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Case Report

A Rare Association of Yellow Nail Syndrome and Elephantiasis Nostras Verrucosa

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Abstract

We report the case of a 64-year-old man with diabetes mellitus who developed progressive dyspnoea, recurrent bilateral pleural effusions, and chronic lower-limb oedema with recurrent cellulitis over 15 years. Imaging revealed bilateral pleural effusions with right-sided bronchiectasis, and skin biopsy showed tissue oedema with dilated lymphatic channels. The combination of persistent pleuropulmonary disease and lymphedema, along with classical nail changes, strongly suggested the possibility of Yellow Nail Syndrome with secondary elephantiasis nostras verrucosa. The patient partly improved with targeted antimicrobial therapy, anticoagulation, and supportive care. There was no definite association of yellow nail syndrome with elephantiasis nostras verrucosa reported in the literature. But chronic lymphatic malformation and lymph edema were considered risk factors for elephantiasis nostras verrucosa. This case underscores the need to consider lymphatic disorders in adults with recurrent pleural effusions and chronic lower-limb changes, even when nail abnormalities were subtle.

Keywords: *Elephantiasis Nostras Verrucosa; Yellow Nail Syndrome; Lymphedema; Pleural Effusion*

Introduction

Yellow Nail Syndrome (YNS) is an uncommon disorder defined by the triad of nail changes, lymphedema, and respiratory involvement. As its features may appear at different times and nail abnormalities can be subtle or absent, the diagnosis is frequently overlooked. Pleural effusions linked to YNS are often bilateral, recurrent, often chylous, and resistant to routine management.

Elephantiasis nostras verrucosa (ENV) is a non-filarial complication of chronic lymphedema or venous stasis marked by hyperkeratotic, verrucous skin changes, frequently associated with diabetes, venous stasis, lymphedema or recurrent cellulitis.

We report a case of yellow nail syndrome with the characteristic dystrophic yellow coloured nails, bilateral pleural effusions, bronchiectasis, and chronic lower limb lymphedema. These features were consistent with YNS with secondary ENV.

Case Report

A 64-year-old retired police officer, a known case of diabetes mellitus on insulin, presented with worsening dyspnoea aggravated by postural changes and a productive cough for one week. He had a history of recurrent pleural effusions and repeated episodes of bilateral lower limb edema and cellulitis.

Fifteen years earlier, he reported recurrent bilateral pedal oedema. Five years ago, he developed progressive dyspnoea and a dry cough. Evaluation revealed bilateral pleural effusion (right > left). Diagnostic evaluation of the right-sided effusion suggested granulomatous pleuritis, and anti-tubercular therapy was initiated but discontinued due to lack of improvement.

One year later, he presented with worsening respiratory symptoms and was found to have a multiloculated effusion on the right side. Chylous fluid was drained and he underwent right-sided decortication. Postoperatively, he continued to experience recurrent episodes of cough, dyspnoea, bilateral pedal oedema, and cellulitis, with over 20 hospital admissions in the past 3-4 years. He described progressive lower limb oedema with thickening, wrinkling, and nodular eruptions extending up to the knees, resembling elephantiasis, along with recurrent erythematous episodes.

During the present admission, the patient was haemodynamically stable with room-air oxygen saturation of 94%. General examination showed a malnourished, dyspnoeic individual. Nails of both upper and lower limbs were curved, discoloured, and atrophic (Figure 1). Lower limbs were oedematous with thickening, wrinkling, and nodular formation extending up to the knees, resembling elephantiasis (Figure 2). Respiratory examination revealed reduced chest movement on the right side, stony dullness over infra-axillary and infrascapular areas, and bilateral crackles (right > left).

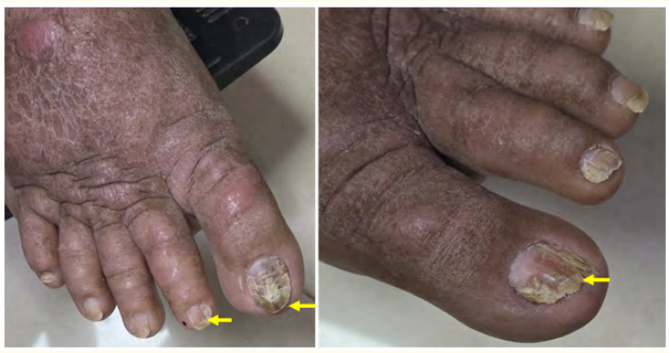

Figure 1: *Nail changes characteristic of Yellow Nail Syndrome. Yellow arrows show curved, discoloured and atrophic nails in the toes.*

