

Incidental finding of a malignant peritoneal mesothelioma in an inguinal hernia sac: Report of a case

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Summary

We report the case of a 46-year-old female who underwent a routine operation of herniorrhaphy for a right inguinal hernia. Physical findings revealed a moveable tender mass in the right lower inguinal area. Pathological examination revealed a mass with multiple nodules scattered in the inguinal hernia sac. The histological features of these nodules were compatible with malignant peritoneal mesothelioma (MPM). The incidental finding of MPM in the inguinal hernia sac is very rare. The incidence of this neoplasm is 2-2.6 cases per million annually, and the prognosis is very poor.

Key words: Malignant peritoneal mesothelioma; Inguinal hernia.

Introduction

Peritoneal mesothelioma is a rare, benign or malignant primary tumor, arising from the peritoneal membrane. The most frequent histological form is the malignant one with an incidence of 2-2.6 new cases per million per year [1]. In most cases it appears in individuals past 40 years of age and a definite male predominance has been noted. Abdominal pain with little or no ascites is the most common symptom, with increased abdominal girth as a second. Abdominal girth and pain and a new onset hernia are less frequent [2, 3]. The prognosis for malignant peritoneal mesothelioma (MPM) is extremely poor. A more favorable prognosis occurs in women with a small disease volume [2]. We report here the clinicopathological features of an incidental finding of a MPM in an inguinal hernia sac.

Case Report

A 46-year-old female had a progressively enlarging firm mass in her right lower inguinal area two years prior to admission. The lesion was painless, but moveable and disappeared into the abdominal cavity. Physical findings in this woman revealed a moveable tender mass in the right lower inguinal area, which was also moveable with cough or Valsalva manoeuvre.

Ascites or an abdominal mass was not observed. X-rays of the chest and routine blood analysis were normal. The gynecological history of the patient was also free from disease. The clinical diagnosis was non-complicated inguinal hernia. Finally, the patient was scheduled for surgery. During the operation, a tumor mass was observed in the right inguinal hernia sac.

Pathological examination revealed a specimen of a 9 cm mass in the inguinal hernia sac with multiple nodules 0.5-1 cm (Figure 1). Microscopically these nodules contained tumor cells which were arranged in papillae and tubules or in small clusters and were divided by fibrous septae into fibrous compartments (Figure 2). The atypical tumor cells were generally uniform in

size and polygonal or oval in shape, with pink granular cytoplasm. The nuclei were bigger than normal cells, oval in shape with occasionally prominent nucleoli. A few mitoses were found and invasion of fat was observed. Immunohistochemical stains were: keratin 5/6 (+), EMA (+), calretinin (+) (Figure 3), mesothelin (+), thrombomodulin (+), vimentin (+), keratin 7 (-), CAM 5.2 (-), AE1/AE3 (-).

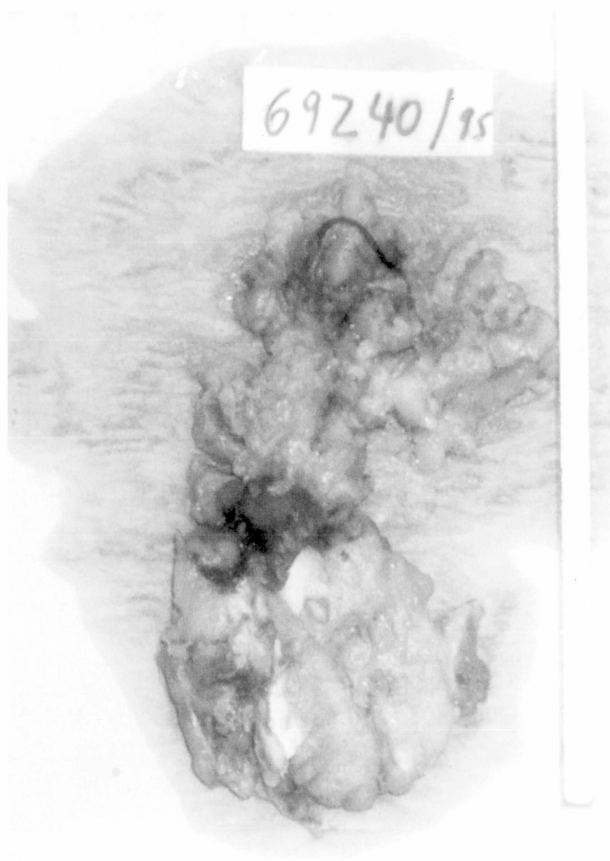


Figure 1. — Macroscopic image of inguinal hernia sac content.

