

Unusual Presentation of Tuberculosis as a Splenic Mass

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DEDE, F., DOĞAN, E., DEMİR, M., ŞENER, D., KÖŞ, M., TAD, M. and ESKİOĞLU, E. *Unusual Presentation of Tuberculosis as a Splenic Mass.* Tohoku J. Exp. Med., 2006, **210** (1), 79-82 — Tuberculosis is an important health problem in developing countries, with varying clinical presentations depending on the organs/systems involved. Tuberculosis is mostly seen in immuno-compromised individuals, such as those with acquired immune deficiency syndrome or malignancies. Here we report a case of a spleen tuberculoma in a 29-year-old male patient with no known immune deficiency. He first presented with abdominal pain, and subsequent ultrasonographic examination revealed a splenic lesion of 10 cm in diameter. A computerized tomography scan of the abdomen confirmed the presence of a solitary, hypodense, septated cystic lesion. Lack of evidence supporting the presence of a splenic infection or a primary/metastatic malignancy prompted explorative surgery where a septated abscess formation was discovered and splenectomy was performed. Histopathological examination revealed granulomatous inflammatory changes with Langerhans-type giant cells, which are consistent with tuberculosis. For a period of two months, antituberculosis therapy with four drugs, isoniazid, rifampicin, pyrazinamide, and ethambutol, was carried out. Pyrazinamide and ethambutol were quitted at the end of two months. Therapy with isoniazid and rifampicin was planned for an additional 10 months. We would like to call attention to yet another atypical presentation of extrapulmonary tuberculosis. ——— spleen tuberculosis; abdominal tuberculosis; splenic abscess

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Tuberculosis (TB) is an affective disease on different age groups ranging from children to elderly (Ceylan and Gencer 2005). TB is known to influence different systems of the human body; in particular its common abdominal sites are the ileocaecal region, mesenteric lymph nodes, and the peritoneum (Dawson 1995; Bozer et al. 2001). With its insidious course and nonspecific symptoms, overt TB infection usually presents itself at a much later stage of the disease. The typical

symptoms are weight loss, fever, and fatigue. In the case of splenic involvement, left flank pain is characteristic (Winternitz 1912). The spleen is enlarged significantly and nodules may even be palpated (Jain et al. 1993)

CASE REPORT

A 29-year-old male patient presented to our clinic with widespread abdominal pain and distention of 6 months duration. He was a farmer with

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an unremarkable past medical history. He was of average height at 164 cm and weighing 57 kg. On physical examination, the patient had subicteric sclera and his body temperature was 36.6°C. Palpation of the abdomen revealed epigastric tenderness and a palpable spleen. Routine blood analysis revealed a hemoglobin level of 9.4 g/dl and an erythrocyte sedimentation rate of 64 mm/hr. Other biochemical and blood count parameters were within normal ranges. A subsequent abdominal ultrasound (US) demonstrated a cystic mass lesion (3 × 3 cm) in the upper pole of the spleen along with minimal ascites, and the patient was promptly hospitalized for further workup. Investigations into a possible etiology included C-reactive protein, 58.2 mg/l (0-5); Hepatitis C antibodies (-); Hepatitis B surface antigen (-); Toxoplasma IgM (-) and Toxoplasma IgG (-); Rubella IgM (-) and Rubella IgG (+); Cytomegalovirus (CMV) IgM (-), CMV IgG (+); and Wright agglutination (-). Levels of serum immunoglobulin (Ig) G, IgM, and IgA antibodies were normal. Serum iron, folate, and vitamin B12 levels were also normal. Total iron binding capacity was not elevated, and ferritin levels were 374.6 ng/ml (normal 24-336.2). Antinuclear antibodies and rheumatoid factor were negative (-), and serum α -fetoprotein, carcinoembryonic antigen, and CA 19-9 levels were normal. CA 125 and CA 15-3 were mildly elevated at 36.61 U/ml (0-35) and 34.45 U/ml (0-25), respectively. Serum Echinococcus granulosus IgE and its indirect hemagglutination were both negative. There was no paranchymal infiltration on a chest roentgenogram. A second abdominal US showed a further enlarged, thick walled, centrally necrotic mass lesion (6 × 10 cm) at the upper pole of the spleen, surpassing the rib margin by 2 cm. An abdominal computerized tomography (CT) scan (Fig. 1) revealed hepatomegaly (max. width 17 cm) and splenomegaly (max. width 13 cm), also confirming the presence of a septated, hypodense, cystic lesion (10 × 11 × 10 cm) on the anterior aspect of the upper pole of the spleen.

