Blastomycosis presenting as multiple splenic abscesses: Case report and review of the literature

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A 31-year-old Canadian Aboriginal man from northwestern Ontario presented with left upper quadrant pain and a tender left upper quadrant mass. Evaluation with a computed tomography scan showed multiple lesions within the spleen, a collection between the splenic tip and splenic flexure of the colon, and several small adrenal lesions. Computed tomographic-guided needle biopsy showed necrotizing granulomatous inflammation and multinucleated giant cells. Gomori's methenamine silver stain showed broad-based budding yeast consistent with *Blastomyces dermatitidis*. Abdominal symptoms resolved after two months of oral itraconazole. Multiple splenic abscesses are a rare presentation of blastomycosis and should be considered in the differential diagnosis of left upper quadrant abdominal pain in a patient with a history of travel or residence in a region endemic for *B dermatitidis*.

Key Words: Abdominal pain; *Blastomyces dermatitidis*; Fungus; Spleen

Blastomycosis is an uncommon granulomatous infection caused by the thermally dimorphic fungus *Blastomyces dermatitidis*, which exists in the mycelial form in the soil of warm, moist, wooded areas that are rich in organic debris (1-6). Conidia are inhaled when the mycelia are disturbed. At body temperature, they convert to thick-walled budding yeast (7,8). Hematogenous dissemination is presumed to result in extrapulmonary blastomycosis. Blastomycosis presents most commonly with pulmonary disease but may also present with skin lesions, abscesses or osteomyelitis, which may lead to misdiagnosis or delay in diagnosis. The endemic area for blastomycosis includes the Ohio and Mississippi river basins, the regions that border the Great Lakes and northwestern Ontario (Figure 1) (9).

Blastomycosis may have a variety of different clinical manifestations (10), of which the pulmonary manifestations are the most frequent (11,12). There are a few reports of individuals presenting with splenomegaly and left upper quadrant discomfort as the initial manifestation, subsequently leading to a diagnosis of splenic abscess caused by *B dermatitidis* (13-17). We report a case of blastomycosis presenting with multiple splenic abscesses and review the previously published reports regarding this condition.

Une blastomycose prenant la forme de multiples abcès spléniques : Rapport de cas et analyse bibliographique

Un homme autochtone canadien de 31 ans du nord-ouest de l'Ontario a consulté en raison de douleurs au quadrant supérieur gauche et d'une masse sensible dans la même région. L'évaluation par tomodensitométrie a révélé de nombreuses lésions dans la rate, un épanchement entre le bout et l'angle du côlon et plusieurs petites lésions des surrénales. Une biopsie à l'aiguille orientée par tomodensitométrie a révélé une infection granulomateuse nécrosante et des cellules géantes multinucléées. La coloration à l'argent-méthénamine de Gomori a circonscrit un bourgeonnement de levure à large base compatible avec un *Blastomyces dermatitidis*. Les symptômes abdominaux ont disparu après un traitement de deux mois à l'itraconazole par voie orale. De multiples abcès spléniques constituent une présentation rare de la blastomycose et devraient être envisagés dans le diagnostic différentiel de douleur du quadrant supérieur gauche abdominal chez un patient ayant déjà voyagé ou habité dans une région où le *B dermatitidis* est endémique.

CASE PRESENTATION

A 31-year-old Canadian Aboriginal man from northwestern Ontario presented with a two-month history of left upper quadrant pain beginning in early spring. He denied cough, fevers, chills and weight loss. His medical history was negative for diabetes or immune system disorders. There was a history of alcohol abuse until six years before presentation. The patient worked as a carpenter in the winter and forest firefighter from May through September. Physical examination revealed a tender left upper quadrant mass. Chest radiography was normal. A computed tomographic (CT) scan showed multiple low-attenuation lesions within the spleen, a collection between the splenic tip and splenic flexure of the colon, and several small adrenal lesions (Figure 2). There were no signs or symptoms of adrenal insufficiency.

