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Case Report

Dermatophytic Disease with Ganglion Extension: A Rare Case of a Young Moroccan Man

Abourak Chaimae[1]*, Elmaati Maha[2], Oukassem Siham[1], Guennouni Asmae[1], Laamrani Fatima Zahra[1], Jroundi Laila[1], Senouci Karima[2] and El Aoufir Omar[1]

[1]Department of Emergency Radiology, CHU Ibn Sina, Faculty of Medicine and Pharmacy of Rabat, Mohamed V University, Rabat, Morocco

[2]Dermatology Department, CHU Ibn Sina, Faculty of Medicine and Pharmacy of Rabat, Mohamed V University, Rabat, Morocco

***Corresponding Author:** Abourak Chaimae, Department of Emergency Radiology, CHU Ibn Sina, Faculty of Medicine and Pharmacy of Rabat, Mohamed V University, Rabat, Morocco.

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Abstract

Dermatophytic disease is a rare form of generalized and chronic fungal infection.

It affects the skin and lights, sometimes associating secondary dermohypodermal, ganglionary, or even visceral localizations.

It is probably related to a cellular immunodeficiency in common dermatophytes.

It is mainly described in the Maghreb countries, where it is linked to high consanguinity.

We report a rare case of a Moroccan young man suffering from this disease with secondary ganglion localizations.

Through this article, we discussed the peculiarities of this pathology, the place of imaging, and the bases and difficulties of therapy.

Keywords: *Dermatophytic Disease; Dermatophytes; Ganglion Invasion; Imaging*

Introduction

Dermatophytic disease is a rare and ancient condition; the first case described by Sequeira dates back to 1912 [1]. In 1959, Hadida and Schousboe published the first Algerian case and defined the characteristics of this condition as "exuberant skin lesions associated with multiple visceral localizations" [1,2]. In 1964, Rollier first described, in Morocco, a scalp scab associated with nodosities of the subcutaneous tissue with polyadenopathies and the presence of *T. violaceum* [1]. In 1978, Puissant., *et al.* described the first case in Tunisia in a 35-year-old man with dermatophaneary and ganglionary impairment due to *T. violaceum* and *T. schoenleinii* [1].

In literature, it has been known by various names: dermatophytic disease, Hadida and Schousboe's disease, generalized wart trichophytia, and chronic granulomatous dermatophyta [1].

Since its first description by Hadida and Schousboe, about fifty observations have been in the literature [3]. It is described mainly in North Africa (95.6%). Algeria remains the country with the highest number of cases (48.8%), followed by Morocco (22.2%) and Tunisia (17.8%) [1].

Case Report

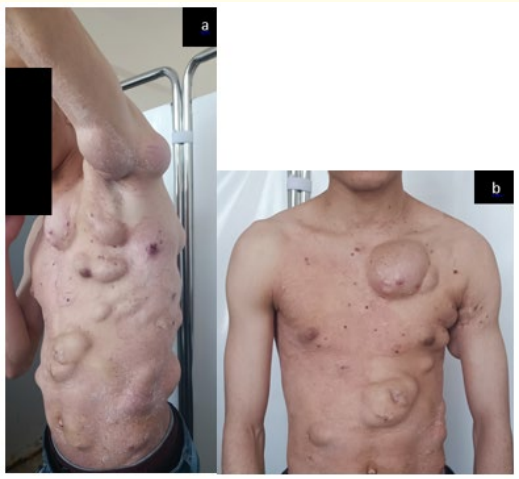
This is a 23-year-old Moroccan young man from an unknown degree of parental consanguineous marriage. 3rd of a well-being 5 brotherhood. There is no notion of tuberculosis contagion or immunodepression.

The history of the disease dates back to the age of 3 years, with the installation of an alopecia plaque treated as a reign with a favorable development for the first time, followed by numerous recidivism with the appearance of itchy erythematoskeletal lesions at the level of the trunk and large folds for which the patient continued in self-medication by taking oral antimycotics at the discretion of recurrences, resulting in a diffuse alopecia of the scalp.

In 2009, at the age of 9, he was operated on for surgical drainage of a subcutaneous abdominal wall abscess, for which he was given Terbinafine 250 mg/day. In 2014, the patient developed multiple nodules fistulating with an outpouring of pus in the scalp and trunk with the extension of erythematous lesions. He consults with many doctors who put him under various treatments without improvement before he was referred to the university hospital center in 2018, where a skin biopsy objectivized an aspect of mycosis with the mycological examination of pus, the biopsies and squamous were a *T. rubrum*, and he was put under different treatments: Terbinafine 250 mg/day from 08/01/2018 to 06/01/2018; Terbinafine 500 mg/day from 06/06/2018 to 03/10/2018; Griseo 1g from 03/10/2018 to 12/12/2018; Griséo 1,5g from 12/12/2017 to 14/09/2022.