Enduration after a tuberculosis skin test was 8 × 6 mm and sputum specimens were thrice negative for acid resistant bacteria. Colonoscopy and

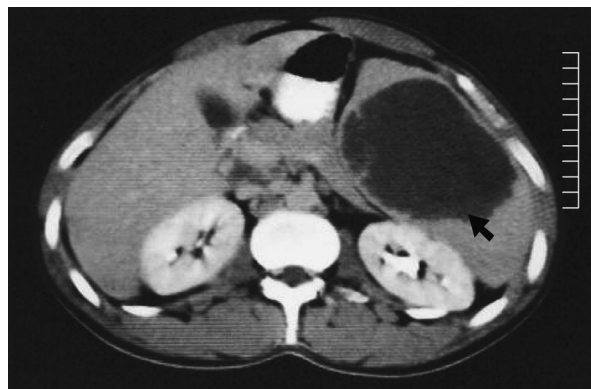


Fig. 1. Abdominal CT scan before treatment. Abdominal CT scan revealed a single, hypodense, 10 × 11 × 10 cm in size lesion in the spleen (Shown with small black arrow).

upper gastrointestinal endoscopy were normal. There were no signs of abnormal cell infiltration on bone marrow aspiration, and bone marrow biopsy was normal. As the patient did not have any lymph nodes appropriate for excision, diagnostic laparotomy was performed. Macroscopically, there were widespread adhesions and peritoneal deposits within the abdominal cavity, with a markedly enlarged (approximately 20 cm diameter) spleen. Splenectomy was performed, and multiple biopsies were obtained from the right and left lobes of the liver. Sampling was also performed on the lymph nodes from the peritoneum, hepatoduodenal ligament, and the hepatogastric ligament. Histopathological examination of these samples revealed granulomatous inflammation of the parietal peritoneum, liver, and spleen, with signs of caseous necrosis in the latter two. Histological examination of the spleen lesion biopsy showed Langerhans-type giant cells (Fig. 2) within wide caseous necrosis field, and inflammatory cell infiltration. Similarly, on examination of hepatoduodenal lymph nodes, findings consistent with caseous granulomatous lymphadenitis were observed, with reactive changes in the paraaortic lymph nodes. The findings described above prompted the initiation of antituberculosis therapy with four drugs; isoniazid (300 mg once daily), rifampicin (600 mg once daily), pyrazinamide (1,000 mg twice daily), and ethambutol (1,500 mg once daily) for a period of

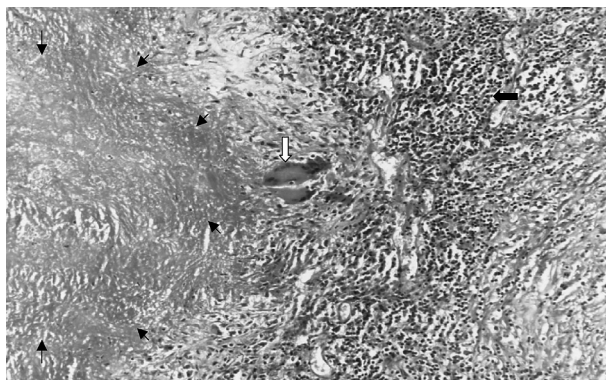


Fig. 2. Histological appearance of the hypodense lesion extracted from spleen after laparotomy. Histological examination of this lesion biopsy shows Langerhans-type giant cells (marked with white arrow) within wide caseous necrosis field (bordered with little black arrows), and inflammatory cell infiltration (marked with big black arrow) (Hematoxylin and eosin, original magnification $\times 100$).

two months. Pyrazinamide and ethambutol were quit at the end of two months. Therapy with isoniazid (300 mg once daily) and rifampicin (600 mg once daily) was planned for an additional 10 months.

