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Immature Testicular Teratoma Post-operatively Presenting as Chemotherapy Resistant Retroperitoneal Mature Teratoma

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Abstract:

Introduction: Growing teratoma syndrome (GTS), first described in 1982, presents as newly growing metastatic masses in patients previously treated for immature teratoma. Since 1994, only 13 cases have been reported in India. *Case Report:* A 21 year old male who had undergone orchidectomy for immature teratoma presented with retroperitoneal mass 6 months later. He received 4 cycles bleomycin-etoposide-cisplatin based chemotherapy without change in size of mass. He underwent excision of tumor and retroperitoneal lymph nodes. Histopathology revealed mature teratoma predominantly cartilage, respiratory and enteric epithelium and adipose tissue without immature elements. *Conclusion:* These tumors cause adjacent vascular and organ invasion causing compressive symptoms. GTS mimics tumor recurrence and patients need to be on regular follow up for detecting these tumors, for they are resistant to chemoradiotherapy and are amenable to surgical resection.

Key words: Abdominal Pain, Bleomycin, Nephrectomy, Teratoma, Testicular Neoplasms.

Introduction

"Benign maturation" of teratomas were first described in 1969 [1]. The term growing teratoma syndrome (GTS) was first coined in 1982 by Logothetis *et al*. in their description of six patients with metastatic mixed germ cell tumors (GCT) [2]. The incidence of GTS according to various studies is 1.9%-7.6% [3]. Since 1994, only 13 cases (6 ovarian, 7 testicular) of GTS have been reported from India [4-9]. We report a testicular immature teratoma presenting postoperatively, as a retroperitoneal mass refractory to chemotherapy.

Case Report

This 21 year old male had been referred to the Urology department for the evaluation of a retroperitoneal mass in November 2015. He had undergone treatment for a painless left-sided testicular swelling at another hospital in November 2014; left orchidectomy had been performed, and post-operatively, he had remained asymptomatic for about 6 months. His β-human chorionic gonadotropin (β HCG) and α-fetoprotein (AFP) prior to his surgery in November 2014 were 34.8 mIU/mL (normal <0.8) and 6393 ng/mL (normal

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<50) respectively. He was finally diagnosed to have an immature testicular teratoma [Fig.1a]. Six months postoperatively, he had developed left-sided abdominal pain of a nonspecific nature. There was no abdominal distension or other gastrointestinal complaints. There was no history of hematuria, decreased urine output or fever. He did not have any other chronic illness.

At the time of his visit, he was conscious and oriented, with pulse rate 82/minute, blood pressure 116/80 mm Hg and respiratory rate 18/minute, with otherwise normal general examination. On systemic examination, respiratory and cardiac systems were normal; a firm mass was palpated in the left hypochondrium, umbilical and left lumbar regions. His penis and right testis were normal, while his left-sided scrotum was empty with a surgical scar. His laboratory investigations were as follows: hemoglobin: 12.1 g/dL, total leukocyte counts: $8.63x10^9$/L, platelets: $324x10^9$/L, random blood sugar: 82 mg/dL, urea: 36 mg/dL and serum creatinine: 1.1 mg/dL. Contrast-enhanced computed tomography of the abdomen revealed a large well-defined heterogeneous mass in the retroperitoneum, measuring 16x9x9 cm with specks of calcification. This mass appeared to displace the left kidney superiorly, causing a mild degree of left-sided hydroureteric nephrosis [Fig.1b].

In view of his previous history of immature teratoma, metastasis of immature teratoma was suspected and he was referred to our hospital's Oncology department. Thereafter, he received four cycles of bleomycin-etoposide-cisplatin (BEP) based chemotherapy between September and November 2015. Post-chemotherapy, his β-HCG and AFP were 5 mIU/mL and 131.3 ng/mL respectively. Even after chemotherapy, the size of the mass had remained unchanged (both clinical and radiological) and hence he had been referred to the Urology department. Following evaluation, he was planned for tumor excision. Per-operatively, a retroperitoneal mass encasing the lateral border of the renal artery and vein, aorta, small intestine, colon, gonadal vein, ureter, left common iliac artery and psoas fascia was noticed. Tumor excision, left nephrectomy and aortic resection with aortic graft placement was performed. Histopathological evaluation revealed a mature teratoma. He is still under follow up.