A tentative diagnosis of lymphoma was considered and the patient was referred to a surgeon, but he chose not to present for his evaluation for the next five months. He presented to the surgeon because the left upper quadrant pain had become severe during the two weeks before surgical evaluation and was associated with nausea and vomiting. Weight loss (18 kg) was noted, but there were no constitutional symptoms, respiratory

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Figure 1) Geographical regions endemic for blastomycosis (modified from references 12 and 34). USA United States of America

complaints or night sweats. The patient also noted a 2.5 cm palpable mass in the midline of the neck, present for the previous two weeks. The patient reported that a chest radiograph two months earlier was normal.

Although a repeat CT scan was requested, the patient did not attend for the study until four months later. At this time, the CT scan revealed persistence of the splenic and adrenal lesions. A percutaneous needle biopsy of the spleen showed necrotizing granulomatous inflammation and multinucleated giant cells. Gomori's methenamine silver stain showed broad-based budding yeast consistent with *B dermatitidis* (Figure 3). It was presumed that the neck mass was also due to blastomycosis.

The patient was treated with oral itraconazole (200 mg twice daily). He did not return for follow-up evaluation, but was successfully contacted six months after the biopsy. He stated that after two months of itraconazole, the left upper quadrant pain and the neck mass had completely resolved. He had independently discontinued the itraconazole after two months and did not return for further evaluation because of loss of insurance coverage as a result of marital separation. Further attempts to contact the patient were unsuccessful.

DISCUSSION

The current patient's presentation, with abdominal pain and splenic lesions, is an infrequently described manifestation of blastomycosis. He responded well to oral antifungal chemotherapy and did not require splenectomy.

All of the reported patients with splenic abscesses resulting from *B dermatitidis* had contact with geographical regions endemic for *B dermatitidis* (Table 1). The diagnosis of splenic abscess resulting from *B dermatitidis* in previously reported cases involved varied radiographic investigations, including radionuclide investigations (Table 1). More recently, CT has led to a decreased need for radionuclide studies, and CT-guided splenic aspiration has facilitated biopsy, as in the present case, obviating the need for surgical intervention.

Several conditions, such as sickle cell hemoglobinopathy, nonpenetrating abdominal trauma, gastrointestinal malignancy,

Figure 2) A contrast-enhanced computed axial tomographic scan of the abdomen revealing numerous tiny avascular low-density lesions that appear to be confluent

Figure 3) Microscopic evaluation of splenic biopsy tissue with Gomori's methenamine silver stain (original magnification $\times 400$) revealed broad-based budding yeast forms (diameter: 10 μm to 20 μm) diagnostic of *Blastomyces dermatitidis*. The Ziehl-Nielsen stain (not shown) did not reveal acid-fast bacilli

infectious endocarditis and injection drug use may predispose to splenic abscesses (18-26). Circulating microorganisms may be filtered by the spleen, resulting in solitary and multiple abscesses. The spectrum of microorganisms recovered from chronic splenic abscesses is broad, including *Staphylococcus aureus*, streptococci, Enterobacteriaceae including *Salmonella* species, organisms of chronic infection such as *Brucella* species (27), anaerobes (*Clostridium perfringens* and *Clostridium difficile*), and other Gram-negative bacteria such as *Burkholderia pseudomallei* (28-31). The microbiology of splenic abscesses compiled from case series before 1986 noted the following breakdown of pathogens recovered from splenic abscesses: sterile 28%, coliforms 23%, streptococci 22%, staphylococci 20%, *Salmonella* species 11%, anaerobic bacteria 5% and fungi 1% (14).

