

# Well Differentiated Papillary Mesothelioma of the Ovarian Surface – A Case Report –

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Well differentiated papillary mesothelioma (WDPM) is an unusual variant of epithelial mesothelioma. Most WDPMs exhibit either benign or indolent behavior. Making the differential diagnosis between this rare tumor and serous papillary carcinoma can be problematic. We report here on a case of a 43-year-old woman with a WDPM of the surface. She presented to our hospital for a routine gynecologic evaluation, and she had no specific symptoms or a history of asbestos exposure. Gynecologic ultrasonography revealed a right ovarian mass that measured 6 × 3.8 × 3 cm in size. No ascites was detected. Right salpingo-oophorectomy was performed; grossly, the tumor was a yellowish firm, multinodular mass. Microscopically, the tumor consisted of numerous papillae that were lined by a single layer of uniform mesothelial cells. Nuclear pleomorphism and mitoses were not found. On immunohistochemical study, the tumor cells were positive for calretinin and cytokeratin, but they were negative for CEA. It is important to differentiate WDPM from serous papillary carcinoma or other malignant tumors to avoid treating them as malignant tumors.

**Key Words :** Mesothelioma; Ovary

Well differentiated papillary mesothelioma (WDPM) is an unusual variant of epithelial mesothelioma. It occurs mainly in the peritoneum, and this tumor is most commonly seen in young women who have no history of asbestos exposure; it is often found incidentally at laparotomy that is done for other indications.<sup>1,2</sup> WDPM rarely occurs at other sites, including the ovary,<sup>3,4</sup> pericardium,<sup>5</sup> and the tunica vaginalis.<sup>6,7</sup> The tumor is occasionally found on as a frozen sectioning because surgeons suspect this tumor to be other malignant tumors, such as diffuse malignant mesothelioma, or peritoneal dissemination from other tumor sites or ovarian serous papillary carcinoma.<sup>1</sup> Most WDPMs exhibit either a benign or indolent behavior.<sup>1-8</sup> Making the differential diagnosis between this rare tumor and serous papillary carcinoma can be problematic. WDPM must be distinguished from the significantly more aggressive lesions to avoid unnecessary treatment.<sup>1,8</sup> We report here on a case of a 43-year-old woman with a WDPM of the surface epithelium of the ovary.

## CASE REPORT

A 43-year-old woman presented to Department of Gynecology for routine gynecologic evaluation. She was a housewife, and underwent a total laparoscopic hysterectomy for leiomyomas two years ago. She had no specific symptoms or history of asbestos exposure. Gynecologic ultrasonography revealed a right ovarian mass that measured 6 × 3.8 × 3 cm in size. No ascites was detected. The serum CA-125 was 17.8 U/mL (normal: <35 U/mL), the serum CA19-9 was 2.7 U/mL (normal: <34 U/mL), and the serum AFP was 2.2 ng/mL (normal: <10.9 ng/mL). Right salpingo-oophorectomy was performed. Grossly, the tumor was a yellowish firm, multinodular mass (Fig. 1). Microscopically, the tumor consisted of numerous papillae that were lined by a single layer of uniform mesothelial cells (Fig. 2). Any nuclear pleomorphism and mitoses were not found. Psammoma bodies were present (Fig. 3). On immunohistochemical study, the tumor cells were positive for calretinin (Dako, Carpinteria, CA, USA) (Fig.