DISCUSSION

TB of the spleen develops as a result of dissemination of advanced pulmonary or miliary disease, following either ingestion of contaminated food or infected sputum, or by dissemination via the portal vein (Dawson 1995; Akhan and Pringot 2002; Gulati et al. 1999; Batra et al. 2000; Ho et al. 2000). Especially in developed countries TB is an issue seen in cases with immune deficiencies depending on human immunodeficiency virus (HIV) infection, malignancies, primarily lymphoma (Ho et al. 2000; Neki et al. 2001), and in developing countries increasing drug resistance and late diagnosis make this disease an important health problem with significant mortality and morbidity (Bozer et al. 2001).

Splenic involvement is rare and is usually associated with miliary dissemination, and involvement of the liver is quite frequent (Jain et al. 1993; Gulati et al. 1999; Ho et al. 2000). Hepatosplenic involvement can be seen as micro-

or macronodular forms (Akhan et al. 2002). In a series evaluated retrospectively, the rate of splenic tuberculosis was reported as 8% (Batra et al. 2000), while in the literature the rate of micronodular type of involvement was reported around 5% (Sheen-Chen et al. 1995). In autopsies of cases with disseminated pulmonary tuberculosis, hepatosplenic involvement was reported to be as high as 80-100% (Thoeni and Margulis 1979). Macronodular involvement is extremely rare and is more frequent in HIV positive patients with a rate of around 15% (Monilla-Serra et al. 1997). The differentiation of macronodular involvement in the form of localized tumoral mass from other solid masses of the spleen is extremely difficult (Akhan et al. 2002; Sheen-Chen et al. 1995). In the differential diagnosis of solitary splenic masses, splenic cysts, hematomas, some fungal infections, pyogenic and amebic abscess infarcts, Crohn's disease, vascular tumors, lymphoma, and metastatic tumors should be taken into consideration (Thoeni et al. 1979; Solbiati et al. 1983; Batra et al. 2000). In our case, the single and solid mass seen on US and CT, at first reminded us of malignant etiologies, primarily lymphoma.

On US, the micronodular involvement usually reveals itself as diffuse hyperechogenicity (hypoechoic if necrotizing), but may also result in a homogenous or heterogeneous appearance, without the presence of organomegaly (Akhan et al. 2002). On CT, splenic lesions typically appear as multiple, rounded, hypodense, uncalcified masses with diameters around 1-2 cm. These findings, however, are non-specific since they may also be associated with pyogenic spleen abscesses and lymphoma (Gulati et al. 1999; Ho et al. 2000).

Being cheap and reliable, US remains the method of choice for evaluating abdominal TB (Batra et al. 2000). On the other hand, CT is ideal for establishing the degree of disease, evaluating the complications, and for follow up (Dawson 1995; Gulati et al. 1999). Needle aspiration is reliable when performed in the assistance of CT and US and smears frequently reveal positive results in terms of diagnosis (Solbiati et al. 1983; Ho et al. 2000). Percutaneous biopsy is frequent-

ly required for confirming the diagnosis of splenic involvement (Akhan et al. 2002), however, definite diagnosis of abdominal tuberculosis may still require minilaparotomy or laparoscopy (Dawson 1995; Bozer et al. 2001).

Though still in debate, antituberculosis therapy of 12 months is deemed appropriate despite the possibility that most patients with splenic TB have a disseminated disease (Bass et al. 1994). However, long-term treatment may still be warranted in immunocompromised individuals, particularly those with HIV (Dawson 1995).

In our case, no immunosuppressive condition that could cause such infection was demonstrated. This type of TB cases seen in developed countries primarily requires thorough investigation of underlying etiologies like HIV and lymphoma. Clinical and routine laboratory findings are non-specific and obscure, despite the usual reliability of common methods like US and CT in distinguishing such lesions, from primary or metastatic malignancies of the spleen. Since both these entities have a varying morbidity and mortality, and because of the obviously different management protocols involved, surgery remains the gold-standard for definitive diagnosis of such cases with an undefined etiology. When the prevalence of TB in Turkey and patients with varying presentations are taken into consideration, we believe that more detailed information about the differential diagnosis, with particular attention to non-invasive imaging techniques should constantly be updated.

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