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# **Case Report**

# Disseminated cat-scratch disease in an adult with selective IgA deficiency

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## ARTICLE INFO

Article history: Received 6 January 2016 Accepted 28 February 2016 Available online 8 April 2016

Keywords: Cat-scratch Bartonella IgA Post-therapeutic

#### ABSTRACT

A 51-year-old man with history of undiagnosed pulmonary nodules 4 years prior, presented with right-sided chest pain. Acute cardiac workup was negative, and a chest computed tomography examination demonstrated marked improvement in bilateral pulmonary nodules. A concordant abdominal computed tomography examination showed new subcentimeter hypodense lesions throughout the liver and spleen, mild progressive abdominopelvic lymphadenopathy, and new small lytic lesions of T11 and L4 vertebrae. A positron emission tomography examination demonstrated hypermetabolic activity of these abdominopelvic lesions suggesting metastatic disease. Extensive laboratory workup was negative, aside from IgA deficiency. Eventually, biopsy of a hepatic lesion was performed and compatible with Bartonella species. An elevated Bartonella IgG titer was noted, consistent with Bartonella Hensalae infection, or "cat-scratch disease." Radiographic findings showed marked improvement after clinically appropriate antibiotic therapy.

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## Case report

A 51-year-old man presented to the emergency room with right-sided chest pain. Initial workup, including electrocardiogram and troponins were negative for acute myocardial ischemia. Of note, the patient had a medical history of multiple pulmonary nodules 4 years before the current admission (Fig. 1A). Subsequent wedge resection during that admission demonstrated giant cell reaction and caseation necrosis. Acid fast and fungal pathologic workup were negative, and there was no evidence of

primary neoplasm or other metastatic disease. During the current admission, the computed tomography (CT) chest examination demonstrated resolution of the pulmonary nodules (Fig. 1B).

A concurrent abdomen and/or pelvis CT examination revealed multiple ill-defined hypoenhancing hepatic lesions (Fig. 2A), splenomegaly with hypoenchancing lesions (Fig. 2B), T11 lytic osseous lesion, and L4 lytic osseous lesion (Fig. 2C). A subsequent positron emission tomography scan demonstrated hypermetabolic activity within the hepatic lesions (Fig. 3A), splenic lesions (Fig. 3B), and osseous lytic lesions

Competing Interests: All the authors have declared that no competing interests exist.

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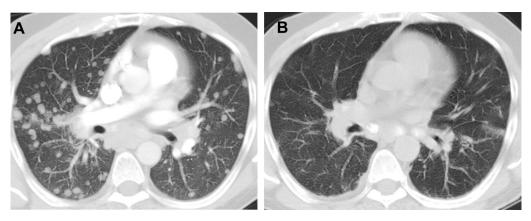


Fig. 1 - (A) Axial CT 4 years before current admission showed numerous bilateral noncalcified pulmonary nodules and (B) CT chest at the time of admission demonstrating interval resolution.

(Fig. 3C). Medical evaluation for lymphoma, metastatic disease, and atypical infection was performed. All pertinent laboratory workup, including but not limited to human immunodeficiency virus, aspergillus, acid-fast bacillus, fungal culture, antineutrophil cytoplasmic antibody, and perinuclear antineutrophil cytoplasmic antibody was negative. However, IgA deficiency with a value of less than 10 was discovered (normal range  $\geq$ 15).

A CT-guided liver biopsy of the largest hepatic lesion was performed and demonstrated granulomatous hepatitis with scattered non-necrotizing granulomas, associated granulation tissue and/or fibrosis, and scattered apoptotic hepatocytes. A special silver stain (Steiner) was positive for organisms morphologically compatible with Bartonella species. A concordant elevated Bartonella IgG >1:1024 was diagnostic for Bartonella Hensalae infection.

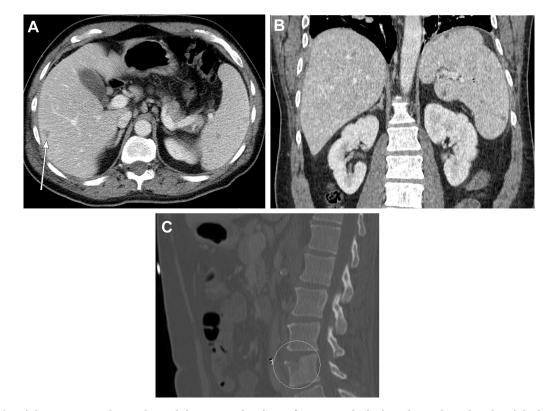


Fig. 2 – (A) Axial scontrast-enhanced CT abdomen at the time of current admission shows low-density right hepatic lobe lesion (arrow delineating low density hepatic lesion), (B) coronal contrast-enhanced CT imaging demonstrates mild splenomegaly with multiple low-density lesions, and (C) sagittal CT imaging demonstrates low attenuation lytic osseous lesions at L4 (circle encompassing L4 lytic osseous lesion).