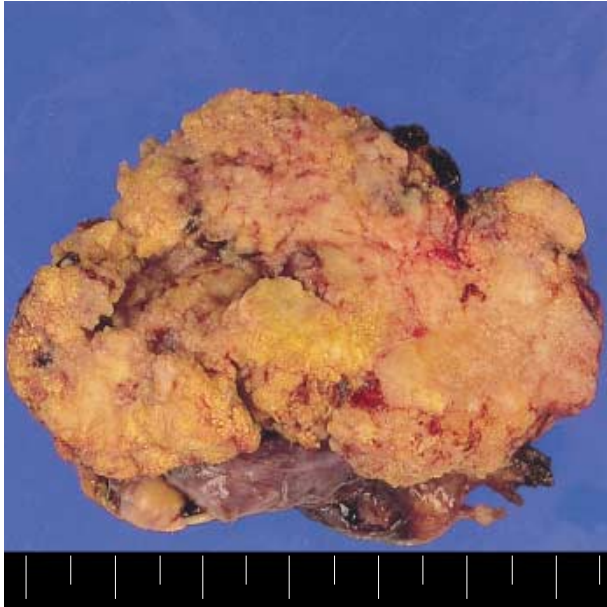


Fig. 1. The tumor consists of yellowish firm, multiple nodules with a papillary architecture.

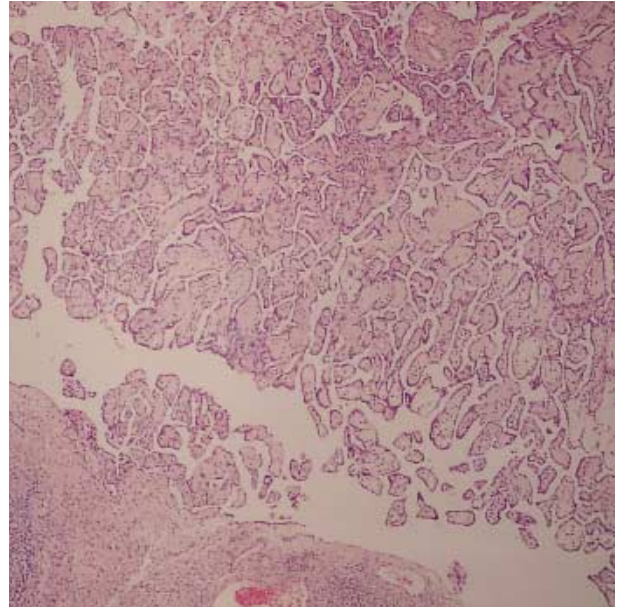


Fig. 2. The tumor shows slender fibrovascular papillae lined by uniform mesothelial cells.

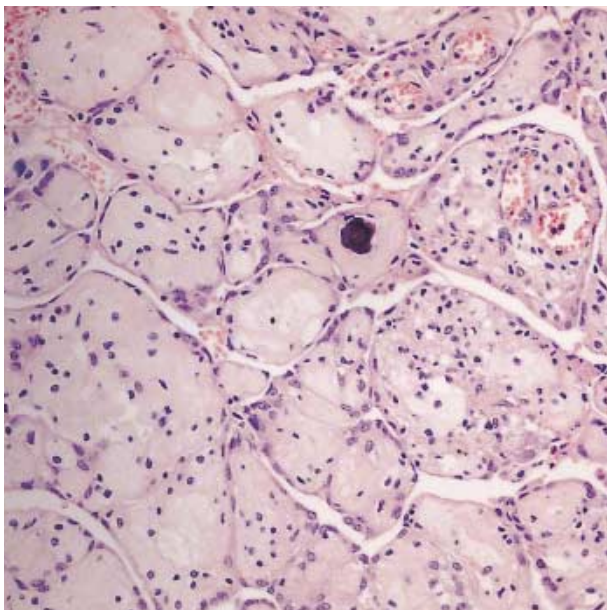


Fig. 3. Fibrous papillae show a single layer of cuboidal mesothelial cells with minimal cytologic atypia and a psammoma body.

