CASE REPORT

MYXOID ADRENAL ADENOMA WITH FOCAL PSEUDOGLANDULAR PATTERN

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ABSTRACT

Adrenal cortical tumors with myxoid change are rare tumors. To our knowledge, only 22 cases have been described so far in literature, which include 13 carcinomas and 9 adenomas. A pseudoglandular pattern has been described in 9 of these tumors. We report a case of a myxoid adenoma of the left adrenal gland in a 67-year-old woman, with a focal pseudoglandular pattern involving about 20% of the studied tumor. Rest of the tumor was composed of anastomosing cords of tumor cells. Abundant myxoid stroma was present, which stained positively with alcian blue and was weakly focally positive with periodic acid Schiff. Immunophenotype was consistent with an adrenal tumor, i.e., positive for vimentin, inhibin, and melan A. Cytokeratin AE1/AE3 and chromogranin were negative. MIB-1 index was <0.1%.

Key words: Adenoma, adrenal, myxoid, pseudoglandular, tumor

INTRODUCTION

Majority of adrenal cortical adenomas occur in adults. Architectural patterns commonly seen are trabecular (or short cords) and alveolar with rounded nests of cells. We report a case of an adrenal adenoma with abundant myxoid stroma and a focal pseudoglandular pattern. Brown *et al.* reported the first 6 cases of myxoid adrenal adenoma in their series of 14 myxoid adrenal tumors, with a pseudoglandular pattern in 4 of these cases.[1] Subsequently, 3 myxoid adrenal adenomas have been reported, with a pseudoglandular pattern in 2 of them.[2-4]

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CASE REPORT

An adrenal tumor was incidentally detected on abdominal ultrasound examination in a 67-year-old woman during a routine health checkup. She had a history of mild hypertension since 10 years. There was no history of abdominal discomfort or any other complaints. On examination, the blood pressure was 160/90 mm Hg. CT study showed a large, well-defined mixed-density Space occupying lesion occupying left suprarenal region. Post–contrast enhancement study showed variable enhancement. Delayed scans showed

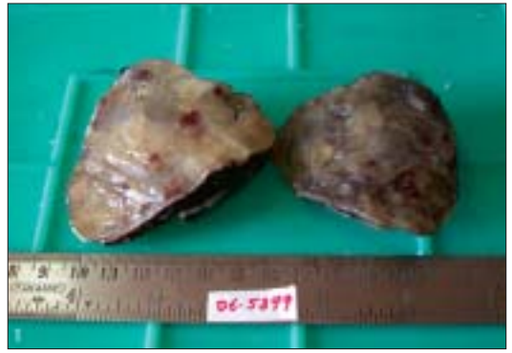

Figure 1: Gross: Tumor with mucoid and glistening cut surface

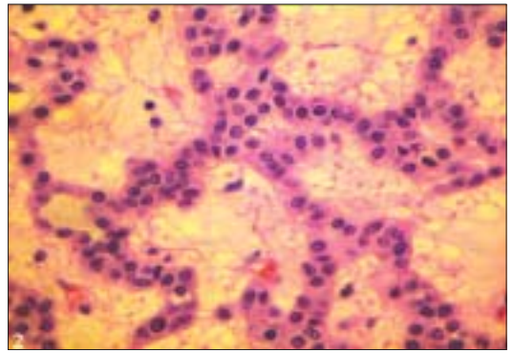

Figure 2: Myxoid stroma with cords of neoplastic cells (H and E, ×10)

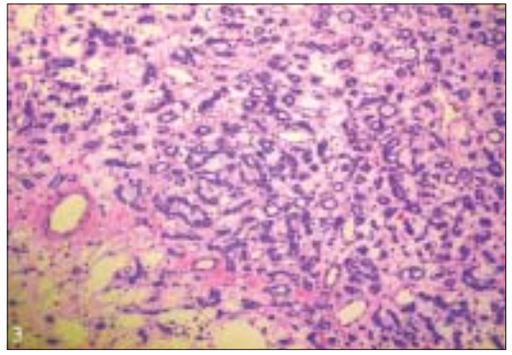

Figure 3: Neoplastic cells arranged in pseudoglandular pattern (H and E, ×10)

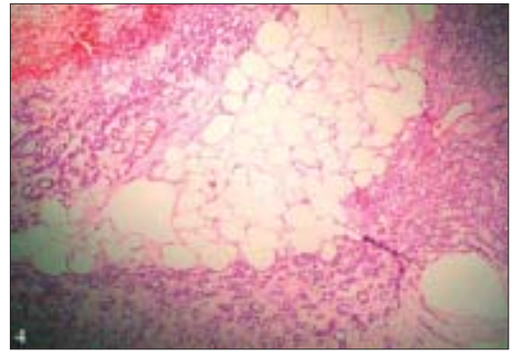

Figure 4: Focal lipomatous metaplasia within the tumor (H and E, ×10)

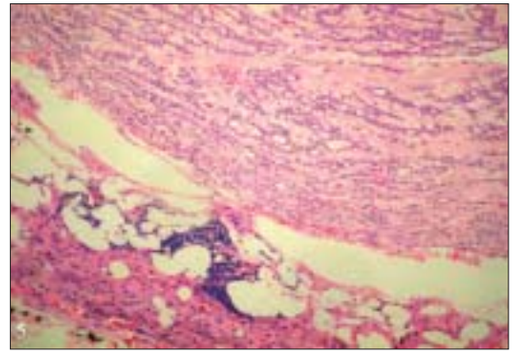

Figure 5: Rim of adrenal tissue showing myelolipomatous metaplasia (H and E, ×10)

