Case study: oral paracoccidioidomycosis with a long latency period

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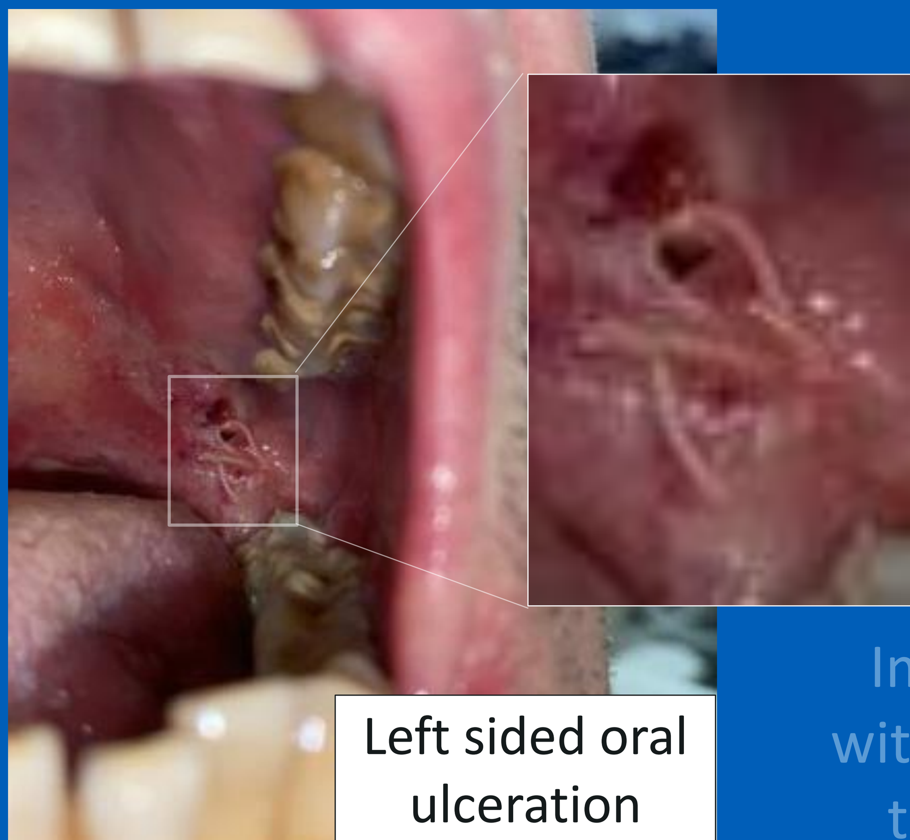
Initial presentation

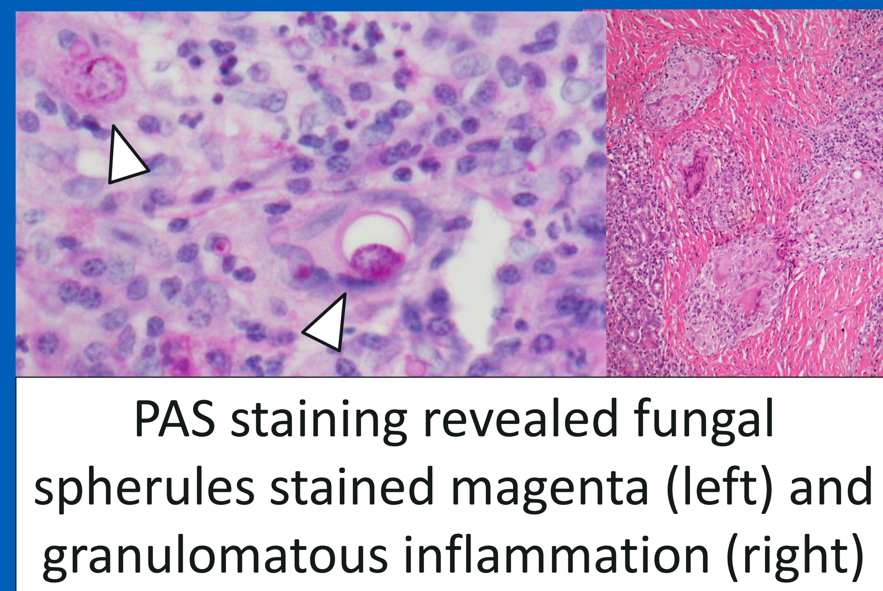
- A 49M presented to his dentist with a 1-year history of a slowly developing oral ulcer involving the left mandibular alveolus, loosening teeth and trismus
- Incisional biopsy of the ulcer and histological examination revealed non-necrotising granulomas with no evidence of neoplasia or fungal elements
- 4-5 month history of worsening breathlessness, cough and weight loss. Chest CT revealed bilateral ground glass changes and nodules
- A presumptive diagnosis of sarcoidosis was made, but no immunosuppressive therapy was commenced because he was relatively stable