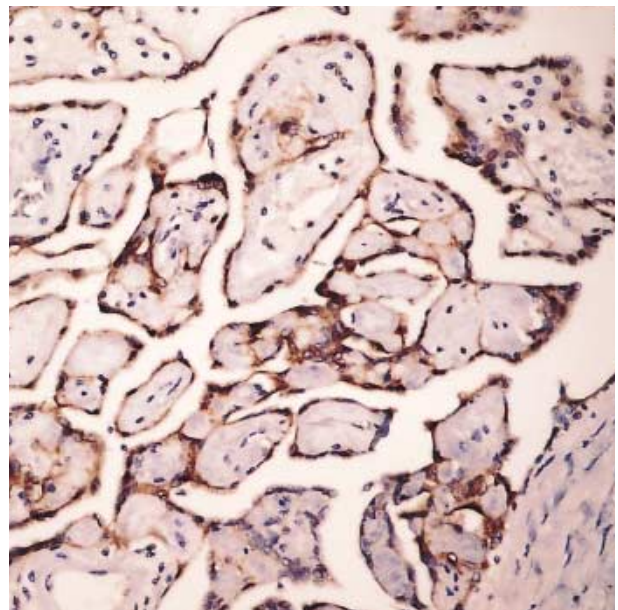


Fig. 4. The tumor cells stain positively for calretinin (Immunohistochemical stain).

4) and cytokeratin (Dako), but they were negative for CEA (Dako). Histochemical stains demonstrated focal alcian blue staining-positive intracytoplasmic material, that was negative by periodic acid schiff staining with and without diastase predigestion. The patient has remained well after a follow-up of seven months.

## DISCUSSION

WDPM is considered to be a benign subtype of mesothelioma that predominantly affects the peritoneum of women in the third and fourth decade.<sup>1,2</sup> There are only two reports of WDPM of the ovary.<sup>3,4</sup> The histologic appearance of WDPM is that of thin fibrovascular papillae lined by relatively uniform cuboidal meso-

thelial cells.<sup>1</sup> Psammoma bodies are occasionally encountered.<sup>1,9</sup> WDPM must be differentiated from metastatic peritoneal adenocarcinoma, serous papillary adenocarcinoma, diffuse malignant mesothelioma and reactive mesothelial hyperplasia.<sup>1</sup>

The most important point in the differential diagnosis of this case was to rule out serous papillary adenocarcinoma of ovary. The absence of multilayering, cell stratification, cytologic atypia and mitotic activity supported a diagnosis of WDPM.<sup>1</sup> Calretinin is a 29-kDa calcium binding protein that is expressed in central and peripheral neural tissues, and in a limited number of non-neural tissues including normal and neoplastic mesothelium.<sup>10</sup> Calretinin was previously identified to be the most sensitive (88%) and specific (100%) mesothelial marker to differentiate diffuse peritoneal mesothelioma in woman from serous papillary ovarian carcinoma and serous papillary peritoneal carcinoma.<sup>11</sup> This case showed reactivity for calretinin and cytokeratin, but any reaction for CEA was negative. The immunoreactivity for CEA and CD15, and another glycoproteins, such as Ber-EP4, TAG-72, and MOC-31 is quite useful in supporting a diagnosis of adenocarcinoma.<sup>12</sup>

Diffuse malignant mesothelioma (DMM), in which well-differentiated papillary elements are prominent, can also be confused with WDPM. DMMs are usually bulkier than WDPMs and they demonstrate diffusely infiltrative cytologically atypical cells.<sup>9</sup> Abnormal mitoses and bizarre cells are also found in DMM.<sup>13</sup> Patients with WDPM generally have a far better prognosis than those with DMM.

WDPM must also be distinguished from reactive mesothelial hyperplasia. Papillary structures may occur in mesothelial hyperplasia.<sup>14,15</sup> However, in comparison the conspicuous papillary pattern of WDPM, such changes are rare in with hyperplasia. The lack of inflammation, the accompanying reactive changes in the adjacent serosal tissue and the relatively uniform or monotonous tumor cells are essential points for the diagnosis of WDPM.

In conclusion, it is important to diagnose this tumor in order to refrain from an aggressive therapeutic approach. The histological findings that include the cytologic features and the immunohistochemical study results are important for making the correct diagnosis of WDPM.

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