Chryseomonas luteola: an unusual clinical infection mimicking a mediastinal malignant lymphoma

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Abstract: Chryseomonas luteola is an infrequent human pathogen. We describe a case of mediastinal location showing Splendore–Hoeppli phenomenon in the abscess clinically mimicking a malignant lymphoma.

Keywords: Chryseomonas luteola, mediastinal lymphoma, botryomycosis, diagnosis

Introduction
Botryomycosis, initially described by Bollinger in 1870 in a pulmonary nodule in a horse, 1 is a chronic supplicative infectious disease characterized by the presence of eosinophilic fungus-like granules in abscesses, displaying Splendore–Hoeppli phenomenon and mimicking fungal infections. 2 Instead of fungi, these sulfur granules contain bacteria that may be highlighted by Gram or Giemsa staining. The pathogenesis of botryomycosis is not known, but it may be related to the dose and attenuate virulence of the bacteria, to an immune dysfunction or an altered inflammatory response caused by inadequate antibiotic therapy. 3 The more frequent agents causing botryomycosis are Staphylococcus aureus (40%) and Pseudomonas aeruginosa (20%); less frequently, other agents might be involved. 4, 5 Both cutaneous and visceral forms of botryomycosis are known; particularly, visceral diseases may cause serious differential diagnostic problems with malignancies. 6 All organs might be affected, and the disease has been described in the liver, kidney, brain, prostate, orbit, tongue, ear, bowel, and lung. We herein describe a striking thoracic botryomycosis mimicking mediastinal lymphoma in a young patient due to an uncommon bacterium.

Case report
A 16-year-old girl affected by autoimmune thrombocytopenia and on high-dose steroid therapy (average dose: 0.7 mg/kg/day for 1 year) presented with a 1-month history of fever and evidence on computed tomography scan of a mediastinal mass extending into the lung (Figure 1). Slight pericardial effusion was also found, but no investigations on the pericardial effusion were done. The patient had also leucopenia with lymphocytopenia: the whole presentation was considered very suspicious for a mediastinal lymphoma, more than for a germ cell tumor. The patient underwent surgery for histological diagnosis of the mass. Fresh samples were sent for intraoperative diagnosis to evaluate the adequacy of tissue for diagnosis: on frozen section, an abundant inflammatory infiltration of neutrophils was disclosed and a diagnosis
of mediastinal abscess was done as there was no evidence of neoplastic cells with the appearance of Reed–Sternberg cells or Hodgkin cells featuring a neutrophilic-rich variants of Hodgkin or anaplastic large cell lymphomas. Due to the intraoperative diagnosis, pathological tissue was sent fresh for microbiologic investigations also. On formalin-fixed and paraffin-embedded sections, histological examination revealed a diffuse granulomatous suppurative process showing the so-called Splendore–Hoeppli phenomenon: bundles of peculiar bacillary structures were frequently embedded in an eosinophilic substance, which formed a peripheral ring (Figure 2A). The microorganisms stained blue with Giemsa (Figure 2B), whereas the substance was strongly periodic acid-Schiff positive (Figure 2C). Microbiological cultures from the tissue yielded growth of lobated colonies, with a ‘fried egg’ morphology and a cheese-like texture, identified as Chryseomonas luteola. A final diagnosis of botryomycosis due to C. luteola was rendered. The patient immediately started treatment with meropenem 70 mg/kg tid and ciprofloxacin 500 mg bid for 6 weeks and rapidly recovered. The possible source of infection was not found: history revealed that the patient lived in the countryside in close contact with many domestic animals.

**Discussion**

*C. luteola* is an aerobic, motile, nonspore-forming gram-negative rod, ubiquitous in water and soil, which produces a characteristic yellow pigment. First described by Tatum and coworkers in 1974, it has been reported in few publications as the causing agent mainly of bloodstream infections associated with intravenous indwelling catheters, prosthetic valve endocarditis, foreign bodies, pancreatitis, and cutaneous abscesses. Rarely, nonbacteremic cases have been described as postneurosurgical infections, fatal meningitis, peritonitis complicating appendicitis or peritoneal dialysis catheters, femur abscess, subphrenic abscess, endophthalmitis, facial cellulitis, leg ulcer in a patient with sickle disease, and hand infection. The use of steroids, immunodepression, the presence of a foreign body, and postsurgical instability have been suggested to predispose to infection with *C. luteola*. Infections are therefore more frequently nosocomially acquired than community acquired. In our patient, no previous history of catheterism or surgery was present, but we think that previous steroid therapy for thrombocytopenia could have predisposed the infection. As in our case, the outcome of infection caused by *C. luteola* is usually good. The organism is generally resistant to first- and second-generation cephalosporins, but sensitive to third-generation cephalosporins, aminoglycosides, ureidopenicillins, and quinolones, with variable sensitivity to ampicillin and tetracycline.

Finally, we would point out that a pulmonary infection similar to that in humans is regularly found in ferrets (*Mustela putorius furo*), a small carnivorous animal that is becoming increasingly popular in Western countries as a pet animal. Although our patient did not keep such an animal, ferrets and squirrels are numerous in the area where the patient lived and we cannot exclude a possible contact with contaminated water or soil or with other animals. Animals can be a potential
source for uncommon infections, especially in contact with immunocompromised patients.

Disclosure
The authors report no conflict of interest in this work.

References