Between 2019 and 2022, he was out of sight with worsening injuries. Reviewed on September 14, 2022, where he was given Fluconazole 300 mg/week until February 20, 2023, when he stopped the treatment by himself with aggravation of lesions and appearance of other abscesses.

Dermatological skin objective examination (Figure b and d): a phototype III. Several gums spread over the trunk, scalp, and root of the two upper limbs at different stages of evolution. Several lesions of renitent consistency evoking subcutaneous abscesses, the most voluminous measuring 15 cm of the large axis at the level of the left hypochondrium. Large erythematous calamus wardrobe with circular edges extended on the trunk in the geographical map. Hypochromo-atrophic scars on the back Varicose hamartomas in the left hip Confluent nodules at the conica of the two ears at the level of the scalp: scarring alopecia; persistence of a few hairy regions at the occipital and frontal levels; appearance of pseudo-comedones open across the entire scalp; two large renitent masses of the occipium; disseminated subcutaneous nodules (Figure c). Painful submandibular, pre-tragial, cervical, axillary, and inguinal adenopathies (Figure a).



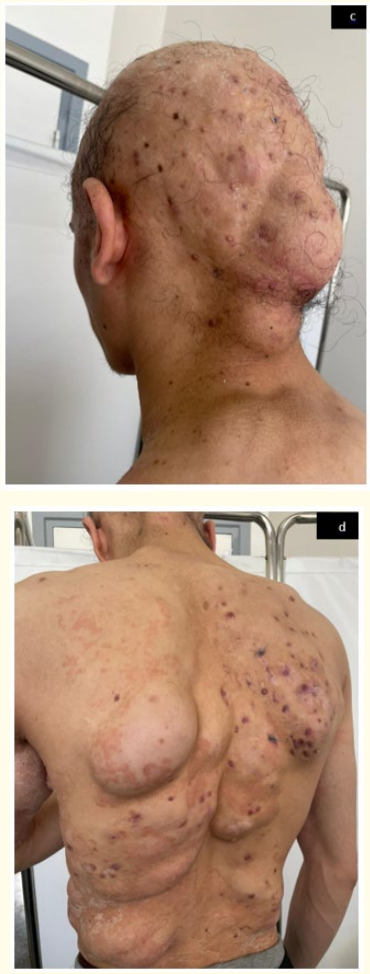


Figure a-d: *a: Clinical picture of the axillary cavity: massive painful axillar adenopathies. c: Clinical image of the scalp: scarring alopecia, persistence of a few hairy regions at the occipital and frontal levels, appearance of pseudo-comedones open across the entire scalp, tow large renitent masses of the occipital, disseminated subcutaneous nodules. b and d clinical images of the trunk and back: Several gums spread over the trunk, scalp, and root of the two upper limbs at different stages of evolution. Several lesions are evoking subcutaneous abscesses. Large erythematous calamus wardrobe with circular edges extended on the trunk in the geographical map. Hypochromoatrophic scars on the back.*

An ultrasound of the ganglionary areas was performed, identifying several of the necrosis adenopathies at the cervical and axillary levels. A cervico-thoraco-abdomino-pelvic CT was requested, objectivating multiple collections of subcutaneous parietal soft parts, ovals with elevated walls after injection of contrast product, sitting at the cervical level, the chest, and the abdomino-pelvic and axillary walls, but remaining superficial are signs of muscle extension or in depth. A cerebro-facial MRI was requested to study the relationships of the facial nerve with a parotid collection, returning in favor of two abcess lesions of the superficial lobe of the right parotid, which are at a distance from the anatomical location of the face nerve. Multiple abscess formations under the scalp and posterior cervical skin with suppurated lateral-cervical adenitis No intracerebral extension (Figure e-h).

