



A Rare Case of Well Differentiated Papillary Mesothelioma of the Tunica Vaginalis

Authors

Megha Yadav¹, Mukesh Agrawal², Jaya Bhaskar Reddy³, Sujani Madabhushi⁴,
Mujeeb Siddiqui⁵

^{1,3,4,5}Department of Pathology, VIMTA LABS Ltd, Cherlapally, Hyderabad, Telangana, India.

²Lab Director, VIMTA LABS Ltd, Cherlapally, Hyderabad, Telangana, India

Corresponding Author

Megha Yadav

Head, Dept of Pathology, VIMTA Labs Ltd, Phase-II Cherlapally, Hyderabad- 500051, Telangana, India

Email drmegha1852@gmail.com

Abstract

Well Differentiated Papillary Mesothelioma (WDPM) is an uncommon neoplasm of the paratesticular area usually presents as hydrocele. Here we report a rare case of benign papillary mesothelioma of the tunica vaginalis presented as hydrocele with singly polypoidal grossly papillary lesion with multiple foci in the hydrocele sac. Histologically tumor shows branching papillary structures with fibrovascular core and cytologically bland nuclei without any mitosis, necrosis or area of invasion. In addition histochemistry is done to differentiate it with other papillary neoplasm. WDPM must be distinguished from the aggressive lesion to avoid unnecessary treatment.

Keywords- *Well Differentiated Papillary Mesothelioma (WDPM), Paratesticular area, Hydrocele, Tunica Vaginalis.*

Introduction

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 ^[1]. WDPM occurs rarely at other sites including pericardium, pleura and tunica vaginalis. Unlike the conclusive link between asbestos exposure and diffuse malignant mesothelioma an association of asbestos and WDPM has not been definitely established ^[2].

The majority of tumors with histological features of WDPM have behaved in a benign or indolent fashion. However several WDPM have pursued a more aggressive course resulting in death after the development of DMM ^[2]. We report a case of WDPM in a 50 year old male presented with hydrocele.

Case Report

A 50 year old patient presented with scrotal swelling in a surgical OPD. The swelling was gradually increasing in the size within a period of 8 months. No history of trauma or asbestos

exposure. Local examination revealed hydrocele. Ultrasonography of the testicular region showed papillary growth of 1.5cm in the paratesticular area. Orchidectomy was done and sent for histopathological diagnosis.

Macroscopically testis was normal with one pedunculated papillary mass of 1.5x1cm in diameter found in the tunica vaginalis (Fig 1). The hydrocele sac was of 10x6x4 cm. Cut section of the sac was showing diffuse infiltrating nodules of varying sizes ranging from 0.5 to 0.8 cm. Hydrocele sac was showing hemorrhagic areas.

Microscopically the papillary mass and the nodules from the sac wall were showing multiple branching papillae with fibrovascular cores. These papillae were lined by single layer of flat cuboidal to columnar epithelium (Fig 2). No cellular pleomorphism, mitotic activity, necrosis or stromal invasion was noted. Immunohistochemistry of the tumor was showing positivity for EMA, Cytokeratin, Vimentin and calretinin.---- of cells show positive for p53 and negative for CEA (Fig 3,4)



Fig 1. Pedunculated papillary mass of 1.5x1CM in the tunica vaginalis

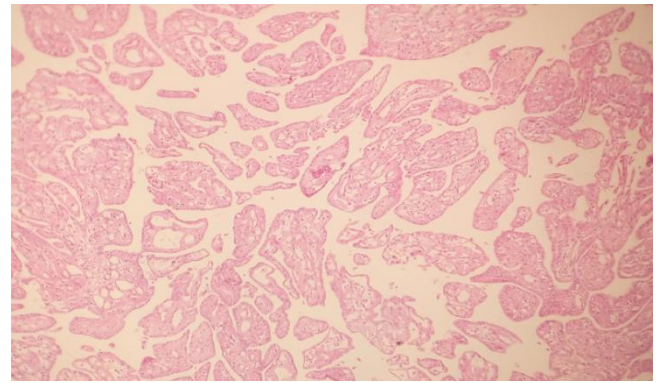


Fig 2. Papillary mass with branching papillae with fibrovascular cores lined by single layer of cuboidal epithelium (10x)