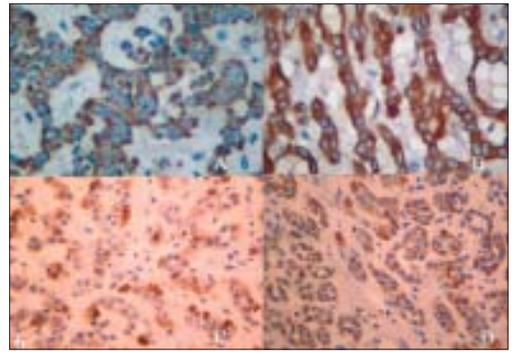

Figure 6: Immunohistochemistry: Positivity with vimentin (A), synaptophysin (B), inhibin (C), melan A (D)

low-density areas centrally, with uniform density in rest of the SOL. Laparoscopic left adrenalectomy was done.

Pathologic findings

Gross: The resected adrenal gland measured 8×7×5.5 cm and weighed 120 gm. On sectioning, the adrenal gland was entirely replaced by a well-circumscribed tumor with a mucoid and glistening cut surface [Figure 1]. Normal adrenal gland was not recognized grossly. Few areas of hemorrhage were noted.

Microscopy

The tumor was entirely encapsulated and composed of polygonal tumor cells with uniform round nuclei with fine chromatin and a moderate amount of eosinophilic cytoplasm. Predominant pattern noted was that of thin cords, one-to-two cells thick [Figure 2]. A focal pseudoglandular pattern with cell nests acquiring a distinct central lumen and intraluminal myxoid contents was noted, involving about 20% of the studied tumor [Figure 3]. Extensive myxoid change was noted, which stained positively with alcian blue. Weak focal staining with periodic acid Schiff was present. There was no evidence of nuclear atypia, mitotic activity, necrosis, vascular invasion, or capsular invasion. Focal lipomatous metaplasia was seen within the tumor [Figure 4]. A thin compressed rim of normal adrenal tissue was identified, showing a focus of myelolipomatous metaplasia [Figure 5]. Immunohistochemical examination showed positivity of the tumor cells for vimentin, synaptophysin, inhibin, and melan A [Figure 6]. Chromogranin and cytokeratin AE1/AE3 were negative. MIB-1 index was <0.1%. The features were consistent with an adrenal cortical adenoma with myxoid change.

DISCUSSION

Myxoid tumors of the adrenal gland are rare. Tang *et al.* reported the first case of a myxoid adrenocortical carcinoma in 1979[5] Subsequently, 22 cases of myxoid adrenal tumors have been reported, 12 of which were carcinomas[6-9] and 9 were adenomas.[1-4] Fine *et al.* described a myxoid adrenal tumor which was difficult to classify as either benign or malignant.[10] These reports indicate that myxoid change did not have any effect on the clinical behavior. Another unusual histologic feature seen was the focal pseudoglandular pattern. The usual architectural patterns noted in adrenal adenomas are trabecular (or short cords) or alveolar with rounded nests of cells. A pseudoglandular pattern has been described in 9 of all myxoid adrenal tumors (carcinomas and adenomas) described so far.[1,3,4,10] It could be possible that myxoid change promotes the formation of pseudoglandular structures, since it has not been described in the conventional adrenal tumors. Small foci of lipomatous metaplasia were noted, a feature which has been described in adrenal adenomas.[11] Myelolipomatous metaplasia in the residual adrenal gland was also present. Alcian blue staining highlights the extracellular acidic mucosubstances in all reported myxoid adrenal tumors, whereas focal Periodic acid schiff positivity has been reported only in few cases.

Immunohistochemistry studies revealed positivity for inhibin, synaptophysin, and melan A. Inhibin is a dimeric 32 kDa peptide hormone that is composed of an α subunit and a β subunit. The value of inhibin in the diagnosis of ovarian sex cord stromal tumors has been established. The vast majority of adrenal tumors also stain positively for inhibin.[12] A103 is a monoclonal

antibody directed against an antigen on melanoma cells recognized by T lymphocytes termed melan A or MART-1.5 Although this antibody has been most commonly used as a marker for malignant melanoma, it has also been proved to be positive in adrenocortical tumors.[13] Vimentin is commonly expressed in adrenal tumors and was positive in our case. Stainings with cytokeratin (AE1/AE3) and chromogranin were negative. The findings were similar to those described by Brown *et al.* in their series of 14 cases.[1]

The differential diagnosis between benign and malignant can be a difficult task. Several multiparametric systems have been published so far.[14,15] The Weiss system appears to be most utilized due to its simplicity and reliability. Our case did not reveal any of the features of malignancy.

To conclude, we have reported a case of an adrenal adenoma with extensive myxoid change and focal pseudoglandular pattern. Although rare, myxoid adrenal tumors must be considered in the differential diagnosis of retroperitoneal myxoid tumors. An immunohistochemical profile which includes inhibin and melan A will aid in the diagnosis.

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