Unusual presentation of more common disease/injury

Iliopsoas coccidioidomycotic abscess with associated intra-abdominal extension in an immunocompetent patient

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Summary

Exposure to the fungal pathogen *Coccidioides immitis* in normal hosts causes primarily self-limited pulmonary disease. We report a case of an immunocompetent patient who developed a *Coccidioides*-associated iliopsoas abscess with rare intra-abdominal dissemination at least one decade after primary exposure in an area endemic for this fungus.

BACKGROUND

The dimorphic fungus *Coccidioides immitis* is endemic in dry arid regions of the Southwestern USA, Mexico and Central and South America. The fungus lives predominantly in sandy soil as mycelium which matures into arthrospores that are easily transmissible through dispersion in the air. Human exposure usually occurs by inhalation of air-borne spores but can also occur by their direct inoculation into the skin.¹ Germination of the spores in the pulmonary tree gives rise to spherules containing endospores that cyclically mature into new spherules. The growth of this parasitic phase in the lungs leads to a self-limited pulmonary syndrome commonly characterised by shortness of breath, dry cough and fever. Dissemination can occur to distant sites such as skin, joints, bone and meninges manifesting as fever, various types of rash, joint pain, myalgia and headache, the constellation of which is termed 'valley fever' or 'desert rheumatism'. Dissemination is more common in the immunosuppressed, such as those with HIV, solid-organ transplant recipients and those on steroids and other drugs that diminish cell-mediated responses. Although reported, dissemination to the abdominal cavity is relatively rare where it causes granulomatous inflammatory involvement of a variety of structures. Here, we report an unusual presentation in a previously healthy immunocompetent patient with a non-healing iliopsoas abscess who lived in an endemic area for this fungus 13 years prior to his disease presentation.

CASE PRESENTATION

The patient was a 36-year-old previously healthy Mexican man who was settled in the Bakersfield, California area where he worked as a manual labourer for approximately 4 years before moving to central Ohio. During this time describes a 2-month-long self-limited pulmonary illness characterised by fever, dry cough and pleuritic chest pain. No definitive diagnosis was made at that time and he was not treated with antimicrobials. After resolution, he remained completely asymptomatic for 13 years after

moving to central Ohio. Recently, he presented to a regional hospital with a 2-week history of dull right lower quadrant pain which was not associated with fever, diarrhoea or other systemic symptoms. CT revealed a right-sided iliopsoas fluid collection which was drained percutaneously with some symptom relief. Oral ciprofloxacin was started empirically. The patient continued to have significant pain and underwent open drainage and drain placement and was started on oral amoxicillin-clavulanate empirically. Cultures of fluid taken at this time grew light yeast but no bacteria. Despite drainage fluid reaccumulated 4-weeks later and he was transferred to our institution for re-evaluation. On presentation he was asymptomatic other than right lower quadrant abdominal pain. He reported ~20 lbs weight loss over the prior 2 months since his initial presentation.

INVESTIGATIONS

Routine blood work was within normal limits as was his chest x-ray. His HIV test was negative. His CD4 T-cell and total immunoglobulin A (IgA) and IgM levels were normal and his IgG was 3220 mg/dl (normal 700–1600). CT scan again revealed a moderately sized iliopsoas abscess (figure 1A). The patient underwent a second exploratory laparotomy and washout which revealed diffuse small bowel and omental studding. Omental tissue submitted for histopathological examination revealed exuberant necrotising and non-necrotising granulomata and with microabscess formation surrounding spherules of *C immitis* (figure 1B). Gomori methenamine silver stains showed several fungal organisms, morphologically also compatible with *C immitis* (not shown). Stains for acid fast organisms were negative. Fungal, bacterial and acid-fast cultures of the biopsy specimen were negative; however, fluid from the abscess grew moderate amounts of filamentous fungi identified as *C immitis*. Serum antibody testing specific for this fungus using both immunodiffusion and complement fixation methods were positive (CF titre 1 : 8).

