DERMATOLOGY PRACTICAL & CONCEPTUAL
www.derm101.com

Nodules on the right ear

Carolina Talhari, M.D.[1], Alexandra Maria Giovanna Brunasso, M.D.[2,3], Sinesio Talhari, M.D.[1], Cesare Massone, M.D.[4]

[1] Department of Dermatology, Institute of Tropical Medicine, Manaus, Brazil
[2] Division of Environmental Dermatology and Venereology, Medical University of Graz, Graz, Austria
[3] Department of Dermatology, Galliera Hospital, Genoa, Italy
[4] Division of General Dermatology, Medical University of Graz, Graz, Austria

Key words: nodule, ear, mycosis, lobomycosis

Citation: Talhari C, Brunasso AMG, Talhari S, Massone C. Nodules on the right ear. Dermatol Pract Conc. 2012;2(1):5. http://dx.doi.org/10.5826/dpc.0201a05.

Editor: Harald Kittler, M.D.

Received: June 2, 2011; Accepted: July 20, 2011; Published: January 31, 2012

Funding: None.

Competing interests: The authors have no conflicts of interest to disclose.

All authors have contributed significantly to this publication.

Corresponding author: Cesare Massone, M.D., Department of Dermatology, Medical University of Graz, Auenbruggerplatz 8, A-8036 Graz, Austria. Tel. +43.316.385.3235; Fax. +43.316.385.4957. Email: cesare.massone@klinikum-graz.at.

Case report

A healthy 17-year-old boy with a one-year history of asymptomatic, slowly enlarging nodules affecting his right ear was seen. The patient's past medical history was unremarkable, he was not immunosuppressed, he was not taking any drugs, and he was employed as a rubber worker in the Amazon region. Physical examination revealed ill-defined, smooth, shiny, hard nodules on his right ear (Figure 1). The consistency of the skin lesions was hard, and pain was absent. Regional lymph nodes were not enlarged. Laboratory examination showed unremarkable complete blood count, urinalysis, hepatic and renal function tests, serum glucose, Elisa-HIV and erythrocyte sedimentation rate. Chest X-rays were normal.

Histopathologic examination showed a diffuse granulomatous infiltrate involving the entire dermis, constituted by histiocytes and multinucleated giant cells (Figures 2, 3). The histiocyte's cytoplasm contained rounded yeast-like hyaline cells with a thick double birefringent membrane. These cells were forming chains of multiple organisms. The Grocott-

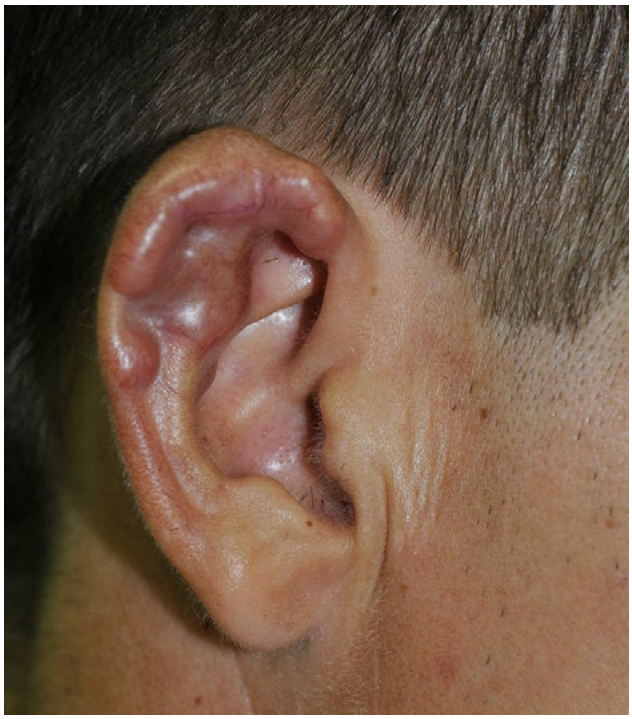

Figure 1. Multiple, non-ulcerated nodules and plaques on the right ear. [Copyright: ©2012 Talhari et al.]