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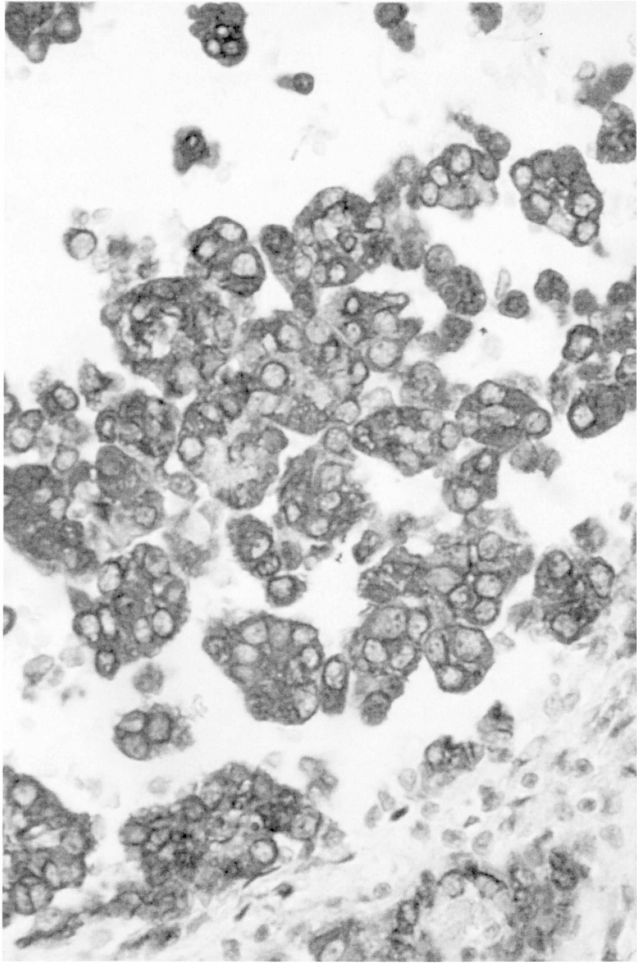


Fig. 2

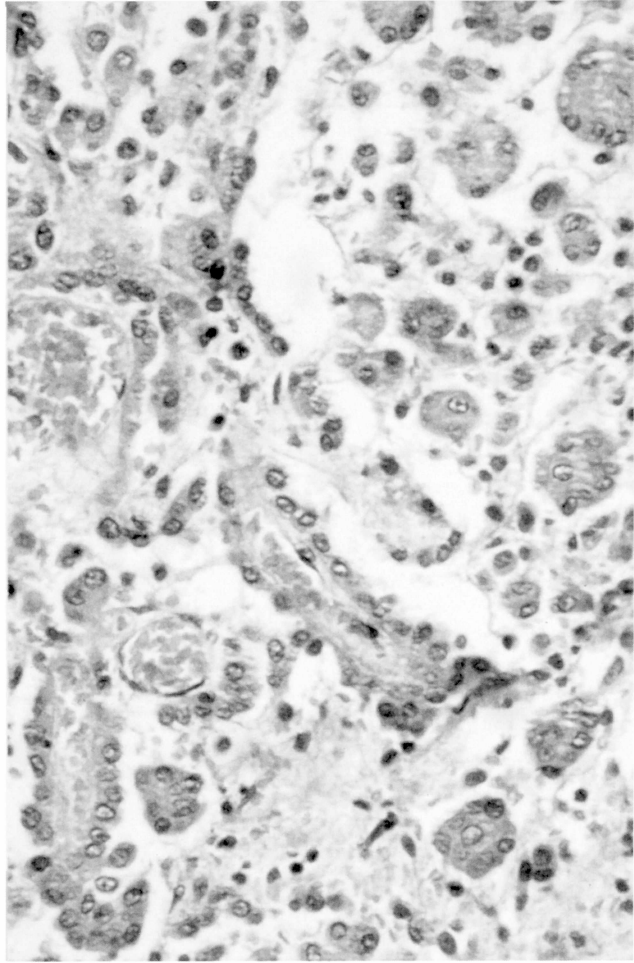


Fig. 3

Figure 2. — Histologic image of mass consisting of papillae with atypical tumor cells (hematoxylin and eosin x16).