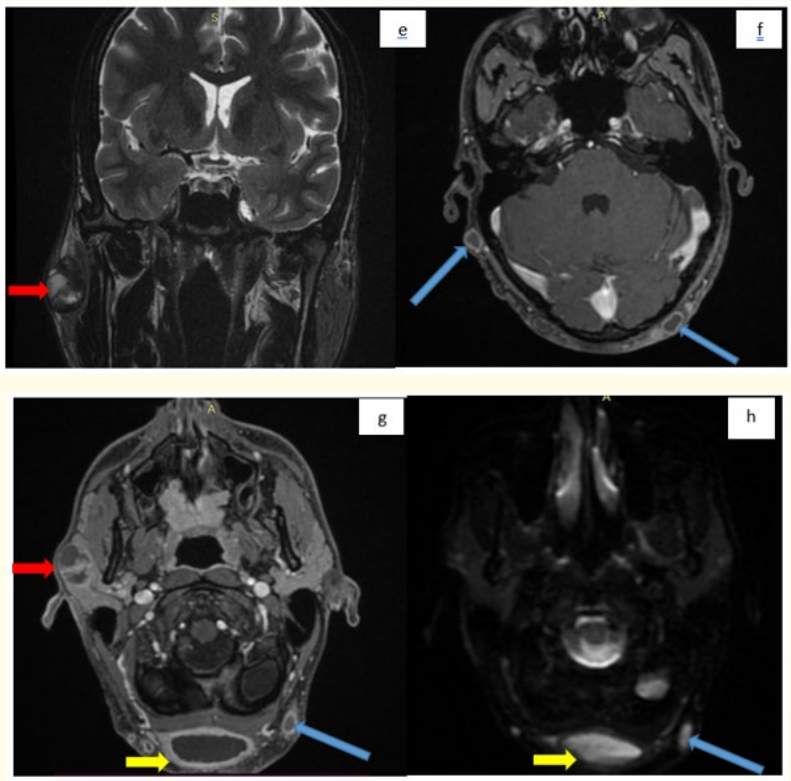


Figure e-h: *Cerebro-facial MRI.*

e: T2 fine coronary plan: two abscessed lesions of the superficial lobe of the right parotid heterogeneous signal (red arrow).

f and g: T1 axial plans after gadolinium injection: the two parotid abscess lesions (red arrow) and multiple abscess formations under the scalp skin (blue arrow), enhanced on the periphery after gadolinium, the largest of which is at the occipital level (yellow arrow).

h: Diffusion: Restriction of the diffusion of abscess collections under the scalp skin (yellow and blue arrows).

In the face of these bands of arguments, the diagnosis of a dermatophytic disease with ganglionary extension and without visceral extension was held.

From the therapeutic point of view, the patient was operated on for a flat placement for cervical and chest abscesses twice with mycological and bacteriological studies, and then he was released under 1.5g of Griséo.

Discussion

Dermatophytic disease is an autosomal recessive genetic disease characterized by a selective deficit of cellular immunity in dermatophytes. The CARD9-deficient gene has recently been described [1-7].

It is mainly described in the Maghreb countries, where it is linked to high consanguinity [1,2,5-7]. Multiple injuries to members of a family are significant, at 34.7% [1,2,6]. The frequency of consanguinity and the notion of family cases suggest the existence of an immune deficit of recessive transmission, causing a state of tolerance towards dermatophytes [1,2,6,7]. Indeed, our patient came from a parental

consanguineous marriage of unspecified degrees. But his brothers and sisters are well-being. Some sporadic cases have been in Central Europe, Japan, and Aboriginal Australia [1,2].

As far as we know, the immune mechanisms that can explain both the spread of the lesions and the deep invasion of the dermatophytes have not been studied extensively, and the few studies concern cases of extensive or disseminated dermatophyties in immune-depressed patients [1,2].

In the case of dermatophytic disease, if humoral immunity does not appear to be impaired, as evidenced by the elevation of total and specific IgE and the presence of antitrichophytin antibodies, a cellular immunity deficit has been confirmed [1,7].

In fact, according to Wagner and Sohnle and Kaaman., *et al*. patients with chronic *T. rubum* infections have a negative Trichophytine IDR, while the response to *in vitro* lymphocyte transformation is positive [1]. In rare cases where an immunological scan was performed, it showed the presence of a functional abnormality of cellular immunity with normal CD4 and CD8 lymphocytosis, preservation of polynuclear function, and increased production of IL-1 and TNF. More recently, IL-4 and IL-5 secretors suppressing TC2-type CD8 lymphocytes were thought to be the cause of immunodeficiency [1,7].

Dermatophytic disease mainly affects male subjects (83.3%). It has a predilection for young adults (27.9 years old) but often begins at a pediatric age (93.7%), on average, between the ages of 11 and 26 [1,3]. Indeed, our patient is a young man of 23 years, but the onset of symptoms in his home dates back to the age of 3.

The dermatophytic disease begins with a recurring scalp tint (51.7%), as was the case in our patient, or with glabrous skin damage (41.4%), and rarely with onyxies (6.8%) [1-5].