In early reports, chronic abscesses were reported to be either sterile or caused exclusively by aerobic bacteria, but the spectrum of microorganisms reported from chronic abscesses has changed, likely as a consequence of improved microbiological techniques for the recovery of anaerobes and fungi and a heightened awareness that parasites may be responsible for

TABLE 1
Cases of blastomycosis with splenic abscess

	Case (reference)				
	13,14	15	16	17	Current
Age, years	29	54	32	36	31
Sex	Male	Male	Male	Female	Male
Ethnicity					
Aboriginal	No	Yes	NR	NR	Yes
African American	Yes	Yes	NR	NR	No
Immunological risk factors	Injection drug abuse	Alcohol abuse	None	None	Alcohol abuse
Pre-existing pulmonary disease	TB, pneumonia	TB	NR	NR	None
Symptoms and signs*					
Constitutional	Yes	Yes	Yes	No	No
Weight loss (kg)	10	5	30	Yes (amount NR)	None initially; 18 kg 5 months later
Pulmonary	Yes	Yes	Yes	No	No
Abdominal	Yes	Yes	Yes	Yes	Yes
Musculoskeletal	No	Knee pain, effusion	No	No	No
Presumed place of acquisition	Alabama, USA	Ontario	Boston, USA/ Eastern Canada	Minnesota, USA	Northwestern Ontario
Presumed diagnosis	Not stated	Not stated	Hodgkin's disease	Not stated	Lymphoma
Diagnostic studies†					
Chest radiographic findings	Consolidation, pleural effusion	Pleural effusion	Normal	NR	Normal
Ultrasonography	Yes	Yes	Yes	Yes (endoscopic)	No
Abdominal CT scan	Yes	No	Yes	Yes	Yes
Radionuclide scan	Yes	No	No	No	No
Fine-needle aspiration	No	No	Yes	Yes	Yes
Management					
Splenectomy	Yes	Attempted‡	No	NR	No
Antifungal chemotherapy	Yes	Ketoconazole, 5 to 6 months	Yes	NR	Itraconazole, 2 months

*Constitutional: night sweats, fever, chills, malaise and/or fatigue; Pulmonary: thoracic pain, decreased air entry or cough; Abdominal: pain, tenderness, guarding and/or palpable splenomegaly; †Ultrasonographic findings included splenic enlargement, hyper- or hypoechogenicity, and cysts; Abdominal computed tomography (CT) scan findings included splenic enlargement and low-density lesions replacing parenchyma; Radionuclide scanning in one case included a gallium 67 scan (photopenia in spleen) and technetium 99m sulfur colloid scan (splenomegaly and photopenia); Fine-needle aspiration in both cases done was diagnostic of blastomycosis; ‡Laparotomy showed ascites; splenectomy could not be done because of dense adhesions. NR Not reported; TB Reactive tuberculin test

splenic abscesses (18-32). The apparent increase in reports of fungi, mainly *Candida* species isolated from splenic abscesses, may be a result of intensified chemotherapy and corticosteroid regimens for patients with neoplasms and inflammatory conditions requiring immunosuppression. Abscesses from parasites and other unusual microorganisms such as *Nocardia* species and mycobacteria have also been reported, attributed to immunosuppression, injection drug use and HIV infection (23).

The patient presented in the spring after having reported two months of symptoms. The time of presentation and clinical manifestations are compatible with previous studies reporting a seasonal variation of blastomycosis (33). Localized pneumonias typically present one to six months after initial exposure, with the presumed exposure occurring in the summer months. Four to nine months after the primary exposure, reactivation or slow progression of asymptomatic infection resulting in isolated extrapulmonary or disseminated hematogenous disease is observed. Our patient's presentation is compatible with these time lines (33).

Although rare, splenic abscess caused by *B dermatitidis* should be considered in an individual with left upper quadrant abdominal pain who has a history of travel or residence in a region endemic for *B dermatitidis*. With CT scanning, CT-guided biopsy, ultrasonography and antifungal chemotherapy, splenectomy may be avoided in some cases (14-15).

The present patient was lost to follow-up, but if possible, patients should be followed because of the potential for chronicity or recurrence.

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