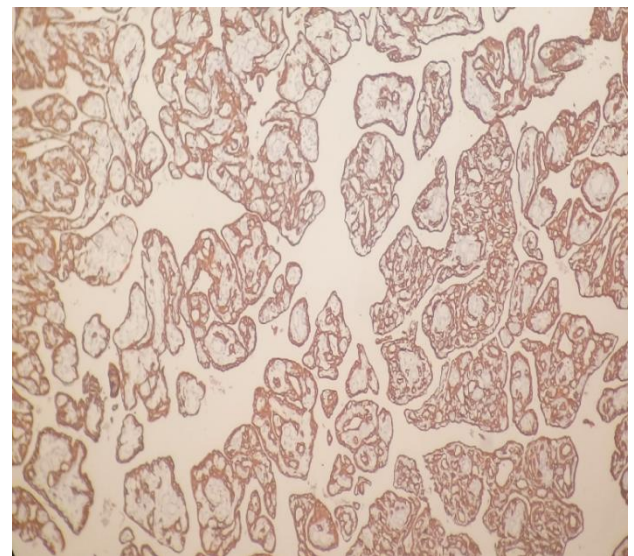


Fig 3. Positive for cytokeratin

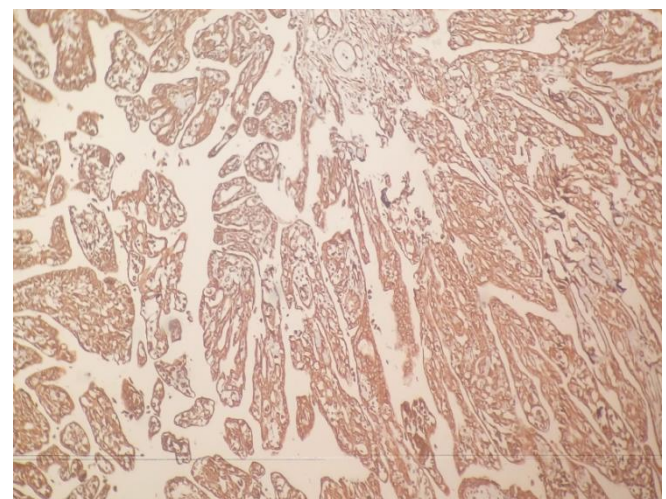


Fig 4. Positive for Vimentin

Discussion

Mesothelioma can arise from the pleura, peritoneum or paratesticular area in decreasing

order of frequency. Paratesticular mesothelioma can arise from the tunica vaginalis formed by an out pouching of the abdominal peritoneum. Only 0.3-5% of cases of mesothelioma occur in the tunica vaginalis [3]. WDPM of tunica vaginalis is considered benign mesothelioma in the World Health Organization fascicle on the tumors of the urinary system and male genital organs, primarily due to indolent behavior. Main mesothelial lesion of the paratesticular area include mesothelial hyperplasia, mesothelial cyst, adenomatoid tumor, WDPM and malignant mesothelioma. Barbera and Rubino reported the first case of WDPM in 1957. To the best of our knowledge 19 cases of paratesticular WDPM have been published to date. WDPM were described in men between the ages of 18-70 years (median 56). All reported cases with WDPM had a good outcome with no evidence of disease recurrence or progression [4].

The definition of WDPM has been controversial, but most experts currently restrict the term only to localized, solitary and exophytic tumors of delicate papillae lined by bland cuboidal cells [2],[4],[5]. Some of the cases were designated as "papillary mesotheliomas with borderline features or localized mesotheliomas of low grade malignancy, cautioning about their low malignant potential, despite the benign follow-up [2],[4].

Main differential diagnosis of WDPM includes mesothelial hyperplasia, malignant mesothelioma and papillary cystadenoma. Mesothelial hyperplasia sometimes also composed of papillary structure but often not contain fibrovascular core and they are always associated with the inflammation and reactive change [3],[6],[7]. Presence of invasion, marked cytological atypia, atypical mitosis, necrosis, big mass are the main differentiating features of WDPM and MM.

At immunohistochemical level, no study to date has compared the expression of different markers between diffuse malignant mesothelioma and well differentiated papillary mesothelioma. Rather studies have focused on differentiating diffuse malignant mesothelioma from reactive mesothelial proliferation. Although no single

antibody has been shown to carry a discriminatory value, several markers have been reported to be more commonly expressed in diffuse malignant mesothelioma than in reactive mesothelium [8]. Calretinine, Pan ck, EMA and Vimentin shows positivity for the WDPM. CEA will be negative which may be the main differentiating feature to a papillary serous cystadenoma.

The etiology of WDPM has not been clearly elucidated. It has been suggested that proliferative lesions might be related to local trauma, herniorrhaphy or a long term hydrocele. A relationship between asbestos exposure and mesothelioma of the tunica vaginalis is postulated [3].

Our case had no asbestos exposure like the rest of the reported cases.

We describe a mesothelial neoplasm of the tunica vaginalis called as well differentiated papillary mesothelioma of tunica vaginalis. No complex architecture with stromal invasion or coagulative necrosis or increase in the mitotic activity is seen in our case. Sometimes WDPM may be confused with carcinomatosis on surgery, but can be readily distinguished microscopically. Although our case had no invasion, atypia, infiltrative growth or mitotic activity, we recommended close patient follow up because of the association of the multiple nodules in the hydrocele sac showing the same morphological features. The biological behavior of WDPM is not well known because of the limited number of cases.

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