CASE REPORT

Atypical manifestation of cat-scratch disease: isolated epigastric pain in an immunocompetent, 12-year-old child

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Key Clinical Message
We present a 12-year-old immunocompetent girl with hepato splenic cat-scratch disease (CSD). Her sole inaugural complaint was isolated epigastric pain. She fully recovered, with normalized abdominal CT scan following 2 weeks course of Azithromycin®. CSD should be included in differential diagnosis in children with epigastric pain, especially in those with domestic pets.

Keywords
Cat-scratch disease, epigastric pain, liver abscess, management, prognosis.

Introduction
Cat-Scratch Disease (CSD) is a known zoonosis that causes subacute lymphadenitis in children. In its typical form as encountered in around 90% cases, CSD often associates subacute lymphadenitis with low grade fever in children with positive history of cat contact and/or scratch. The disease is caused by the bacterium Bartonella henselae inoculated to humans following cat scratch or contact. It may also be the cause of prolonged fever of unknown origin.

In immunocompetent children, the clinical syndrome is usually a self-limiting disease which resolves spontaneously, without antibiotic therapy. With the increasing awareness of its clinical spectrum, several other less common manifestations of CSD such as osteomyelitis, meningococcal meningitis, optic neuritis, granuloma annulare, urticarial eruptions, anemia, endocarditis, pneumonia, and hepatosplenic abscesses have been reported [1–8].

We report a case of CSD with hepatic involvement which occurred in a child who complained of epigastric pain, as the only clinical manifestation at presentation.

Our aim is to emphasize that this atypical presentation should be considered and screening for CSD included in the work-up for children with epigastric pain, especially those having pets in their homes.

Case Report
A previously healthy 12-year-old girl, with unremarkable personal and familial medical history, was referred to us with a month history of epigastric pain that increased with deep breath, and was enhanced on abdominal palpation. She had received symptomatic treatment by her family doctor (Sodium Alginate and Phloroglucinol®), but this proved inefficient. Twenty days prior to her referral, blood test ordered by her family doctor had shown white blood cell count (WBC) of 6.3 Giga/L, mildly increased erythrocyte sedimentation rate (ESR) at 46 mm/h, and C-reactive protein (CRP) at 19.9 mg/L.

On admission she was afebrile and looked healthy. Her capillary refill time, heart rate, and blood pressure were normal. She reported no history of fever, weight loss, night sweats, fatigue, or recent pallor, but complained of
nonradiating epigastric pain which increased on abdomi-
 nal palpation, and on deep breath. The girl’s physical 
 examination revealed no regional lymphadenitis (axillary, 
 inguinal, or other), no hepatosplenomegaly, no scar from 
 cat scratches, although she admitted having four cats at 
 home and played often with them. Blood tests performed 
 on admission showed: WBC of 10.2 Giga/L without any 
 abnormal cells, ESR was accelerated (90 mm/h), and CRP 
 was increased (32 mg/L). Liver function tests were normal 
 with, alanine transferase level of 21 UI/L and aspartate 
 transferase of 11 UI/L, tuberculin test was not contribu-
 tive. The abdominal ultrasound (US) revealed a nonen-
 larged liver with diffuse homogenous, nodular hypoechoic 
 lesions throughout the organ, with the largest measuring 
 30 mm in diameter. These hepatic lesions were associated 
 with a mass of lymphadenopathy with heterogeneous 
 hypoechoic aspect of necrolysis, located in the hepatic 
 hilar and coelo-mesenteric region. The latter lesions were 
 infiltrating the celiac and the splenic arterial structures 
 (Fig. 1). An unenhanced abdominal CT scan was per-
 formed, it confirmed the abdominal US scan findings by 
 showing lesions that were hypodense, with some having 
 a central liquefied zone surrounded by an oedematous 
 halo. The CT scan also showed a significant mass of 
 lymphadenopathy in the coelo-mesenteric region, with 
 the largest, located in the hepatic and periportal hilum, 
 measuring 46 mm (Fig. 2). To confirm the diagnosis, a 
 serological test (FOCUS Diagnostic – IF, Laboratoires 
 Biomnis and IFI, VIRCELL, respectively) was performed 
 to search for bartonellosis. The results showed high titer 
 of IgG to *B. henselae* (1:4096) as well as increased IgM 
 antibodies against *B. henselae* (at level of 96). The 
 patient was started on a 2-week course of azithromycin 
 (250 mg twice daily). Clinical review, 12 days after dis-
 charge, showed a well-looking child whose sole com-
 plaint was very mild residual epigastric pain that she 
 quoted at 3 of 10 (vs. 8 of 10 on admission) on the 
 pain scale (Evendol pain Scale®, France), and was 
 slightly enhanced by deep abdominal palpation. The 
 patient was again reviewed three and half months after 
 hospitalization, her physical examination this time, was 
 unremarkable.

