and immediately relieve the pain and discomfort manifested by the patient.

In idiopathic cases that fail to respond to local hydroxypropyl chitosan, a surgical technique provides relief from pain. After avulsion of 5 to 6 mm of the distal nail plate, a strip of nail bed and hyponychium 3 to 4 mm wide is resected and replaced by a split-thickness graft.<sup>24</sup>

In conclusion, our good result using simple topical treatment now needs to be confirmed.

**FIGURE 3.** Flowchart for the screening, follow-up, and treatment of pterygium inversum unguis patients.**AUTHOR CORRESPONDENCE****Roberta Marinho Falcão Gondim PhD**

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**DISCLOSURES**

The authors have no relevant conflicts of interest to declare.

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