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Benign Cystic Mesothelioma of the Peritoneum

—A Case Report—

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A case of benign cystic mesothelioma in a 53-year-old woman is presented. The patient had abdominal pain and a palpable mass for 4 days. This mass was noticed incidentally by an obstetrician. A computed tomographic scan of the abdomen and pelvic sonography showed a cystic mass, 6×4cm, in the right lower quadrant close to the cecum and suggested cystadenocarcinoma of the ovary. Operation showed that the mass involved peritoneum in region of the appendix. Complete removal of the mass was done. Grossly it consisted of multilocular cyst containing clear, serous, gelatinous fluid. The light microscopic examination revealed that this lesion consisted of cystic spaces of various size and intervening connective tissue stroma. The cells lined the cysts varied from flattened to cuboidal with occasionally a picket-fence or hobnailed appearance in areas. Brush borders were seen on the luminal surface of some cells. Electron microscopic examination confirmed that the cells were mesothelial origin.

This lesion mimics cystic lymphangioma of the abdomen grossly and light microscopically, from which differential features are discussed.

Key words: *Benign cystic mesothelioma, Cystic mesothelioma, Benign mesothelioma, Peritoneal tumor*

INTRODUCTION

Benign cystic mesothelioma of the peritoneum is a rare tumor and is recently established as mesothelial cell derivation by electron microscopic investigations (Moore et al., 1980; Schneider et al., 1982). This lesion is considered as a clinically borderline variant between the benign adenomatoid tumor and malignant mesothelioma because of the presence of potential for recurrence and absence of any fatal result (Katsube et al., 1982). Two cases were reported in terms of multicystic peritoneal mesothelioma in the Korean literature (Jung et al., 1986). Because we have been unable to find detailed description, the cliniopathologic findings of present case are described in detail.

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CASE HISTORY

A 53-year-old female was admitted to the Department of Surgery because of abdominal pain and a mass in the right lower quadrant of the abdomen for 4 days. She had been taking some medicines for severe gastritis. This mass was noticed incidentally by an obstetrician. Physical examination on admission showed no specific findings except for tenderness in the right lower quadrant of the abdomen. The bowel sounds were decreased. Colon study was unremarkable. A computed tomographic scan of the abdomen showed a round low density mass, 6×4cm, in the right lower quadrant close to the cecum with displacement of the cecum and small bowel. There was no bowel wall change. The mass was largely cystic with focal solid area at the superior lateral portion, containing low density fluid. Pelvic sonography showed a cystic

mass with ill defined septation and suggested cystadenocarcinoma of the ovary. Exploratory laparotomy was done. Laparotomy revealed a multicystic mass, 7cm in diameter, between the cecum and retroperitoneum. The mass involved partially peritoneum in region of the appendix. The liver, pancreas, ovaries and appendix were unremarkable. Complete removal of this mass was done.

PATHOLOGIC FINDINGS

Gross Findings:

The mass consisted of translucent, multilocular cyst containing clear, serous, gelatinous fluid. It measured 7cm in diameter. The wall was largely thin and smooth. The focal solid area seen on CT scan consisted of small cystic spaces (Fig. 1).

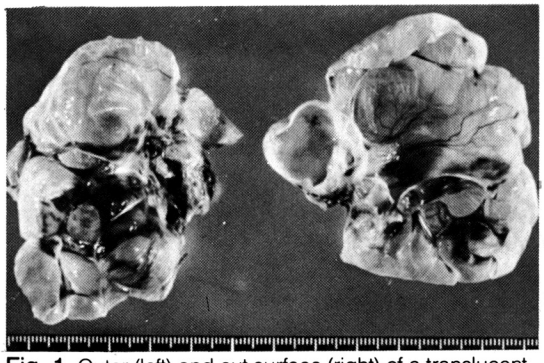

Fig. 1. Outer (left) and cut surface (right) of a translucent, mutilocular cystic mass. The cyst is smooth and thin-walled.

Light Microscopic Findings:

The tumor showed cystic, honeycombed structures of various size. The cells lining the cystic spaces varied from flattened to somewhat, taller, more plump cells. The larger cysts were lined by a single layer of flattened cells that were indistinguishable from the endothelial cells (Fig. 2), while the cells lined small cysts appeared cuboidal to polygonal with vesicular nuclei and amphophilic cytoplasm. Some of cysts were lined by flattened cells alternating with plump cells. The plump cells had a "picket fence" regularity or "hobnail" appearance in areas (Fig. 3). Some exfoliated papillary clusters of cells were seen in the cystic lumen and these cells contained intracytoplasmic eosinophilic secretory materials. There were some cells that had luminal brush borders and intercellular bridges. Some of cystic spaces contained eosinophilic, amorphous precipitate, which stained positive focally

