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CASE REPORT

A case of abdominal mesothelioma diagnosed by indium-111 leucocyte scintigraphy

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ABSTRACT

We present a case of peritoneal mesothelioma that presented with fever of unknown origin and an elevation in the inflammatory parameters. Radiological imaging did not reveal a diagnosis. Because of tumour-associated inflammatory activity, indium-111 leucocyte scintigraphy enabled us to establish a diagnosis. To our knowledge, the use of indium-111 leucocyte scintigraphy in peritoneal mesothelioma has not been reported previously.

KEYWORDS

Peritoneal mesothelioma, indium-111 leucocyte scintigraphy

CASE REPORT

A 40-year-old man was referred to our outpatient clinic due to fever and weight loss. Because of a productive cough, he was first analysed by a pulmonologist. Chest radiograph, ECG, Mantoux and spirometry were normal. History revealed a fever of up to 39.5°C during the last three months and severe night sweats. He complained of fatigue, thirst, loss of appetite and a weight loss of 12 kg in two months. Other symptoms were nausea and vomiting in the morning. His stools were normal. Medical history was unremarkable. He stopped smoking 20 years ago. His alcohol intake was 0 to 1 unit a day. There was no history of drug abuse or exposure to toxic agents, including asbestos. He was taking 500 mg of paracetamol three to four times a day to suppress his fever. His father and grandfather died of lung cancer. His occupation was forklift truck driver. Physical examination showed a blood pressure of 120/80 mmHg, a regular pulse of 95 beats/min and a temperature of 38.7°C. Enlarged lymph nodes were not detectable. Physical examination of the heart, lungs and abdomen was normal.

Routine laboratory tests revealed a microcytic anaemia (haemoglobin 6.1 mmol/l) and a remarkable elevation of inflammatory markers (C-reactive protein 219 mg/l, erythrocyte sedimentation rate (ESR) 77 mm/h, leucocytes 15.9 x 10^9/l, granulocytes 95% and thrombocytes 663 x 10^9/l). Urinalysis, including cultures on tuberculosis, was negative. Further diagnostics were aimed at infections, autoimmune diseases and malignancy, particularly malignant lymphoma. High-resolution computed tomography (HRCT) scanning of the thorax, ultrasound and a CT scan of the abdomen were reported normal.

The patient was admitted to hospital for further analysis. Repeated cultures of blood, urine and sputum were sterile. Colonoscopy was performed and showed no intraluminal pathology. Echocardiography revealed no endocarditis. Bone marrow investigation was normal. Finally, the case fitted the classical criteria of fever of unknown origin.[1]

In a further search for a possible inflammatory origin of his symptoms, indium-111 leucocyte scintigraphy was performed. Autologous leucocytes were labelled *in-vitro* with the radioactive isotope indium-111 and subsequently reinjected into the patient. Accumulation of leucocytes may occur in infectious or noninfectious inflammatory lesions. Images were obtained with single photon emission computed tomography (SPECT) scintigraphy and showed several hotspots in the upper abdomen. A repeated CT scan 3.5 weeks after the first CT scan clearly showed intra-abdominal fluid and extensive masses in the mesenterium and omentum (*figure 1A*). Comparison of the SPECT scan and the CT scan (*figure 1B*) revealed that the hotspots on the SPECT scan corresponded to the intra-abdominal masses on CT. CT-guided biopsy of one of the tumours established the diagnosis of malignant mesothelioma of the epithelial type.

Figure 1A. *CT scan of the abdomen showing widespread tumoural changes and thickening of the small intestine and mesenterium and ascites*

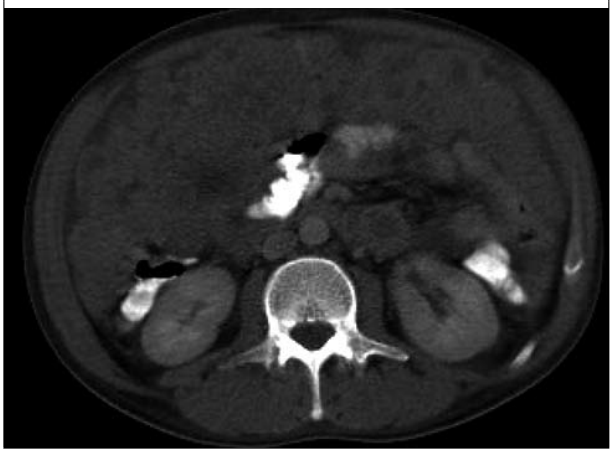
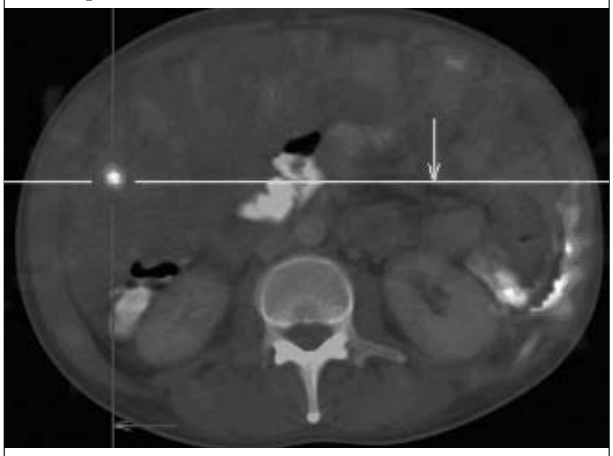


