Adenomatoid mesothelioma of the peritoneum (AMP) is a rare benign tumor originating from mesothelial cells. Most frequently, AMP occurs between 26 and 55 years of age, at a mean age of 41 years. In contrast to diffuse malignant mesothelioma, which has been linked to asbestos exposure, the etiology of AMP has not been established. Only a minority of patients have symptoms related to the tumor. AMP may present local recurrence, but it has no potential for malignant transformation. Although there are many case reports of abdominal mesotheliomas, to date, there have been no reports of MR imaging features of AMP. In this article, we present the MR imaging features of a case of AMP with histopathological correlation.

**CASE REPORT**

A 25-year-old woman presented with pelvic pain. The patient had had a cesarean section 3 years before her visit and an appendectomy about 6 months earlier than her onset. Routine ultrasound exam showed an expansive pelvic lesion suggesting an adnexal origin, most likely an ovarian neoplasm, and this finding was confirmed by two other ultrasound exams, each one performed at a different facility. A pelvic-abdominal MR exam was requested for lesion characterization. Laboratory tests were within normal values, except for CA125 of 37.9 µ/ml (normal range 0.0 to 35.0 µ/ml).

**Imaging Findings**

Transvaginal pelvic sonography showed a retrouterine adnexal mass extending towards the left parauterine region with a complex echotexture containing a homogeneous solid component about 5.0 cm in size and small cystic areas intermingled with linear septa. The mass measured 10.8 x 6.1 x 10.5 cm (volume of 359.7 cm³). Color Doppler imaging demonstrated vascularization of the solid area and of some septa in the cystic region, with a resistive index ranging from 0.60 to 0.70. Ovaries were identified in neither ultrasound exam. There was no sign of ascites (Figure 1).

The MR images demonstrated a large, expansive, and well-delimited lesion with lobulated contours; T1-weighted images showed homogeneous signal intensity predominantly with a low signal, whereas T2-weighted sequences were heterogeneous with small high intensity foci (Figure 2). The lesion measured 9.2 x 7.2 x 8.0 cm and was located close to the ovaries, which were dislocated anterolaterally. The contours, dimensions and signal intensity of the ovaries were normal. The uterus was normal in shape, dimensions and signal intensity, and it was also dislocated anteriorly. A small amount of free fluid was present in the peritoneal cavity surrounding the lesion. The upper abdominal MR evaluation showed the extension of the free peritoneal fluid and normal anatomy of abdominal organs. Computed tomography of the pelvis without...
intravenous injection of contrast agent was performed to search for calcification and showed a low homogeneous coefficient of attenuation for the lesion, ranging from –2 to +10 U.H, with no evidence of calcification (Figure 3).

**Patient outcome**

The patient was submitted to open abdominal surgical resection of the lesion, which had adherences to the rectal anterior wall but no invasion of the uterus or adnexa. Macroscopic anatomopathological examination revealed various irregular fragments of brownish, friable tissue that measured 15.0 x 12.0 x 4.0 cm and weighed 170.0 g. Microscopic analysis revealed a well-differentiated mesothelial neoplasia, which is detailed in Table 1. Immunohistochemistry evaluation was carried out in the histological sections with an immuno-peroxidase reaction via the avidin-biotin peroxidase method, with the primary antibodies calretinin + CEA- and BerEp4- (Figure 4).

A follow-up MR exam revealed an expansive, predominantly cystic lesion with high protein content located in the posterior cul-de-sac; this lesion had shown a progressive increase in volume over 3 years and was characterized as a recurrent tumoral lesion (Figure 5).