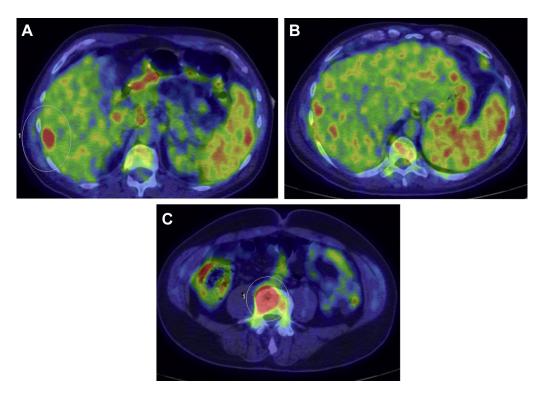


Fig. 3 – (A) Axial CT and/or positron emission tomography imaging at the current admission demonstrates increased metabolic activity at the nonenhancing right hepatic lobe lesion (circle encompassing metabolically active hepatic lesion), (B) axial CT and/or positron emission tomography imaging with increased metabolic activity throughout multiple nonenhancing hypodense splenic lesions, and (C) axial CT and/or positron emission tomography imaging demonstrates low attenuation lytic osseous lesion with hypermetabolic activity at L4 vertebral body (circle encompassing metabolically active lytic L4 lesion).

After diagnosis, the patient was placed on home intravenous therapy (Gentamicin) for 2 weeks, followed by oral Doxycycline 100 mg tablet twice a day for 4 weeks, providing 6 total weeks of antibiotic therapy. A follow-up abdomen and/or pelvis CT examination was obtained 8 weeks after discharge and showed improvement in hepatic lesions (Fig. 4A), splenomegaly and/or splenic lesions (Fig. 4B), and lytic osseous lesions (Fig. 4C). The patient was scheduled for follow-up care with infectious disease on an "as needed" basis for management.

# Discussion

Bartonella Hensalae infection, or "cat-scratch disease," is an unusual but well-documented infectious disease that typically affects children. The infection often presents with nonspecific clinical features, such as fever and lymphade-nopathy, but is typically self-limiting and asymptomatic [1,2]. However, systemic disease with hematologic and lymphatic dissemination may result in multiorgan system involvement. [3]. Radiologic manifestations are well described in pediatric patients, including involvement of lymph nodes, liver, spleen, osseous structures, and neural tissue but rarely documented in adult patients [4]. In some instances with widespread involvement of disease, antibiotic therapy may be warranted [3].

Cat-scratch disease in the adult is much less common and generally occurs in an immunocompromised patient. Infection rarely occurs in an immunocompetent host [5]. Immunocompromised patients generally have immunodeficiency virus or are intentionally immunosuppressed with adjuvant therapy for other systemic disease. Selective IgA deficiency is a relatively common immunoglobulin deficiency, with known predilection to frequent infections and exacerbations of common associated organ dysfunction, such as nephropathy [6,7]. However, chronic Bartonella Hensalae infection has not been previously identified in an adult patient with selective IgA deficiency.

This case is particularly unusual because of the patient's history of caseating pulmonary granulomas 4 years prior. Previous studies have demonstrated that cat-scratch disease can manifest as caseating pulmonary granulomas, but these are exceedingly rare on presentation [8]. We cannot definitively determine if this was a chronic Bartonella infection. However, with a negative workup for other granulomatous diseases, newly diagnosed IgA deficiency, biopsy-proven Bartonella, and elevated IgG titers, it is likely that cat-scratch disease was responsible for the original pulmonary granulomatous involvement.

Although clinical manifestations are nonspecific, the morphologic findings in pediatric patients are well known and

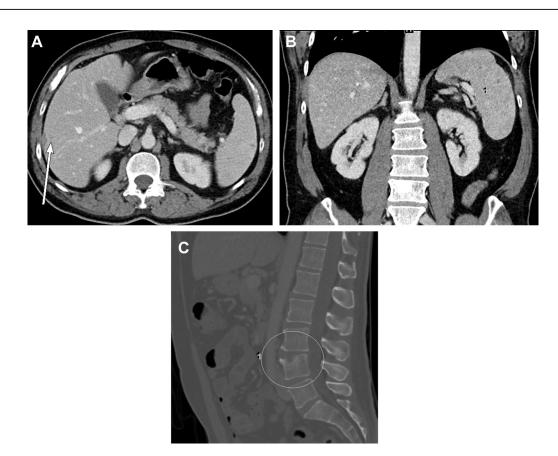


Fig. 4 — (A) Follow-up contrast-enhanced axial CT 8 weeks after admission demonstrates decreased size of low attenuation right hepatic lobe lesion (arrow denoting decreased size of hepatic lesion), (B) contrast-enhanced coronal CT demonstrates post-therapeutic improvement in splenomegaly with decrease in size of multiple low attenuation splenic lesions, and (C) Sagittal CT imaging demonstrates interval increased osseous incorporation with decreased size of osseous lytic lesion at L4 vertebral body (circle encompassing increased osseous incorporation of lytic L4 lesion).

often lead to correct diagnoses. Yet, because it is rare in adults, the diagnosis of cat-scratch disease is seldom considered. If the diagnosis is considered early in the medical evaluation, elevated IgG titers may allow a noninvasive diagnosis and appropriate therapy. As demonstrated in our case, radiographic improvement in systemic lesions can provide conclusive evidence of successful therapy.

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