The serological follow-up showed a decrease in anti-
 Bartonella IgM antibodies titer (at 12), 2 months after 
 the first screening. Control abdominal US, 5 months after 
 the inaugural signs, showed a complete regression of 
 hepatic lesions along with, total disappearance of lymph-
 adenopathy in the hilar region (Fig. 3).

**Comments**

CSD is a common disease which affects persons of all 
 ages, and has a self-limiting course in a large majority of 
 patients. Its causative agent is *B. henselae*, a gram-negative 
 aerobic intracellular bacillus that is difficult to grow, mak-
 ing its routine culture of poor yield and not recommend-
Domestic cats are the main reservoirs of *B. henselae* so that transmission to humans results from close contact and/or a scratch from a carrier pet. The disease commonly presents as chronic lymphadenopathy associated with mild fever and sometimes, with a nontender papule at the inoculation site [2]. Fever above 38°C is present in one-third of patients; in over 50% cases, it is frequently associated with malaise, fatigue, headache, and anorexia. CSD can also be the cause of prolonged fever, or fever of unknown origin. With the increasing knowledge of its clinical spectrum, several other atypical manifestations such as Parinaud’s oculoglandular syndrome, endocarditis, urticarial eruptions, granuloma annulare, erythema nodosum, glomerulonephritis, haemolytic anemia, encephalopathy, osteomyelitis, thoracopulmonary, and hepatosplenic CSD have been reported in both immunocompetent adults and children [3–11]. These atypical manifestations reportedly affect 9–14% patients. In the classical hepatosplenic form of the disease, regional lymphadenitis and/or identifiable inoculation site are rarely present on clinical examination, but more than half of patients present with enlarged liver and/or enlarged spleen [1]. Over 60% patients with hepatosplenic CSD also have associated dull episodic abdominal pain which is localized in periumbilical region [11, 12]. Our patient, in addition to not having hepatosplenic enlargement, had only a clearly epigastric pain as sole complaint. Careful clinical history (contact with cats) oriented the diagnosis, which, was then aided by both abdominal US and CT scans, and confirmed by specific blood tests.

Presence, on abdominal US and CT-scans, of multiple hepatic granulomata evokes several other differential diagnoses that should be considered, among which: acute leukemia, lymphomas, hepatic hemangiomas, bacterial infections (Staphylococcal, Streptococcal, Salmonella, Nocardia, Yersinia, Listeria, Campylobacter, Fusobacterium, etc.), *Entamoeba histolytica*, *Echinococcus*, and mycobacterial infections [13]. Isolated epigastric pain without any other clinical signs and symptoms is common complaint in pediatric population, and can be misleading. A careful detailed clinical history taking along with clinical practice experience was, therefore, of tremendous contribution to the diagnosis. We emphasize that our patient had no fever or any other symptoms throughout the course of her illness.

Management of hepatosplenic Cat-Scratch disease (HS-CSD) has no consensus guidelines in the literature as most patients with the commonest forms of the disease recover spontaneously. Antibiotic treatment is discussed in those patients with atypical forms, especially in those with hepatosplenic involvement. Our patient totally recovered after having received a 2 weeks course of Azithromycin®, but other therapeutic modalities have also been reported in the literature [14]. The overall prognosis of this condition is favorable, as treatment is easily available and simple to administer. Based on our past experience of successful management of other forms of CSD in children with Azithromycin®, we tend, like Garcia et al. [11], to recommend this drug as first choice. The treatment duration remains, however, matter of debate with adult patients having been treated from 5 to 21 days as reported by Garcia et al. in their review [11]. Our practice is to offer 5 days course of Azithromycin® for typical cases and 2 weeks for the atypical forms, in the absence consensus guidelines.

The atypical presentation and course of this case highlight the importance of a well-recorded patient clinical history and evaluation. We warrant clinicians to include HS-CSD in the differential diagnosis in children with isolated epigastric pain, especially in those with positive history for household animals.
Conflict of Interest

All the authors have no conflict of interest to disclose.

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