

# **Blastomycosis in the lymph nodes of the neck - a case report and review of literature**

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## **Introduction**

Blastomycosis is an uncommon, male predominant disease caused by the fungus *Blastomyces dermatitidis*. This disease is also called “Chicago Disease” because of the concentration of cases at the southern end of Lake Michigan about 100 years ago<sup>1</sup>. The most important endemic areas besides USA are South America, Africa (Ouganda, Mozambic, Zimbabwe, Tanzania, Congo) and Asia near waterways.

The disease is initiated by the inhalation of spores aerosolized from rotting or wet wood in the lungs and than transformed into the yeast, which is responsible for inflammation and systemic dissemination<sup>2</sup>. After several weeks of incubation, the initial clinical presentation is that of an acute pneumonia evolving into a non specific chronic pneumopathy. The primary pumlonary infections rarely may present entirely asymptomatic, in most cases they are associated with hemoptysis, cough and fever. From the pulmonary focus it can spread hematogenously. Cutaneous (80% of patients) and skeletal involvement are the most common extrapulmonary localisations<sup>3,4</sup>. Mucosal involvement is uncommon and reports of oral lesions as first sign of Blastomycosis infection are very rare<sup>5</sup>.

## **Case**

A 52 year old male patient was referred to the Department of Craniomaxillofacial and Oral Surgery of the University Hospital Zurich with a 3 week history of intraoral ulceration on the right lower jaw side. There was no history of pulmonary disease, in particular fever, coughing or hemoptysis. He had a nicotine abuse of 30 py. His medical and family histories were not contributory to this problem.

The first 34 years he lived as a farmer on the countryside in Argentina, he moved in 1988 to Switzerland, but he is regularly on holiday in Argentina. Actually, he is working as a worker in a plastics factory.

The physical examination showed a 3.5\*5cm measuring ulceration in region 43-45 (Fig1), no increased lymph nodes in level I-IV palpable.

There was no pathological finding concerning pulmonary infection. The x-ray thorax (Fig 2) revealed no pulmonary lesions or mediastinal lymph nodes .

The PET-scan and also the CT-scan (Fig3) showed no suspicious lymph nodes of the head and neck region. Under general anaesthesia a tumorresection including a neck dissection of level I-III on the right side was performed.

The histopathological examination of the biopsy specimens revealed a marked hyperplasia of the oral mucosa with subepithelial granulomatous inflammation. The granulomas were well formed without necrosis (Fig 4). Because of partial tangential inclusion of the biopsy fragments the marked pseudoepitheliomatous hyperplasia of epithelium was misdiagnosed as well differentiated squamous carcinoma with a granulomatous stromareaction. A partial resection of the right lower jaw and mouth floor, region 43-44 with unilateral neck dissection

level I to III was performed. The macroscopic specimen showed three ill defined, 1.4 to 1.8 cm large, indurations. The histopathological examination revealed a marked pseudoepitheliomatous hyperplasia of the oral mucosa alternating with focal ulcerations. In the submucosa, were numerous well defined non necrotising granulomas often directly underlying the epithelium. The neck dissection revealed a non necrotising granulomatous adenopathie. On special stains (Grocott) (Fig 5) numerous large (8-15  $\mu\text{m}$ ) oval yeasts were identified in the submucosal granulomas and in some granulomas in the lymph nodes. The yeast showed a thick membrane with unipolar budding.

Postoperatively a CT Thorax scan was performed but did not reveal any typical lesions.

## **Discussion**

Blastomycosis is not common. The estimated incidence is 0.6 cases per million persons per year in the USA<sup>6</sup>. There is no case in Switzerland described in literature. It is virtually restricted to persons spending time in Latin America.

Diagnostic methods include serologic assays. However they are insufficiently sensitive to detect the cell wall protein WI-1 antigen on the surface of Blastomycosis dermatitidis that is needed for the attachment of the organism to host macrophages. The most common diagnostic way is by pathohistology of tissue samples. Also fine-needle aspirations can demonstrate the presence of morphologically typical yeast of Blastomycosis dermatitidis

In the present case, the clinical presentation with isolated chronic ulcerations of the oral mucosa, as well as the pseudoepitheliomatous hyperplasia of the oral epithelium were misleading and the diagnosis of Blastomycosis dermatitidis was only established on the operation specimen. The marked pseudoepitheliomatous hyperplasia of the epithelium with features resembling squamous cell carcinoma is a diagnostic pitfall pathologist should be

aware of. In similar way, skin lesions of Blastomycosis are frequently associated with marked hyperplasia of the overlying epidermis, mimicking carcinoma.

The clinical differential diagnosis in the head and neck include tuberculosis, actinomycosis or syphilis.

Different therapies are discussed: the first treatment of choice has been amphotericin B since 1957. Before the availability of amphotericin B the mortality rate was 89-93%<sup>7</sup>. The disadvantage is the longterm intravenous application. As an alternative, Ketoconazole, an oral agent can be used, but is associated with liver toxicity and interactions with other drugs. Nowadays Itraconazole, also an oral agent, is commonly used, because of the little side-effects. Therefore the oral application (longterm of 3 months) of Itraconazole is the therapy of choice.

## References

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