**Table 1 - Histopathological findings of the reported case**

| Parameter                                    | Findings                                      |
|----------------------------------------------|-----------------------------------------------|
| **Origin**                                   | Primitive                                     |
| **Histogenetic lineage**                     | Mesothelial                                   |
| **Structural differentiation**               | Well-differentiated                           |
| **Supporting tissue**                        | Loose – vascularized                          |
| **Predominant cell type**                    | Medium size – Polygonal                       |
| **Cell arrangement**                         | Acinar – Tubular – Papilliform                |
| **Cytoplasm characteristics**               | Abundant – Microvacuolized – Basophilic       |
| **Nucleus characteristics**                 | Medium volume – Round – Discrete nucleoli – Fine chromatin – Homogeneous chromatin |
| **Nucleus/cytoplasm ratio**                  | Maintained                                     |
| **Extracellular material produced**          | Absent                                        |
| **Mitotic index**                            | Low (up to 1)                                 |
| **Degree of necrosis**                       | Absent                                        |
| **Degree of atypia**                         | Mild                                          |
| **Cytologic (nuclear) grade**                | 1                                             |
| **Histological grade**                       | I                                             |
| **Capsular limits**                          | Absent                                        |
| **Tumor limits**                             | Poorly defined                                |
| **Vascularization**                          | Abundant                                      |
| **Forms of infiltration**                    | Absent                                        |
| **Inflammatory infiltrate**                  | Present                                       |
| **Predominant cells**                        | Lymphocytes – Histiocytes                     |
| **Microcalcifications**                      | Present – Focal                               |
| **Desmoplasia**                              | Absent                                        |
| **Hemorrhage**                               | Absent                                        |
| **Vacuolar embolization**                    | Not visualized                                |
| **Lymphatic embolization**                   | Not visualized                                |
| **Perineural infiltration**                  | Not visualized                                |
| **Surgical safety margins**                  | Poorly defined                                |
The lesion was resected surgically, and there was no sign of recurrent disease on subsequent follow-up exams. The patient is currently asymptomatic.

DISCUSSION

Mesotheliomas are rare tumors originating from mesothelial cells of serosal membranes such as the pleura, peritoneum, pericardium, and tunica vaginalis. Simultaneous pleural and peritoneal involvement occurs in 30-45% of cases, whereas disease limited to the peritoneum occurs in 10 to 20% of the patients. Peritoneal mesotheliomas can be classified as benign (adenomatoid, fibrous), borderline (multicystic, well-differentiated papilliferous), and malignant (epithelioid, sarcomatoid or biphasic/mixed), and their characteristics are described in Table 2.

Peritoneal adenomatoid mesothelioma (AMP) is a benign neoplasia of unknown etiology that primarily involves the genital tract of both sexes, occurring more frequently among males. AMP is an uncommon tumor, usually asymptomatic, and is incidentally discovered during radiologic exams, surgeries or autopsies. It is typically a single polypoid or nodular small lesion (2.0 cm or less) that can measure up to 13 cm in diameter in a few cases. Adenomatoid tumors are usually solid, not encapsulated and often contain small cystic lesions (0.4 to 1.5 cm). When present, signs and symptoms are abdominal pain, loss of weight, loss of appetite, nausea, fluid accumulation in the peritoneal space (ascites), and a pelvic mass.

In the present case, the tumor was 15 cm at its widest diameter on pathological examination, exceeding the size of previously reported masses. This might explain why the patient was symptomatic; additionally, after surgical resection, symptoms disappeared for a period of approximately one year. After that, the lesion recurred, and the patient was submitted to a new surgical intervention. The patient has now been free of the disease for 7 years.

Usually, surgical resection is the treatment of choice for AMP. Accurate diagnosis and staging are important because of the obvious therapeutic implications. Although benign, AMP is a source of great concern due to the differential diagnosis of malignant entities.