Figure 1 Radiological and histopathological features of iliopsoas in coccidioidomycosis. (A) A selected coronal CT image showing two fluid collections with peripheral enhancement involving the right psoas and iliacus muscles (indicated with black arrows). (B) H&E-stained section from the omental biopsy showing a microabscess around remnants of a fungal spherule (black arrow) characteristic of *Coccidioides immitis* ($\times 10$ magnification).

TREATMENT

Treatment with liposomal amphotericin B was initiated and was transitioned to oral fluconazole (600 mg daily) after 5 days. The patient left the hospital asymptomatic and is currently stable on therapy. Repeat abdominal CT scan has revealed complete resolution of the intra-abdominal abscesses after 4 months of therapy.

DISCUSSION

Dissemination of coccidioidal infection from the lungs occurs in less than 5% of infected immunocompetent patients,^{2 3} the common sites being the skin, bone, joints and meninges. African-Americans, Hispanics and Filipinos are more prone to severe disease as are men and pregnant women. Immunosuppression is also an important risk factor with disease being more common and severe in those who use TNF- α blockers, steroids and in solid-organ transplant recipients or those with underlying hematological malignancy or HIV. Dissemination into the abdominal cavity is extremely rare, probably occurring in less than 0.05% of cases⁴ and dissemination as a consequence of primary pulmonary infection occurs after antifungal therapy.⁵ Involvement of the peritoneum, liver, appendix, intestine/omentum, biliary tract, ovaries and fallopian tubes has been reported.^{4 6-9} In these cases, only a small number had documentation of prior pulmonary infection, but most had a history of living in endemic regions at the time of diagnosis. The haematogenous spread of the fungus from the primary focus of infection is thought to be the predominant route of abdominal seeding, however paravertebral abscesses can extend into the iliopsoas.¹⁰ While this is a possibility in our case, there was no evidence of paravertebral disease in any of the radiological findings. In our case, the small bowel and omental involvement may have arisen from intraoperative seeding during the initial drainage procedures prior to his presentation to our institution. In some cases of intestinal infection acquisition, an oral route has been postulated.⁶

Our case is unique in two respects; (1) it is one of few describing the iliopsoas as a focus of infection and (2) it has a remarkable time lapse of over a decade between exposure opportunity with primary disease and the development of extrapulmonary disease. We presume that this patient acquired coccidioidal infection through the

pulmonary route followed by haematogenous spread to the iliopsoas. We presume that his intact immune system suppressed the growth of fungus from the time of initial infection giving rise to chronically smouldering disease until his abscess reached a critical size to cause symptoms. Initial concerns of peritoneal involvement were for tubercular disease or peritoneal carcinomatosis. The diagnosis of coccidioidomycosis is established by a variety of overlapping means, most often using histopathology with confirmatory serological testing and culture isolation where possible. Histopathological findings were critical to the diagnosis in this case by facilitating confirmation by serology and culture isolation. This patient's geographical history, however remote, was pertinent to interpretation. This case emphasises the importance of even a remote occupational or travel history in a geographic region endemic for this fungus.

Appropriate treatment of peritoneal coccidioidomycosis is not formalised because there have been very few cases described. Surgical excision or drainage of the affected area is sufficient for cure in some cases.¹¹ In a large number of cases this has been combined with antifungal chemotherapy using amphotericin B and high-dose fluconazole, as we used in our case. The length of therapy is not well defined but at least 1–2 years, and in some cases indefinite maintenance therapy is warranted.

Learning points

- ▶ Coccidioidomycosis is usually a self-limited pulmonary infection in immunocompetent patients but occasionally disseminates.
- ▶ Coccidioidomycosis rarely presents a decade or more after initial exposure with extrapulmonary involvement, including intra-abdominal foci.
- ▶ The diagnosis of *Coccidioides* infection requires a high index of suspicion and is made using combined histopathological, serological and microbiological methods.

Competing interests None.

Patient consent Obtained.

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