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CASE REPORT

Human cervicofacial mycetoma caused by *Streptomyces griseus*: First case report



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Streptomyces griseus causes subcutaneous mycetomas in felines and dolphins; however, human mycetoma caused by *S. griseus* has not previously been reported. Hereby, we report a case of a 50-year-old female presenting with swelling in the left upper cervical region and the left cheek that lasted for 6 months. The fine needle aspiration (FNA) performed on the swelling yielded purulent material; on microscopy, actinomycosis was diagnosed. On culturing, the pus grew *S. griseus*. To the best of our knowledge, this is the first reported case of human mycetoma caused by *S. griseus*.

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Introduction

Mycetoma is a localized chronic suppurative infection of subcutaneous tissue, commonly affecting the foot.¹ It is either actinomycotic or eumycotic in etiology. Actinomycotic mycetoma is most commonly caused by the genus *Actinomadura*, followed by *Nocardia* and *Streptomyces* species.² Among the genus *Streptomyces*, more than 3100 *Streptomyces* species have been described.¹ Of these, *S. somaliensis* has been the commonest etiologic agent causing human infections.³ It has also been identified as

one of the principal etiologic agents of actinomycetoma in South America, Africa, India, Mexico, Malaysia, and the USA.¹ As far as *S. griseus* is concerned, there is scanty literature available on human infection by this organism^{4,5} and no case of human mycetoma by *Streptomyces griseus* has been reported to date, to the best of our knowledge. We report a rare case of cervicofacial mycetoma caused by *S. griseus*.

Case report

A 50-year-old female presented with swelling in the left upper cervical region and left cheek for 6 months. History of suppuration in the swelling with pus exuding from multiple sites was present. There was history of dysphagia, but no history of dyspnea, dysphonia, or fever was present.

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On examination, there was a nodular swelling measuring 8 × 6 cm in the left upper cervical region extending to the left cheek (Fig. 1). The swelling was tender, hard in consistency, and fixed to the underlying tissues. It had tiny ulcerated areas with yellowish discoloration. Intra-oral examination revealed swelling in the floor of the mouth, which was red and indurated. Her hemogram and chest roentgenogram were found to be within normal limits. No other abnormality was detected on systemic examination. The fine needle aspiration (FNA) done from the left cervical swelling yielded purulent material, which was subjected to cytological and microbiological examination.

Cytologic findings

Smears were stained with May-Grunwald Giemsa (MGG), Hematoxylin and Eosin (H&E), Gomori's Methenamine Silver (GMS), Periodic-acid Schiff (PAS), Gram's, and Ziehl Neelsen (ZN) stainings. FNA smears showed mixed inflammatory infiltrate consisting of predominantly neutrophils, a few lymphocytes, macrophages, occasional plasma cells and eosinophils. In addition, numerous aggregates and colonies were seen interspersed within the infiltrate. The colonies were composed of slender and delicate filaments, which were appreciated under higher magnification. These filamentous organisms were PAS positive, Gram positive (Fig. 2), and were highlighted by GMS stains. The filaments were negative with ZN stain. A cytologic diagnosis of actinomycosis was made.

Microbiological findings

The pus aspirated was also sent for culture and sensitivity testing. The specimen was inoculated on blood agar and MacConkey's agar. The growth was visible as chalky white, membranous to granular colonies within 48 hours of incubation at 37°C. It was considered to be belonging to the genus *Streptomyces* and was sent to the Public Health Laboratory Services, Bristol, United Kingdom for confirmation, where it was finally identified to be *S. griseus*. Subsequent to the diagnosis, the patient was put on



Figure 1. Clinical presentation of the patient showing pus exuding from multiple sites.