In the present case, in view of the location of the tumor...
Table 2 - Features of mesothelioma-type tumors as reported in the literature

| Feature                                                                 | Well-differentiated papilliferous peritoneal mesothelioma | Multicystic peritoneal mesothelioma | Fibrous peritoneal mesothelioma | Malignant peritoneal mesothelioma | Peritoneal adenomatoid mesothelioma |
|------------------------------------------------------------------------|-----------------------------------------------------------|-----------------------------------|--------------------------------|----------------------------------|-----------------------------------|
| Mean patient age                                                       | 30 to 50 years (46.1 ± 13.65 years)                       | 37 years and 10 months            | 40 to 70 years                  | 26 to 55 years (average of 41 years old) |
| Sex                                                                    | Predominance in women (65.9%)                             | Predominance in women of reproductive age | Predominance in men             | Predominance in men               |
| Risk factors                                                           | No established etiology                                  | History of abdominal surgery (53% of cases), endometriosis or inflammatory pelvic disease. No etiologic association with asbestos. | Chronic peritoneal irritation and previous laparotomy | Exposure to asbestos (15 to 30 % of cases) | Exposure to beryllium, nuclear radiation, and chronic inflammatory diseases. |
| Macroscopy                                                             | Multiple or single small nodular lesions incidentally detected during surgery (0.5 to 3 cm) | Multiple confluent translucent cysts forming a mass, without hemorrhage, fat or calcifications in their walls | Encapsulated and solid lesions | Solitary bulky lesion, usually small (2cm or less); with no capsule; may present small cystic components |
| Clinical signs and symptoms                                           | Asymptomatic tumor (55%) Abdominal pain (38.3%) Ascites (33.3%) Pelvic mass (11.1%) Chronic pelvic inflammatory disease (11.1%) Constipation (5.5%) | Abdominal mass (29%) + distension/abdominal pain (46%) | Asymptomatic abdominal mass (18%) | Abdominal pain syndrome Ascsites Abdominal mass Alterations of intestinal transit (alternating diarrhea and constipation or symptoms simulating an obstructive crisis). | Asymptomatic (incidental finding) Abdominal pain Weight loss Loss of appetite Nausea Ascites Pelvic mass |
| Survival                                                               | More than five years after diagnosis                     | Mean survival of 8-12 months after diagnosis | Survival                        | Surgical                         |
| Treatment                                                              | Surgical                                                  | The tumor is not sensitive to chemotherapy or radiotherapy | Surgical                        | Cytoreduction surgery with extensive peritoneectomy and perioperative intraperitoneal chemotherapy | Surgical |
| Location                                                               | 26% in abdominal or pelvic organs 22% in the omentum 16% on the pelvic wall 14% in the mesentery 14% in the peritoneum 8% in the Douglas cul-de-sac |                                |                                | Genital tract of both sexes |
| Number of reported cases                                               | 41 cases                                                  | 130 cases                         | 15 cases                        | 1-2 cases/million inhabitants/year |
| Prognosis                                                              | Possibility of malignant transformation                  | Local recurrence                  | Local recurrence                | Local recurrence                  |
| Differential diagnosis                                                 | Serous tumor of the ovarian surface Metastatic tumor usually of the gastrointestinal tract | Cystic lymphangioma Endometriosis Cystoadenoma and cystoadenocarcinoma of the ovary Teratoma Peritoneal pseudomixoma Necrotic leiomyoma - Leiomyosarcoma Peritoneal inclusion cyst | Peritoneal tuberculosis Peritoneal carcinomatosis Peritoneal lymphoma Metastases of an ovarian carcinoma | Malignant mesothelioma Mesothelial hyperplasia Well-differentiated papilliferous peritoneal mesothelioma Metastases |
in the cul-de-sac and its histopathological characteristics (cells clustered in a papillary formation), it was necessary to establish a differential diagnosis with adenocarcinoma.\(^1\) Histology revealed medium-sized polygonal cells in an acinar, tubular and papilliform cell arrangement, a low mitotic index (up to 1), a mild grade of atypia, abundant vascularization, and absence of necrosis, suggesting an adenomatoid tumor.\(^{14}\)