During the course of the disease, clinical manifestations take on several lesional aspects [1,2]. The scalp is the seat of lesions of different aspects, starting first with a peeling and then with a hair loss; indeed, this was the case in our patient. It then forms squamous, ichthyosiform scarring plates. Alopecia plaques, such as a pelade, can affect all pillar areas, even the eyelids and eyebrows [1,2]. Ulcer-vegetative hair-related lesions have been described [1,2]. At the level of the skin, the lesions are erythematous squamous, which will generalize in the form of extensive superficial dermatophytosis or erythroderma. Intense chronic itching is frequently observed and may be accompanied by lichenification; all these evolutionary lesions are compatible with our case. Forms with generalized ichthyosiform erythroderma with papulonodular lesions on the trunk or face giving a leonine-positive face suspect leprosy have been described [1].

At a more advanced stage, tuberculosis papulonodules appear, as do dermohypodermal nodosities, subcutaneous abscesses, ulcers, vegetation, and warts; indeed, this was compatible with our case [1,4]. Rubber injuries have even occurred. Plantar damage has been in the form of keratodermia and an exuberant hyper-keratinizing process that forms giant horns, impeding walking [1]. Damaged skin can be the gateway to bacterial and even fungal infections. Onyx is frequently found early and generally affects all the nails of the fingers and toes. Nails are sometimes deformed and thickened by corneal verrucosities or pachyonychia of all nails with onychogryphosis [1].

On the other hand, the mucous membranes are never affected [1], as was the case in our patient. Adenopathies are found frequently. All ganglionary areas can be affected: axillary, inguinal, cervical, and submaxillary. That was the case with our patient. Deep areas are rarely visited and are delayed. Their volume is variable, which can occur in the form of microadenopathies or reach the size of a mandarine. They may undergo inflammatory outbreaks and fistulate, giving rise to ulcerations resembling tuberculosis adenitis [1-5]. Lymphoedema affecting the limbs and genitals has been reported [1].

At a later stage, the lesions can spread deep and gain deep organs (bone, muscle, brain, lung, liver, rat, and peritoneum, for example) [1-4].

The brain injury is in four cases [1,6,7], of which three in Algeria: the first case, by Hadida and Schousboe, the cerebral invasion was confirmed at the autopsy; the second case, in 1977, by Liautaud and Marill; the third case is particular, associating a tronculocortical lesion. The fourth case, consisting of widespread brain and spinal injuries caused by *T. mentagrophytes*, was in Japan. Staturoponderal retardation and even hormonal disorders have also been observed [1].

Dermatophytic disease is a clinical and immunological entity that remains very poorly defined. It has to be differentiated from other entities, mainly extensive superficial dermatophytes, Majocchi granuloma, dermatophyte mycetoma, and other deep dermatophytes [1].

The interest of imaging is to seek an in-depth extension and to evaluate reports of an injury to neighboring organs, as our case has shown.

On the biological level, in addition to inflammatory syndrome, hyperleukocytosis with hypereosinophilia is often found [4].

The direct examination is often positive for both surface and deep samples (fistula pous, ganglion punctions, biopsies, etc.), confirming the diagnosis of disseminated dermatophytosis. The dermatophytes concerned are of anthropophilic origin in 84% and zoophile in 16% of cases [1,3]. The predominant species is *T. violaceum*, either isolated or in combination with other species of dermatophytes (*T. schoenleinii, T. rubrum and M. canis*). This species remains the main dermatophyte of the sacks in the countries of the Maghreb [1,4,6]. Other species are more rarely affected, such as *T. rubrum, M. canis, T. verrucosum, T. tonsurans*, and *T. mentagrophytes* interdigital variety [1,4,6]. Indeed, a *T. rubrum* was isolated in our patient.

Histologically, the most evocative element is the granuloma found in virtually all deep lesions (tubercules, nodules, hypodermal nodosities, ganglions, etc.) [1]. At the nodular level, the architecture of the ganglionary parenchyma is completely altered by the presence of large areas of tissue necrosis, an important reticular hyperplasia in which giant Langhans-type cells surrounded by lymphocytes, plasmocytes, and histiocytes are observed [1].

Currently, there is no codified therapeutic scheme. Indeed, the different treatments used have allowed only partial control of the disease. However, they appear to prevent or delay the visceral spread of the infection. Griseofulvin is the most commonly administered antifungal, prescribed at a dose of 1 g/day and often combined with local treatments [1,4]. Surgical excision may be indicated in cases of brain injury, tumefaction and large-scale adenopathies [1].

Dermatophytic disease remains a serious condition, triggering the life prognosis after years of evolution [1,4].

Conclusion

Dermatophytic disease is a serious and poorly known physiopathogenic disease that may trigger a life-threatening prognosis. The genetic study of this condition opens up new diagnostic and possibly therapeutic possibilities, as well as genetic counseling.

Author's Contribution

C.A, E.M, A.G and O.S. contributed to the conception, acquisition, analysis, interpretation of data, and drafted the manuscript.

O.E. A, F.Z. L, L.J, and S.K critically revised the manuscript and approved it.

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