Figure 3. — Immunohistochemical stain for calretinin in cytoplasm of tumor cells.

The differential diagnosis for this neoplasm included: nodular mesothelial hyperplasia, MPM, ovarian or extraovarian serous papillary carcinoma and metastatic adenocarcinoma. The clinicopathological features, as mentioned above, helped us identify this tumor as a MPM. Further investigation of the patient (CT scanning, US) showed the uterus and atrophic ovaries, without any evidence of neoplastic disease. Moreover, no exposure to asbestos was reported after additional clinical history investigation.

Discussion

The incidental finding of a MPM in the inguinal hernia sac is very rare. The first case observed – to our knowledge – was in 1976 [4]. The mesothelial lining surface has a great capacity to undergo florid hyperplastic changes when irritated by chronic inflammatory reactions. Hernia sacs exhibit florid foci of nodular mesothelial hyperplasia, following incarceration or some other mechanical insult; this is particularly common in children and may simulate malignancy [5]. The mesothelial cells

may show moderate mitotic activity and nuclear pleomorphism and hence mimic a neoplastic process [6].

As mentioned above, MPM occurs in individuals past 40 years of age with male predominance. Also, the association with asbestos exposure is common [7]. The most frequent clinical presentation, according to Acherman *et al.* [2], is pain in 33% of patients, increased abdominal girth in 31%, and increased hernia in 12%. However in the case of inguinal hernia the symptomatology is insidious and poses difficult problems in diagnosis and treatment [1].

The pathologic examination gives an accurate diagnosis. Grossly, it usually appears as multiple plaques or nodules scattered over the visceral and parietal peritoneum [8]. On rare occasions, the tumor presents as an isolated mass. Such a lesion is distinguished from benign mesothelioma by virtue of its more solid appearance and the presence of atypia, which may be very subtle [9].

The most common architectural pattern of growth is that of papillae or tubules lined by atypical mesothelial cells [10]. The cells are uniform with acidophilic or vacuolated cytoplasm and large vesicular or hyperchromatic

nuclei. Mitoses are occasionally observed. Immunohistochemically, the cells of MPM are positive for keratin (5/6), EMA, calretinin, mesothelin, the WT-1 gene product and thrombomodulin [11, 12].

The differential diagnosis includes reactive mesothelial hyperplasia, adenocarcinoma (primary or metastatic) and serous papillary carcinoma of müllerian type (ovary or uterus). The distinction is based on morphologic features. The multiple plaques or nodules with highly atypical and mitotically active cells raise the possibility of malignancy. The most important distinguishing criterion is invasion of fat or of organ walls, as opposed to linear arrays of atypical mesothelial cells on the free surface, which suggest a reactive process [13].

It is very important to distinguish MPM from serous papillary carcinoma of müllerian type (ovary or extra-ovarian) because there are many differences clinically and therapeutically, and a distinction should always be attempted. Morphologically, features that favor the diagnosis of mesothelioma over serous carcinoma are a prominent tubulopapillary pattern, polygonal cells with eosinophilic cytoplasm, absence of marked nuclear pleomorphism, and absence of a high nuclear rate [14].

Immunohistochemically, calretinin, WT-1 gene product, thrombomodulin and keratin 5/6 are markers for mesothelioma. Serous carcinomas of the ovary are positive for keratin 7, EMA, CAM 5.2, AE1/AE3, but negative for mesothelial markers.

The most characteristic pattern of spread is local with obliteration of the peritoneal cavity. Also, the tumor may locally invade the intestinal wall, the spleen, the liver, the pancreas, the bladder and the gastric wall.

The suggested treatment – as in our case – is surgical excision, associated with intraperitoneal chemotherapy. Over the past decade, the management of these patients has evolved similarly to ovarian cancer treatment and now involves cytoreductive surgery, heated intraoperative intraperitoneal chemotherapy (HIIC), and early postoperative intraperitoneal chemotherapy. This treatment prolongs survival rate to 50-60 months [3].

In our case, the patient was well two years after the initial diagnosis without any sign of disease recurrence, but was lost to further follow-up.

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