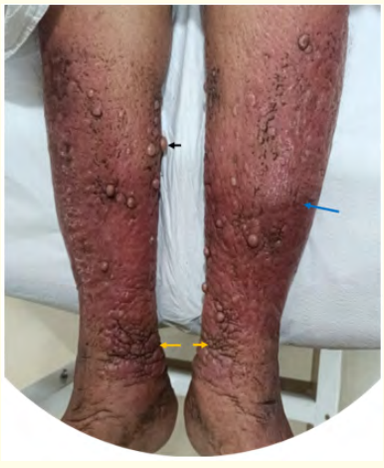

Figure 2: *Chronic edema of the lower limb due to disease affecting the lymphatic channels. Swelling, edema (blue arrow), wrinkling of the skin (Yellow arrows) and nodular formation (Black arrow) can be seen.*

Laboratory investigations revealed leucocytosis with neutrophilia. Sputum culture grew Klebsiella species, and antibiotics were initiated based on culture reports. A dermatology consultation was sought for the skin changes in the lower limbs, and a skin biopsy was taken. Evaluation for filariasis and other causes of persistent lymphedema and hypoproteinaemia was undertaken. Based on dermatology recommendations, antibiotic therapy was modified to crystalline penicillin plus metronidazole. Oral anticoagulation with rivaroxaban was initiated to address possible venous stasis contributing to pedal oedema.

Radiological imaging demonstrated bilateral loculated pleural effusions with volume loss on the right side, bronchiectatic changes of the right lung, and collapse-consolidation of the right lower lobe (Figure 3 and 4). Skin biopsy showed tissue oedema with dilated lymphatic channels. During hospitalisation, the patient showed gradual clinical improvement.

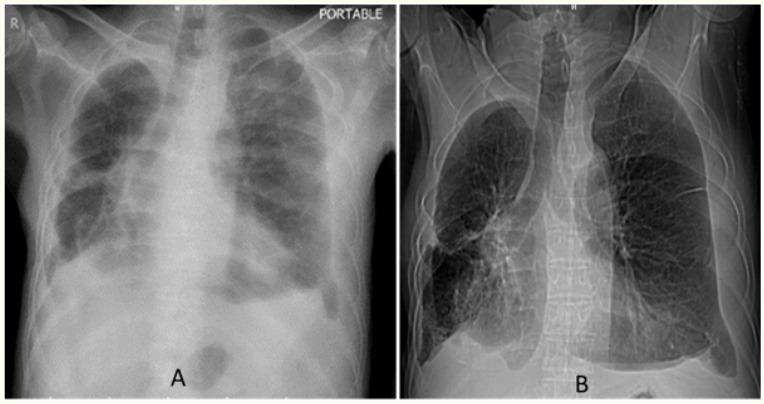

Figure 3: *X-ray chest showing bilateral pleural effusion, bronchiectasis, and pleural thickening. There is volume loss on the right side.*

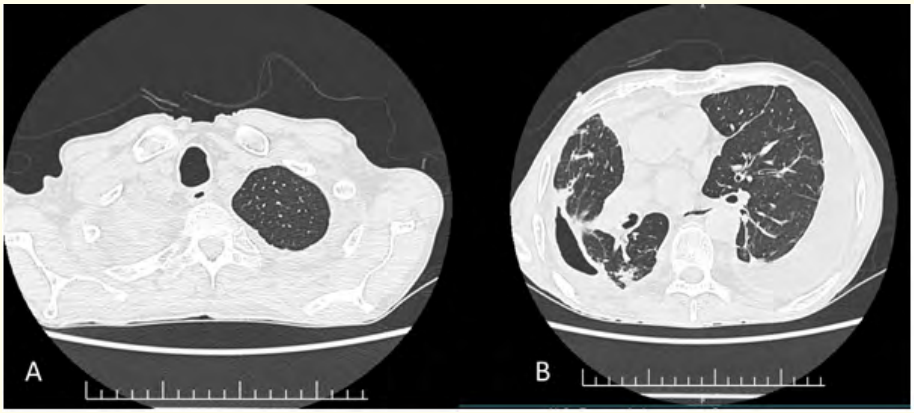

Figure 4: *HRCT Thorax showing pleural effusion, pleural fibrosis, parenchymal fibrosis and nonhomogeneous parenchymal shadows.*

Discussion

Pleural effusion, although a common clinical entity in clinical practice, poses a diagnostic challenge when of a chronic or recurrent nature. Yellow nail syndrome (YNS) is a rare cause of such a presentation. YNS is characterized by slow-growing, yellow, thickened nails, lymphedema, and respiratory tract involvement. The syndrome generally affects adults aged 50 and older. It is defined by the presence of 2 of the following 3 symptoms:

1. Slow-growing, hard, yellow, and dystrophic nails.
2. Lymphedema.
3. Respiratory tract disease [1].

The nails are typically yellowish green in colour, thickened and excessively curved from side to side. They grow slowly, and onycholysis is frequently present. Pulmonary symptoms, such as pleural effusions or bronchiectasis, often accompany the nail abnormalities, while lymphedema may involve the extremities. The pleural effusions are bilateral in about 50% and vary in size from small to massive. Pleural fluid is usually clear, exudate with lymphocyte predominance, or chylous. Once a pleural effusion has developed with this syndrome, it tends to persist and recur rapidly after thoracocentesis. Pulmonary function testing in YNS is typically unremarkable, and biopsies do not usually contribute to the diagnosis [2]. The lymphedema is often mild, usually affects the lower extremities, and can be demonstrated by peripheral lymphangiography [3]. The three separate entities may be manifested at widely varying times [4]. Occasionally, this syndrome may have associated bronchiectasis, sinusitis, pericardial effusion, or even chylous ascites [5].

The syndrome has been reported in association with a variety of other diseases, including immunologic deficiency in siblings, thyroid disease, hypogammaglobulinemia in various forms, nephrotic syndrome, a transient decrease in circulating B lymphocytes, and protein-losing enteropathy [6]. The most widely accepted explanation for the signs and symptoms associated with YNS is a dysfunction of the lymphatic system, specifically lymphatic drainage. Imaging the lymphatic system and lymph transport using lymphoscintigraphy is abnormal in YNS [7].

In our patient, the combination of the classical nail changes, long-standing bilateral pleural effusion with bronchiectatic changes, and biopsy-proven lymphatic dilatation may represent a typical form of yellow nail syndrome. This underscores the importance of considering YNS or related lymphatic disorders in adults presenting with recurrent or unexplained pleural effusions and lower limb lymphedema, when subtle nail changes are present.

Classically, the term elephantiasis applies to swelling of the legs caused by the helminth *Wuchereria* species. The worms block lymphatic channels, leading to severe chronic swelling in the lower legs and genitals. In 1934, the term “nostras” was added to distinguish lymphoedematous disorders of temperate regions not caused by filariasis [8]. In recent papers, however, the term ENV is more often used to denote a dermatological complication of bacterial or non-infectious lymphatic obstruction characterized by dermal fibrosis and epidermal changes consisting of hyperkeratotic, verrucous, and papillomatous lesions [9].

Many causes, such as tumor obstruction, radiation, trauma, hypothyroidism, chronic venous stasis, and some infections, except filariasis, can lead to the development of ENV. Some risk factors are determined, such as obesity, type 2 diabetes, a history of infection, and a low socio-cultural level. The pathophysiology of ENV is not well understood, but it has been observed that the chronic accumulation of lymph and protein-rich interstitial fluid in the tissues increases the volume of the limb and causes tissue deformations. Mossy papules, plaques, and cobblestone-like nodules are clinically impressive features of ENV, but biopsy reveals only moderately abnormal findings such as pseudo-epitheliomatous hyperplasia, dilated lymphatic spaces, fibrous tissue hyperplasia, and chronic inflammation.

In this patient, the progressive lower limb oedema with verrucous and nodular skin changes, supported by biopsy showing dilated lymphatic channels, was consistent with elephantiasis nostras verrucosa. Unlike filarial elephantiasis, this represented a non-filarial form likely resulting from chronic venous stasis with lymphatic obstruction and recurrent cellulitis. Recognition of ENV in such settings is crucial, as timely diagnosis and multidisciplinary care may help reduce the morbidity.

There is no documented direct association between elephantiasis nostras verrucosa (ENV) and yellow nail syndrome (YNS) in the medical literature; however, both are chronic lymphatic conditions that can present with lymphedema [10]. In ENV, the increased volume of the lymphedematous limb is caused by excess lymph accumulation, fibrosis resulting from fibroblast stimulation and excess adipose tissue due to adipocyte stimulation [11,12]. This may lead to thickening and nodular formation as in our case. In chronic/long-standing cases, if lymphedema is severe and recurrent infections occur, theoretically, the skin could undergo ENV-like changes. Here, we postulate that the ENV in this patient may be the result of chronic lymphedema of YNS.

Conclusion

This case highlights a rare but possible association between yellow nail syndrome and elephantiasis nostras verrucosa. YNS and ENV share a common denominator in lymphatic impairment. ENV could develop as a secondary skin complication in a YNS patient with severe, long-standing lymphedema, though this association is rarely reported. Early recognition and multidisciplinary management may mitigate the progression to disfiguring complications such as ENV.

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