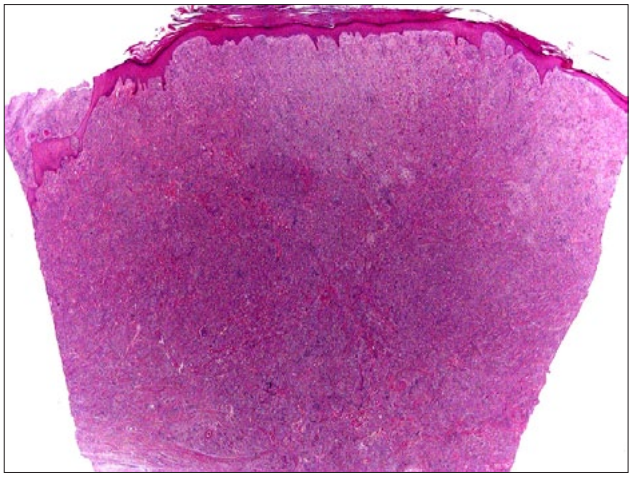

Figure 2. Diffuse granulomatous infiltrate involving the entire dermis (Haematoxilin and eosin [H&E], original magnification: 20X). [Copyright: ©2012 Talhari et al.]

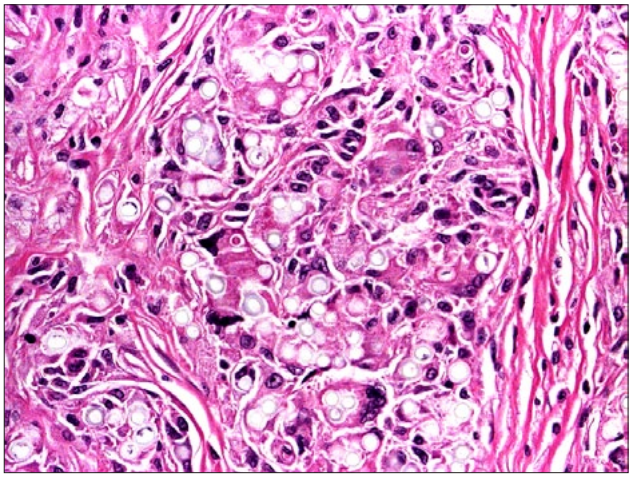

Figure 3. The histiocyte's cytoplasm contains rounded yeast-like hyaline cells with a thick double birefringent membrane. These cells are forming chains of multiple organisms. (H&E, original magnification: 400X). [Copyright: ©2012 Talhari et al.]

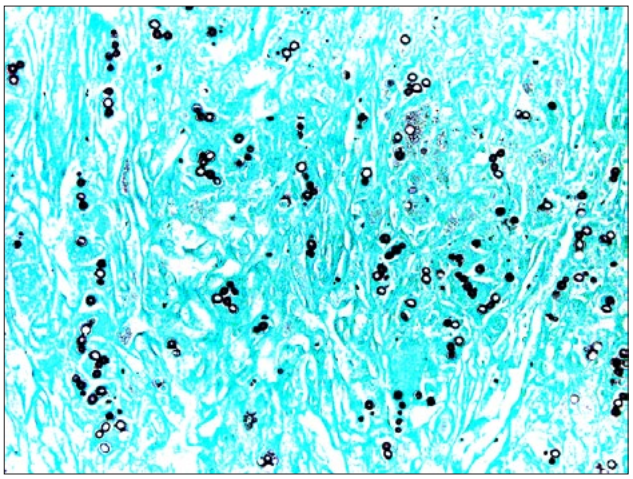

Figure 4. The Grocott-Gomori methenamine-silver nitrate stain shows chains of darkly pigmented, spheroidal, yeast-like organisms (original magnification: 200X). [Copyright: ©2012 Talhari et al.]

Gomori methenamine-silver nitrate stain allowed better visualization of chains of darkly pigmented, spheroidal, yeast-like organisms (Figure 4).

What is your diagnosis?

Answer

Localized lobomycosis

Discussion

Lobomycosis, described by Jorge Lobo, is a deep chronic cutaneous, non-life threatening mycosis restricted to areas of the Amazon, a region characterized by tropical rainy weather [1-3].