Immunohistochemistry revealed positivity for calretinin, which labels mesothelial cells in 60 to 100% of cases\(^{19,13}\) and rarely labels adenocarcinomas (0 to 28%). In addition, the cells of the neoplasia reported here were negative for BerEp4, which labels epithelial cells that are not present in mesotheliomas.\(^2,5\) The present case was negative for CEA immunoreagent, which frequently labels pulmonary and gastrointestinal carcinomas and is detected in only 0 to 35% of serous ovarian carcinomas.\(^2,6\) Thus, negativity of this marker is of no help for differentiation between adenomatoid tumors and adenocarcinomas. The possibility of the latter was ruled out due to immunohistochemistry compatible with an adenomatoid tumor, and by MRI and laparotomy findings that revealed disease-free ovaries. Another possible differential diagnosis for this case, arising from its location in a cul-de-sac, would be a metastatic tumor. However, CEA negativity and calretinin positivity do not favor this possibility, as demonstrated in Table 3. Other differential diagnoses are cysts of peritoneal inclusion, hemangiomas, lymphangiomas,\(^{12}\) mesothelial hyperplasia, malignant mesotheliomas,\(^9\) and well-differentiated papilliferous mesotheliomas.

### Table 3 - Positivity of immunohistochemical markers in adenocarcinomas and mesotheliomas\(^{11,25}\)

| Marker          | Adenocarcinomas (%) | Mesotheliomas (%) |
|-----------------|---------------------|-------------------|
| CEA             | 90-100              | 0-10              |
| B72.3           | 81                  | 0-5               |
| BEREP4          | 90-100              | 0-11              |
| CD15(LEU-M1)    | 58-100              | 0-10              |
| Calretinin      | 6-9                 | 42-100            |

Mesothelial hyperplasia has been associated with peritoneal insults such as hernia, ectopic tubal pregnancy, and abdominal cirrhosis and tuberculosis\(^7\) and is accompanied by adherences and chronic inflammation.\(^{14}\) This entity rarely produces tumoral masses and does not have the tubulopapilliferous complex or the labyrinth architecture of mesotheliomas.\(^9\) The differential diagnosis with malignant mesothelioma and well-differentiated papilliferous mesothelioma is made on the basis of the distinct histological characteristics of these tumors when compared to AMP.\(^{17}\)

MR has become a valuable noninvasive technique for evaluation of the female pelvis,\(^{18,20}\) with advantages over computed tomography and ultrasound for diagnosis and for staging various pathological conditions of the pelvis (leiomyoma, adenomyosis, carcinoma of the endometrium and of the uterine cervix, carcinoma of the vagina, ovarian cysts, endometriosis, teratomas, polycyclic ovaries, and other ovarian masses).\(^{18,20}\)

MR has proven to be a highly sensitive modality for characterization of pelvic masses, allowing physicians to determine whether the pelvic mass is uterine or of adnexal origin and also to characterize most adnexal masses.\(^{20}\) MR can also provide multiplanar information, revealing additional information when compared to CT or US. This is especially true along the pelvic walls and the presacral space.\(^{18,20}\) MR is also especially useful for surgical planning\(^{15}\) and patient follow-up. Low et al\(^{21}\) studied 24 patients with suspected peritoneal tumors and found that MR had higher sensitivity, specificity and accuracy than CT in the detection of tumors (84%, 87% and 86%, compared to 54%, 91% and 74%, respectively, for CT) and was superior for detection of carcinomatosis and of tumors measuring less than 1 cm in diameter (75% to 80% for MR and 22% to 33% for CT). Post-contrast T1-weighted images with fat suppression were proven to be the most sensitive MR technique for detecting peritoneal disease. MR and CT showed identical performance for detection of tumors measuring more than 2 cm and 1 to 2 cm in diameter.\(^{21}\)

Notably, the high sensitivity of the MR exam could depict both ovaries as free of disease and was able to characterize the lesion as not having ovarian or uterine origin; this could not be achieved by ultrasound examination. Also, to date, we believe that this is the first case both to show MR findings for AMP and to correlate these findings to ultrasound and computed tomography. Descriptions of imaging findings regarding AMP are scarce in the literature. AMP seems to have no specific radiological characteristics, and it is important to establish a correlation between clinical presentation and the imaging and laboratory findings. At this point, it is necessary to reinforce that diagnosis can only be confirmed by anatomicopathology and immunohistochemistry.
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