1 year later

- Due to worsening symptoms, was seen by oral-maxillofacial surgeon who found the ulcer had enlarged further
- Repeat biopsy of the lesion revealed numerous budding yeast forms (without pseudohyphae), associated with granulomatous inflammation. 'Ship's wheel' structures seen in the sub-mucosa suggested *Paracoccidioides*
- The case was referred to infectious diseases at Wythenshawe Hospital (Manchester University NHS Foundation Trust)
- Travel history found that he was born in Portugal and then lived in Venezuela from ages 6-28, before moving to the UK
- Smoker; HIV negative

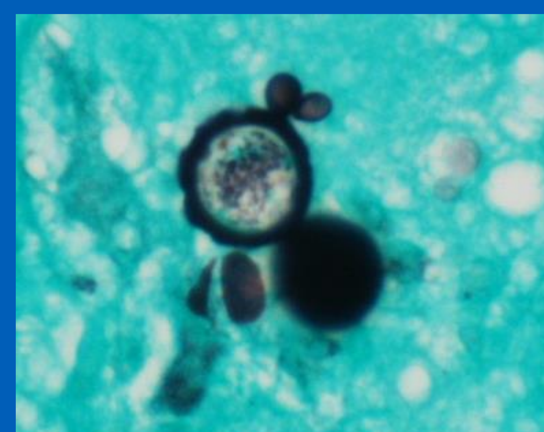


Left sided oral ulceration

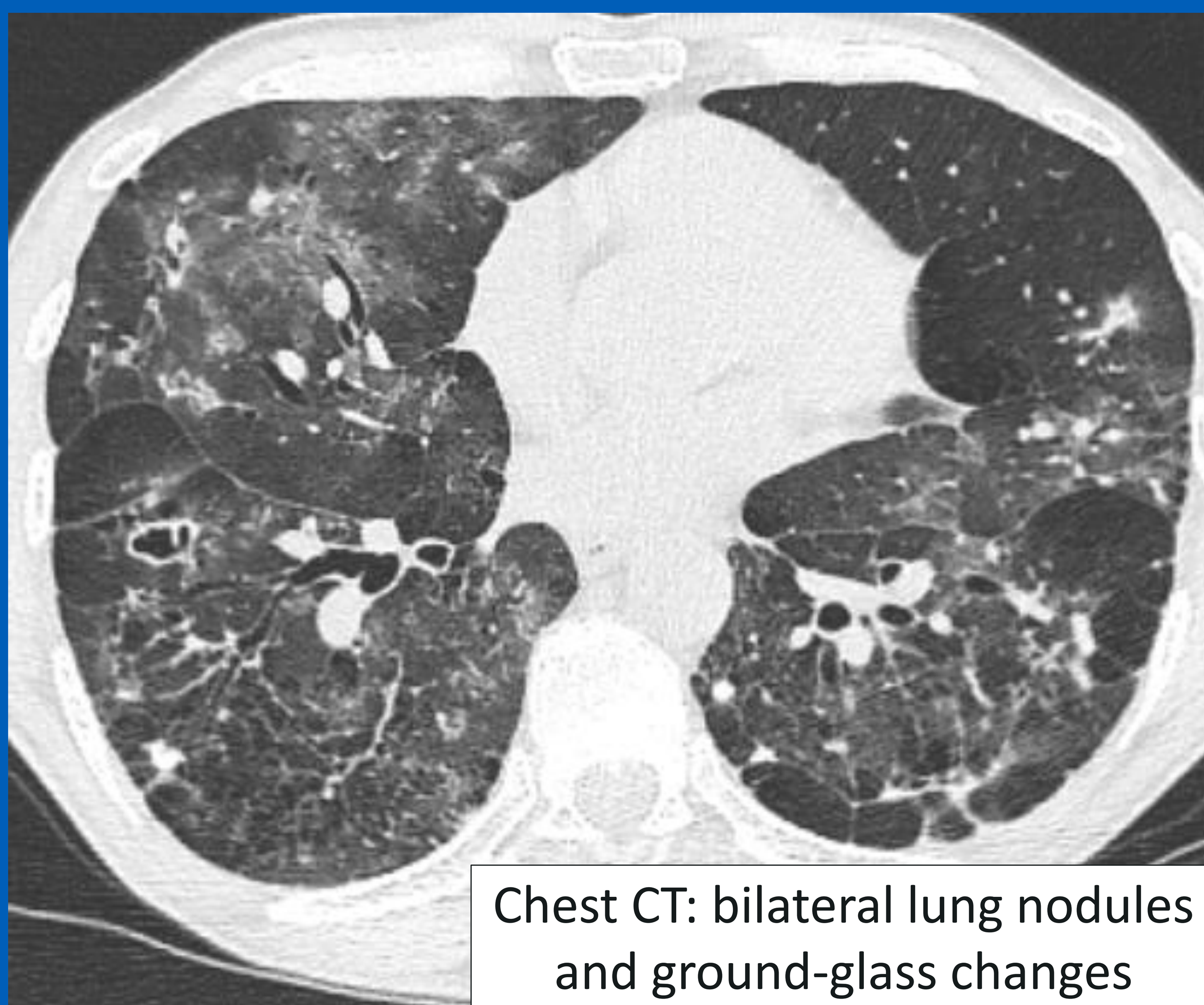


PAS staining revealed fungal spherules stained magenta (left) and granulomatous inflammation (right)

Images used with consent of the patient



Grocott staining showed budding yeast cells (below) and a partial 'ship's wheel' structure with multiple peripheral buds (above)



Chest CT: bilateral lung nodules and ground-glass changes

Test results

- Paracoccidioides serology = positive
- Panfungal PCR (oral tissue biopsy) = positive
- Sequencing = *Paracoccidioides brasiliensis*

Diagnosis

Oral paracoccidioidomycosis with probable pulmonary involvement

Treatment

- Oral itraconazole 300 mg daily
- Removal of several teeth
- After 6 weeks, repeat X-ray showed no change from previous film
- After 2 months, the oral ulceration had improved and he had gained 6kg. He reported less fatigue but remained short of breath
- He remains on intraconazole

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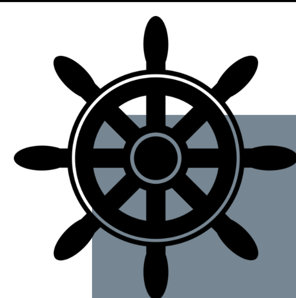

Paracoccidioides brasiliensis

This is a dimorphic (endemic) fungus with characteristic 'ship's wheel' structures. Following inhalation from soil, it causes a transient pulmonary infection which often becomes chronic; oral involvement is relatively common

Extended travel history

Dimorphic fungi are rarely seen in the UK, but are endemic to regions such as Central and Latin America, especially in rural areas. Asymptomatic pulmonary infections may reactivate years later, so a long travel history is essential

Biosafety level 3

Many dimorphic fungi require biosafety level 3 precautions in a specialist laboratory. Please include any relevant travel history and histopathology findings when submitting samples for fungal cultures

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