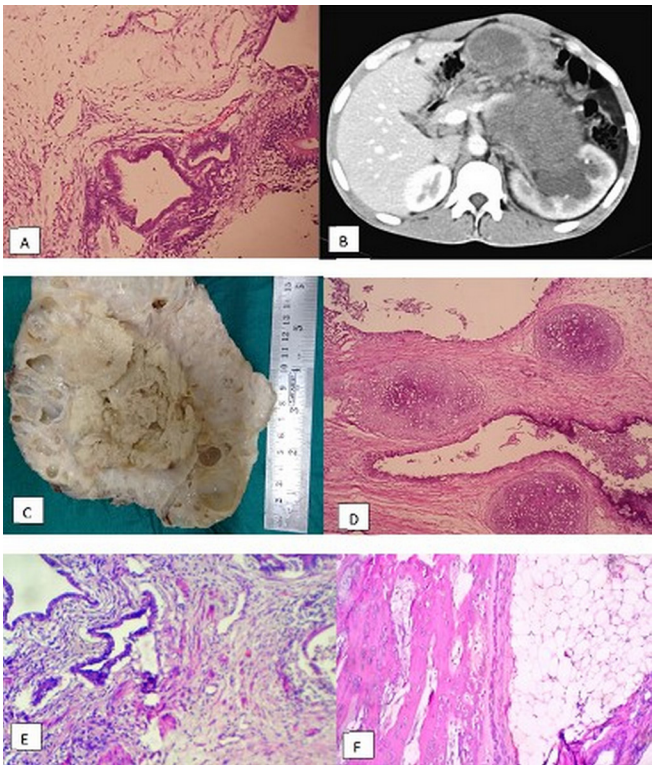

Fig.1(a): Section from testis shows immature neural elements-immature teratoma. **(b):** CECT abdomen revealed retroperitoneal mass adherent to the left renal pelvis. **(c):** Globular mass measuring 18x13x12 cm with cut surface showing solid and cystic spaces with central necrosis and areas of calcification. **(d):** Section from the retroperitoneal mass shows predominantly cartilage and respiratory epithelium. **(e):** Section shows enteric epithelium, muscle and fibro-collagenous tissue. **(f):** Section shows adipose tissue and bony trabeculae.

Discussion

Testicular tumors constitute 1% of all malignancies in males. Among these, 90% are germ cell tumors; half of these are non-seminomatous germ cell tumors (NSGCT) [10]. The usual presentation of GTS is patients with NSGCT, after appropriate chemotherapy and normalization of serum markers, developing metastatic masses [3]. Theories of GTS include: immature malignant elements destroyed by chemotherapy leaving benign mature elements behind; tumor cell kinetics being altered by chemotherapy and totipotent malignant germ cells transforming into mature benign teratoma; spontaneous and inherent differentiation of malignant cells into benign tissues [11]. Another reasonable explanation for GTS is "chemotherapeutic reconversion" of immature teratoma cells into mature tissue [12]. Our patient had a metastatic mass even before chemotherapy but it is also probable that chemotherapy could have also altered the immature elements. Sites for GTS are the retroperitoneum (commonest: 80%), mediastinum, chest, liver, abdomen, sublcavian node, inguinal nodes and the mesentery [1,13].

Three criteria are required for diagnosis of GTS: a patient is receiving or has received chemotherapy, previously elevated serum markers (β-HCG and AFP) become normal, and there is a paradoxical increase in tumor size with absence of immature component [2]. For primary mediastinal NSGCT, Kesler *et al*. proposed that the criteria for GTS also include cardiopulmonary deterioration due to compression of vasculature, heart or lungs [14]. GTS has been commonly reported with the BEP chemotherapy regimen as in our patient [1].

Serum markers for GTS include AFP, β-HCG and lactate dehydrogenase [1]. Radiological exam in GTS reveals masses ranging in sizes between 1 and 25 cm with well circumscribed margins, cystic changes with adipose tissue and calcifications which may be either punctate or curvilinear [1]. Microscopy shows both solid and cystic components and can comprise cartilaginous elements, epithelium (enteric and respiratory), and neurogenic tissue with undifferentiated spindle cell stroma [1]. GTS tumors are chemoradiotherapy resistant and the treatment of choice is surgical excision, as in our case. Complications of GTS occur predominantly due to compression of surrounding structures leading to problems such as hydroureteronephrosis and venous stasis. Our patient had tumor adherence to blood vessels and the left kidney. The fate of GTS, studied in a large French cohort includes relapse of GTS or NSGCT, or second malignancies such as sarcoma, adenocarcinoma, squamous cell carcinoma, PNET, carcinoid and leukemia [15].

Conclusion

GTS is a rare tumor with very few reports from India in the last 22 years. Pathologists, urologists, gynecologists and oncologists need to be aware to prevent misdiagnosis of recurrence, to institute early surgical resection and carefully follow up patients with immature teratoma for detecting new metastatic tumors.

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