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EDİTÖRE MEKTUP / LETTER TO THE EDITOR

Neuroendocrine tumor arising in a retroperitoneal recurrent mature cystic teratoma

Retroperitoneal rekürren matür kistik teratom zemininde gelişen nöroendokrin tümör

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Dear Editor,

Mature cystic teratomas are composed of three germ layers. Particularly, teratomas are located in gonads but extragonadal involvements may be seen[1]. Retroperitoneal teratomas are 4% of all teratomas and 5-10% of all retroperitoneal tumors[2]. The presented case is an extremely rare retroperitoneal recurrent mature cystic teratoma associated with neuroendocrine tumor.

The 40 year-old female was admitted to the hospital with abdominal pain at 2009. The pelvic examination was revealed a retroperitoneal mass. The specimen was composed of multilayered squamous epithelium, follicular epithelium, muscular tissue, adipous tissue. (Figure 1) Microscopic features revealed mature cystic teratoma. Seven years later, the patient was admitted to the hospital with same complaints. Radiologically retrorectal 2,5x2 cm cystic mass was detected on CT. The patient was underwent laparoscopic surgery then laparatomy because of the adhesion of the mass to adjacent tissues. Microscopically the tumor was composed of multilayered squamous epithelium, adipous tissue as mature cystic teratoma and additionally there was a small focus of another neoplasm. (Figure 2,3). This area was 0,9x0,5 cm diameter. The nests were composed of bland looking monomorphic cells with round nuclei, salt and pepper chromatin and eosinophilic cytoplasm. Immunohistochemically, chromogranin, synaptophysin and keratin were positive. (Figure 4). The proliferation index was 1-2% by Ki 67 immunostaining. (Figure 5). Mitotic activity was 1/10 hpf. Necrosis or lymphovascular invasion were not seen. Therefore, the tumor was classified as low grade (Grade 1) NET. Four days after the operation, the patient was discharged from the hospital. At the last control examination, the patient was asymptomatic and disease free for 12 months.

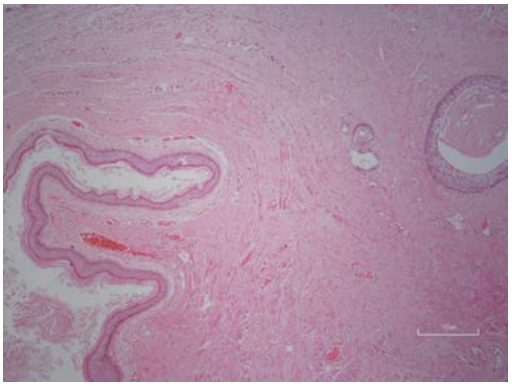

Figure 1. (H&E,X40) Initial biopsy material was composed of multilayered squamous epithelium and skin appendages in this field.

Teratoma is a tumor which contains two or three germ cell layers, endodermal, mesodermal and ectodermal tissues. The mostly accepted theory about teratoma is an origin from the primordial

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germ cell. Retrorectal space is derived from embryonic neuroectoderm, notochord and hindgut, thus a variety of tumors may occur in this region. Extragonadal teratomas tend to occur in midline and retroperitoneal teratomas mostly occur in children than adults[3]. Neonatal teratomas may recur in adulthood. In spite of the initial admission of presented case was at her adulthood, it is recurrred. Malignant transformation has been reported in 0.17% to 3% of gynecologic cases[4]. Most of these malignancies are squamous cell carcinoma which are derived from ectoderm. Malignant transformation interval time is approximately 15 to 20 years and it occurs in postmenoupausal period. Also testicular teratomas may undergo malignant transformation. Neuroendocrine tumors arising in mature cystic teratoma are rarely reported in the literature[5,6]. Particularly these cases are originated from gonadal regions. Primary retroperitoneal teratomas associated with neuroendocrine tumors are extremely rare[7-10] (Table 1). To the best of our knowledge presented case is the fifth case in the literature. The diagnosis of neuroendocrine tumor associated with mature cystic teratoma is important because of its low malignant potential. Pathologic examination is the gold standart to detect this neoplasm, because it is difficult to detect before progression.

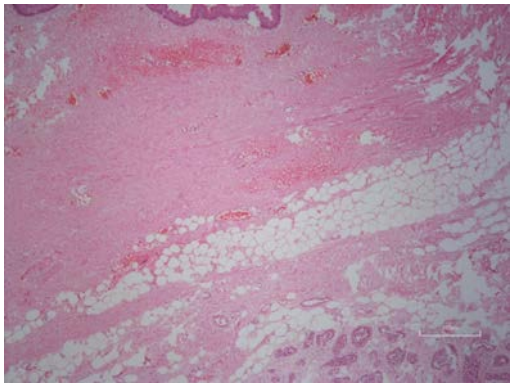
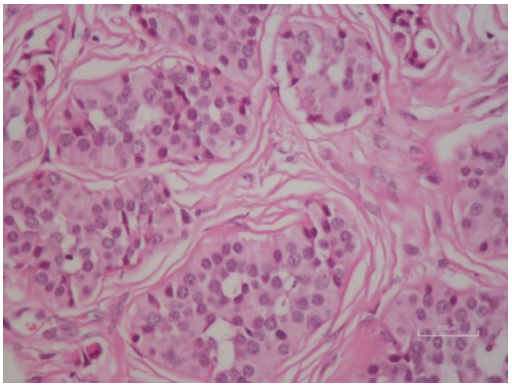


Figure 2. (H&E,X40) The recurrent tumor was composed of multilayered squamous epitheium, adipous tissue, fibrous tissue and small focus of neuroendocrine tumor (at the bottom)



. Figure 3. (H&E,400) The neoplastic cells were composed of small round cells with salt and pepper chromatin, eosinophilic cytoplasm. Neuroendocrine tumor.

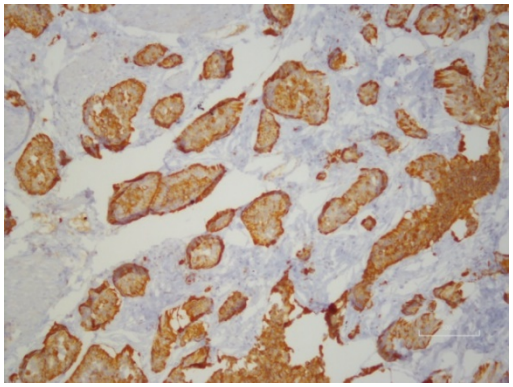


Figure 4. (X100) Immunohistochemically strong membranous, cytoplasmic chromogranin positivity.

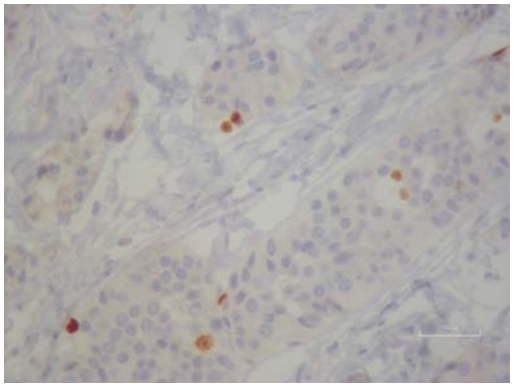


Figure 5. (X400) Immunohistochemically Ki 67 proliferation index was 1-2% at high power.

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