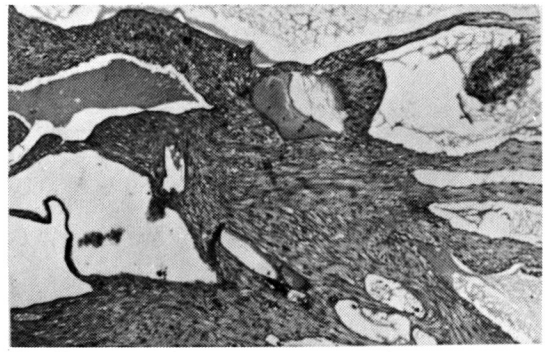

Fig. 2. Microscopically, the mass is composed of cystic spaces largely lined by a single layer of flattened cells that are similar to those of lymphangioma. Note the stroma composed of dense fibous tissue. H&E×40

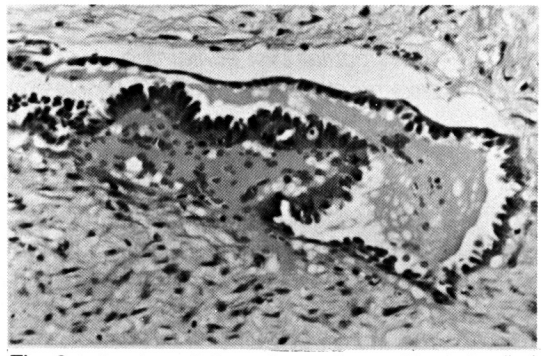

Fig. 3. In focal area lining cells are plump with a hobnailed or picket-fence appearance. The stroma is loose and myxoid with immature stellate fibroblasts. H&E×400

with Alcian blue at pH 2.5, and was negative with Mayer's mucicarmine and PAS stains. There was a variable amount of intervening connective tissue with prominent vascular structure and numerous immature stellate fibroblasts. The connective tissue septa varied in thickness and composition. In many areas loose myxoid stroma was seen and stained positive with Alcian blue at pH 2.5. The serosal aspect of the cyst was covered by partly fibrofatty adhesion and partly a layer of serosal mesothelial cells. The outer wall of the mass was composed of inflamed fibrous and granulation tissue, often admixed with adipose tissue and fibrin: There were embedded mesothelial cells arranged in glands, nests, cords and single cells. These patterns of the mesothelial cells were very similar to those of adenomatoid tumor.

Electron Microscopic Findings:

The cells lined the cystic spaces were of mesothelial

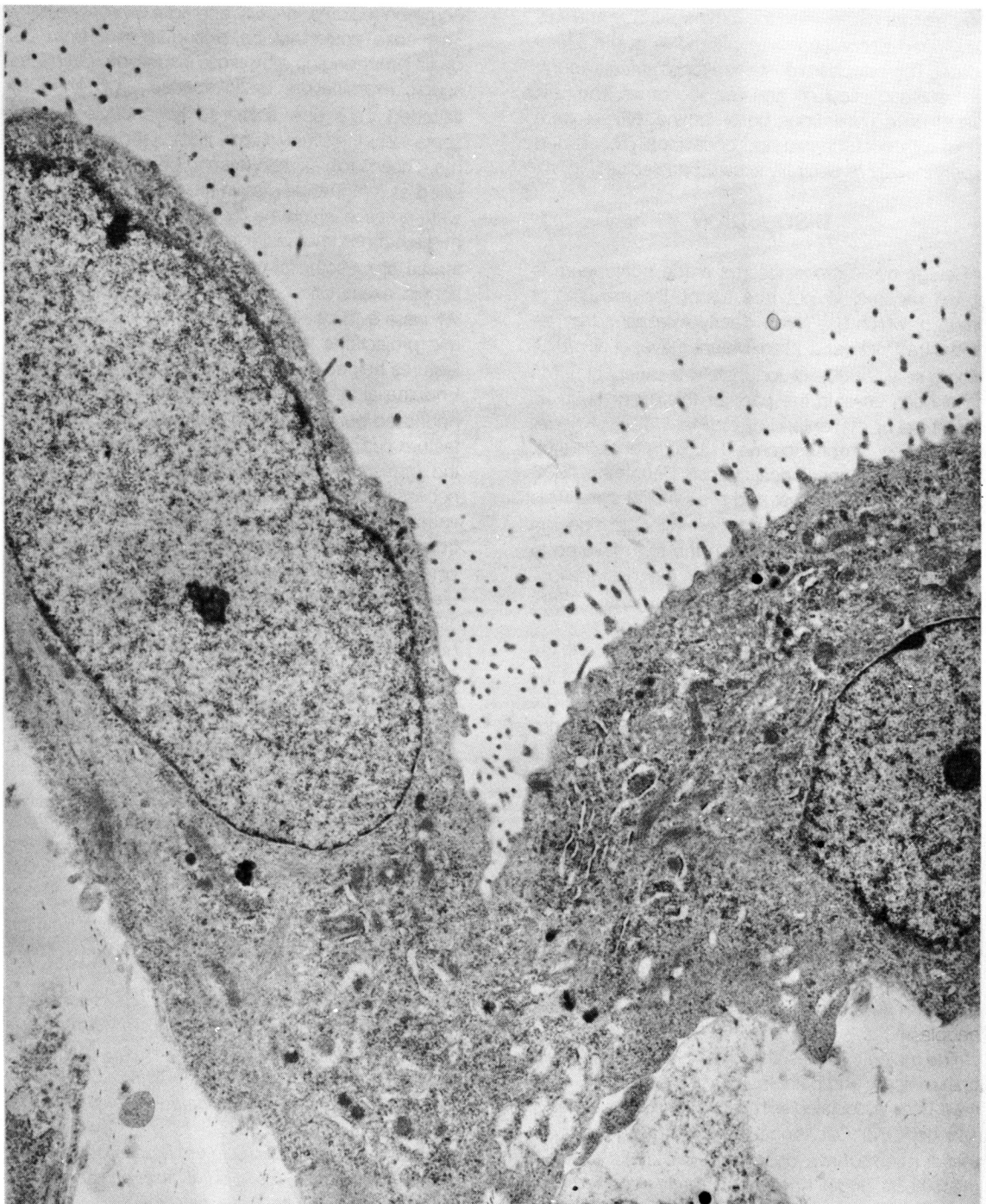

Fig. 4. Electron photomicrograph showing two mesothelial cells with numerous short microvilli along the luminal surfaces of the cells. Note tonofilaments, rough endoplasmic reticulum and free ribosomes in their cytoplasm. Lead citrate-uranyl acetate×10,000.

cells that were characterized by numerous short and long slender microvilli on the luminal surface and well developed desomosomal attachments at the lateral border. The cytoplasm contained tonofilaments, rough endoplasmic reticulum and free ribosomes. The cells rested on a continuous basal lamina. Nuclei were ovoid with euchromatin and occasionally contained a peripherally or centrally situated nucleolus (Fig. 4).

DISCUSSION

Benign cystic mesothelioma of the peritoneum is a well recognized but rare tumor, the mesothelial origin of which has been clearly established by recent ultrastructural studies (Mennemeyer et al., 1979, Moore et al., 1980). Unfortunately, a variety of terms have been used in the past for this entity, such as peritoneal or mesenteric cysts (Plaut, 1928; Krieger et al., 1952), lymphangioma (Hamilton et al., 1946), benign papillary peritoneal cystosis (Jacobson, 1974) peritoneal inclusion cyst and mesothelial cysts. This has led to considerable confusion. Although "benign cystic mesothelioma" has been the term favored in recent publications, others prefer the designation" peritoneal inclusion cyst" (McFadden et al., 1986; Rosai, 1989).

Approximately 50 cases have been reported in the world literature (MaFadden et al., 1986). None of the patients have had a history of asbestos exposure. The typical clinical features are adult female predominance (mean age 37 years: 5:1 female; male ratio), and presenting manifestation of lower abdominal pain, a palpable mass, or both. Approximately 30% of this tumor have recurred locally, once or several times, at postoperative intervals of 4 months to 7 years (mean 32 months). Approximately 30% of patients have had a history of prior abdominal surgery, pelvic inflammatory disease, or endometriosis. For these reasons, it has been suggested that at least some examples of this disorder are a result of pelvic inflammation and adhesion formation and do not represent cystic neoplasm.

The majority of mesothelioma reported have been both clinically and pathologically malignant and many have been associated with prior exposure to asbestos (Moertel 1972). Occasional reports of benign papillary mesothelioma have described, but cystic tumor of mesothelial origin have been rarely reported. The majority of benign cystic lesions of the abdomen had been classified as lymphangioma (Carpenter et al. 1982). While there is little similarity between the more classic manifestations of benign cystic mesothelioma and lymphangioma, the two lesions may share some common features, grossly and light microscopically. This case could not be distinguishable from the cystic lymphangioma by gross inspection. On microscopic examination, cystic spaces largly lined by flattened cells and scattered lymphocytes in the septa were indistinguishable from cystic lymphangioma. These facts caused them to be erroneously classified as lymphangiomas in the past. However there were areas in which the lining cells were plump and had a hobnailed or picket-fence configuration characteristic of mesothelial cells. In areas on light microscopic examination at high magnification, luminal villi were evident. They appeared as fuzzy cytoplasmic projections from the luminal cell surface. This features helped to identify cells as mesothelial cells. Endothelial cells occasionally appear plump or even hobnailed but do not have microvilli. There was other feature supportive of the mesothelial cells, including the presence of papillary tufts. This lesion is also necessary to be differentiated from very rare cystic variant of adenomatoid tumor and a lesion derived from mesonephric rest. Adenomatoid tumors are small, usually asymptomatic, generally limited to the uterus or fallopian tube and do not recur (Katsube 1982). The cyst of mesonephric origin is expected to have cuboidal rather than flattened cells, no papillary clusters, and surrounding cuffs of smooth muscle (Eickhoff, 1978).

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