Figure 1B. *Indium-111 leucocyte scintigraphy superimposed on CT scan showing focal areas of leucocyte accumulation in the abdomen*



The right lateral activity is physiological spleen uptake.

Since there is no evidence-based therapeutic regimen for intra-abdominal mesothelioma, the patient was treated with palliative chemotherapy according to the guidelines for pulmonary mesothelioma, using pemetrexed. After two courses, the disease appeared rapidly progressive, leading to death within ten weeks after the first presentation. Autopsy revealed massive tumour involvement of the peritoneum, without pleural localisation of mesothelioma.

DISCUSSION

Mesothelial cells form a monolayer along the abdominal and pleural cavities and on internal organs. Neoplastic transformation gives rise to mesothelioma, a highly malignant tumour, most commonly localised in the pleura. Abdominal mesothelioma, the only primary peritoneal malignancy, is extremely rare (incidence one/million in the USA).[2] Although 90% of mesotheliomas are associated with exposure to asbestos, cases have been reported in which no exposure has been identified.[3] The commonly accepted mechanism leading to an intra-abdominal localisation is inhalation of asbestos fibres, followed by clearance of those fibres by the mucociliary system of the bronchi. Mucus containing asbestos fibres is coughed up and swallowed. Subsequently, passage of the sharp pieces through the intestinal wall may lead to a peritoneal localisation. A latency period of 20 to 40 years is characteristic.[4]

A presentation with nonspecific and mild symptoms despite an advanced disease state is common. Abdominal pain, fever, a slightly raised ESR or a marked thrombocytosis may be the only presenting signs. In a more advanced stage, fever and raised inflammatory parameters are more common. Less frequently bowel obstruction occurs. Because of the nonspecificity of symptoms, diagnosis often relies on imaging. The most common radiological findings of peritoneal mesothelioma are ascites, irregular or nodular peritoneal or intestinal mucosal thickening and omental or mesentery involvement. The nonspecificity and variety of tumour morphology, site and mode of spread within the abdomen explain the diagnostic challenge of peritoneal carcinomatosis.[5,6]

The presence of high fever of unknown origin (FUO) with night sweats and raised inflammatory parameters suggested an infectious or inflammatory disease. De Kleijn *et al.* described a series of 167 patients with FUO. In this study 26% suffered an infection, 13% a neoplasm (0% mesothelioma) and 24% a noninfectious inflammatory disease.[7] In 30% no diagnosis could be established. In this case, the absence of localising signs and symptoms and abnormalities on radiological imaging delayed the diagnosis. Scintigraphic diagnostics aimed at an inflammatory disease finally revealed the diagnosis. Although the patient suffered from inflammatory symptoms during the course of the disease, hardly any literature on primary peritoneal mesothelioma and FUO is found. However, peritoneal mesothelioma may present as a focal or generalised inflammatory disease and may mimic the signs of acute appendicitis, cholecystitis, incarcerated umbilical hernia or inflammatory bowel disease.[8] Although fluorodeoxyglucose-positron emission tomography (FDG-PET) scans are routinely applied for the diagnosis, staging and follow-up of mesothelioma,[9] this is the first report showing the use of indium-111 leucocyte scintigraphy for this diagnosis.

CONCLUSION

Scintigraphic imaging, in particular FDG-PET, may be useful for the diagnosis of peritoneal mesothelioma, especially when radiological imaging does not reveal a diagnosis. Positivity of indium-111 leucocyte scintigraphy in mesothelioma has not been reported before. The overt clinical signs of inflammation and the accumulation of radioactive-labelled leucocytes in the tumour mass show a tumour-associated inflammatory activity.

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