The agent of lobomycosis is *Lacazia loboi,* an intracellular spherical yeast of 8-12 nm of diameter that resides in macrophage vacuoles [4]. The fungus is easily found in lobomycotic skin lesions but it has never been cultivated [4]. The precise natural reservoir of *L. loboi* is unknown, but soil and vegetation seem to be probable sources of infection [1-3]. The disease can be found in dolphins and is more prevalent in human beings that live near aquatic environments, suggesting that *L. loboi* is an hydrophilic fungus [1,2,5].

The disease affects mostly young, non–immune suppressed, male patients between 21-40 years of age [1-3]. The direct inoculation into the dermis is probably the way of transmission [4]. The incubation period is long and the infection often goes through a quiescent period (months to years) [3]. The lesions often begin as small papules or pustules associated with a burning sensation or mild pruritus [6]. The lack of other systemic involvement is characteristic [1,6]. The typical keloid-like skin lesions appear only after several months and the typical appearance is "keloids over keloids" [1-3,6]. Lesions have well defined margins, are not attached to deeper structures, and can disseminate by contiguity or by auto-inoculation [1]. The process of dissemination lasts several decades. Clinical differential diagnosis of lobomycosis includes lepromatous leprosy, chromoblastomycosis, keloids, xanthomas, fibromas, Kaposi's sarcoma, neurofibromas and dermatofibrosarcoma protuberans [1]. Moreover, in our specific case, the differential diagnosis includes leprosy, relapsing polychondritis, indolent CD8+ lymphoid proliferation of the ear, and borrelia lymphocytoma (noted only from an academically point of view; the Amazon is not endemic for Borrelia infection).

The fungus load on skin smears is usually very large with a high number of rounded cells that do not need to be stained. Skin sections are characterized by a diffuse granulomatous dermatitis involving the entire dermis and extending to the

subcutaneous fat [7]. The Grocott's stain allows the visualization of chains of darkly pigmented, spheroidal, yeast-like organisms with thick (double) birefringent walls [7].

Treatment is very problematic in heavily infected patients with many large lesions [3]. Prompt diagnosis and treatment are mandatory in order to obtained a high cure rate [1,3]. The most successful treatment option is a wide surgical excision, but relapses are common [1]. Systemic therapy with clofazimine has been successfully used [8]. Other reported therapies are ketoconazole, itraconazole, amphotericin B, and 5-fluorocytosine [1,8]. In our patient the nodules were partially excised, he was given itraconazole 200 mg for 8 months, and is still undergoing therapy.

References

1. Talhari S. Doenca de Jorge Lobo. In: Talhari S, Garrido Neves R, eds. *Dermatologia Tropical*. Rio de Janeiro: Ed Medsi, 1997:237–53.
2. Talhari S, Cunha MG, Schettini AP, Talhari AC. Deep mycoses in Amazon region. Int J Dermatol. 1988;27(7):481–4.
3. Lupi O, Madkan V, Tyring SK. Tropical dermatology: bacterial tropical diseases. J Am Acad Dermatol. 2006;54(4):559–78.
4. Taborda PR, Taborda VA, McGinnis MR. Lacazia loboi gen. nov., comb. nov., the etiologic agent of lobomycosis. J Clin Microbiol. 1999;37(6):2031–3.
5. Haubold EM, Aronson JF, Cowan DF, McGinnis MR, Cooper CR Jr. Isolation of fungal rDNA from bottlenose dolphin skin infected with Loboa loboi. Med Mycol. 1998;36(5):263–7.
6. Silva D. Estudo experimental da micose de Lobo. An Bras Dermatol. 1994;69:88–91.
7. Opromolla DVA, Belone AFF, Taborda PRO, Taborda VBA. [Clinico-pathological correlation in 40 cases of lobomycosis]. An Bras Dermatol. 2000;75:425–34.
8. Fischer M, Chrusciak Talhari A, Reinel D, Talhari S. Successful treatment with clofazimine and itraconazole in a 46 year old patient after 32 years duration of disease. Hautarzt. 2002;53(10):677–81.