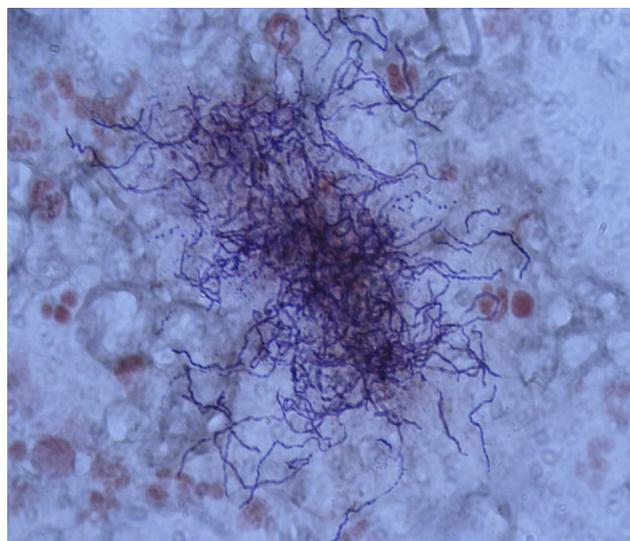


Figure 2. Smear showing aggregates of radiating filaments of *Streptomyces griseus* amidst acute inflammatory cells (Gram's stain, 400×).

intravenous crystalline penicillin. She responded well to treatment, and the lesion subsided within a period of 8 weeks.

Discussion

S. griseus is an aerobic actinomycete that commonly occurs in soil as a saprophyte. *S. griseus* was first isolated by Krainsky in 1914 from Russian soil during a World War I outbreak. In 1915, Waksman isolated a strain from New Jersey soil and called it *Actinomyces griseus*. In 1943, *Actinomyces griseus* was changed to *Streptomyces griseus*, and subsequently the common nomenclature *Streptomyces griseus* (Krainsky 1914) Waksman and Henrici 1948 followed for the organism.⁶ Traditionally, the organism has been known for production of antibiotics used to treat bacterial, mycobacterial, fungal, and parasitic infections.⁷ A review of the literature reveals only a few case reports of human infection by *S. griseus*. Gugnani et al⁵ reported five cases of pulmonary infection due to *S. griseus* on sputum culture. *S. griseus* was also isolated from a case of brain abscess.⁴ *S. griseus* is known to cause subcutaneous mycetomas in felines and captive bottlenose dolphins, and Gugnani et al tried to produce experimental mycetoma in laboratory mice.⁸ However, isolation of *S. griseus* as an etiologic agent in human mycetoma has not been documented to date.

Diagnosis of mycetoma is established on combination of clinical features, cytologic or histopathologic examination, serologic studies, and culture. The *Actinomycetales* include *Actinomycetes*, *Nocardia*, and *Streptomyces*. *Streptomyces* are differentiated from *Actinomycetes* by being aerobic and from *Nocardia* by being non-acid fast organisms that do not fragment into bacillary forms. Furthermore, species of the genus *Streptomyces* are characterized by formation of extensive branched aerial filaments with long chains of conidia and the cell wall made of peptidoglycan

L-diaminopimelic acid.¹ These features, along with culturing, help to differentiate *Streptomyces* from other actinomycetes, particularly *Nocardia* species. However, the identification and speciation of *Streptomyces* is a complex task because these organisms show marked variability and genetic instability. To date, culture remains the gold standard for final etiological diagnosis. On culturing, *Streptomyces* produces dry, chalky, gray-white colonies that emit pungent, musty odor.⁹

S. griseus was previously considered to be of doubtful pathogenicity.³ The reference laboratory of the Centers for Disease Control and Prevention (CDC) has reported incidence of *S. griseus* to be 7.7% of clinical isolates, following *Nocardia* and *Actinomyces* species.¹⁰ This data suggests that *Streptomyces* infection is under-reported and invasive *Streptomyces* infection may be more common. A high index of suspicion is necessary to identify these infections from clinical specimens. It is also stressed that samples should be investigated both cytologically and microbiologically. In the case described here, on cytological examination, an erroneous diagnosis of actinomycosis was made, but it was the growth from culture that helped in reaching the final diagnosis of *S. griseus*. It is also important to note that as such scant data is available on effective treatment of *Streptomyces* infection. Mycetoma caused by *Streptomyces* is often treated with penicillin, sulphonamides, or tetracycline but the cure rate may be low with sulphonamides (the drug of choice for *Nocardia*). Isolates of *S. griseus* referred to the CDC have been found to be resistant to ampicillin (80%), sulfamethoxazole (43%), cotrimoxazole (29%), and ciprofloxacin (57%).¹⁰ Resistance patterns in the case of *S. griseus* must be interpreted cautiously because it can synthesize antibiotics, potentially confounding the results of *in vitro* susceptibility testing. To the best of our knowledge, this is the first case report of mycetoma in humans as a result of *S. griseus* infection.

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