An Atypical Case of Recurrent Benign Retroperitoneal Teratoma in an Adult Patient

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Abstract: We report a case of a forty-five-year-old gentleman presenting with recurrent abdominal swelling within 18 months of undergoing laparotomy with complete excision of benign teratoma, eventually diagnosed again to be mature benign retroperitoneal teratoma. The diagnosis along with its mode of presentation, short interval of recurrence and absence of several characteristic associated features or raised serum AFP constitute a rarity in this age group.

Keywords: Primary retroperitoneal teratoma, benign teratoma, recurrent teratoma, teratoma in adult.

INTRODUCTION:
Benign retroperitoneal teratoma, a subtype of germ cell tumour, is not only uncommon in a forty-five-year-old male subject but also is a rarity since this is recurrent within one and a half years. It is a surgically challenging case too when it’s found very close to kidney and major vessels like external iliac vessels. There are few case reports of recurrent retroperitoneal teratoma but that recurring within 18 months of complete excision without many characteristic associated features and coming out to be benign yet again is extremely unique in this age group. Hence we are reporting this case.

Teratomas are congenital tumours arising from pluripotent embryonic cells with tissue or organ components resembling normal derivatives of all three germ layers and therefore have several recognizable somatic tissues [1]. Generally arising from the gonads, they may be found in extra-gonadal sites such as sacrococcygeal region, mediastinum, neck and retroperitoneum [2, 3].

Teratomas constitute less than 10% of all primary retroperitoneal tumors and hence are relatively uncommon [4]. Retroperitoneal teratomas comprise 3.5 – 4% of all germ cell tumors in children [5]. Retroperitoneal teratomas are seen in females twice more commonly than males.

The symptoms can manifest themselves by abdominal pain, mass or swelling, and abnormal uterine bleeding [6]. Bladder symptoms, gastrointestinal disturbances, and back pain are less frequent. Teratomas are usually benign if they are cystic and contain sebum or mature tissue.

CASE REPORT:
A forty-five-year-old gentleman presented with a history of an intra-abdominal swelling for the last 6 months. The swelling was gradually increasing in size. There were no associated features like pain, fever, vomiting, bowel habit abnormalities and melena. He had a past history of abdominal surgery for an abdominal lump 18 months ago, histopathology report of which confirmed it to be benign teratoma.

General survey was within normal limits. Abdominal examination revealed a firm, immobile, non-tender swelling in the left lumbar region and left iliac fossa. Intestinal peristaltic sound (IPS) was present. Both testes found present in scrotum. Routine blood parameters (e.g. complete hemogram, electrolytes), tumor markers (e.g. serum alpha fetoprotein) and other preoperative investigations like plain abdominal X-ray were found unremarkable.

Ultrasoundography demonstrated a left sided 13 x 9.6 cm soft tissue mass in the retro-peritoneum. It was heterogeneous, well circumscribed with sharply defined borders and cystic areas. Contrast enhanced CT scan showed a left sided retroperitoneal SOL (13.2 cm x 10.1 cm) having a variegated (hypo-dense areas with fatty attenuation) appearance along with hydronephrotic changes in left kidney. Digital IVU (Intravenous Urogram) showed non-visualization of left kidney, even after 50 minutes. DTPA Renogram showed less than 10% function in left kidney.

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Patient was adequately prepared. Exploratory laparotomy performed and the mass was found very close to kidney and major vessels like external iliac vessels. Kidney was not found viable also on per-operative inspection. Excision of the tumor along with left nephrectomy done. After removal of the tumor, the tumour bed was found having major vessels forming its medial boundary. Hemostasis was secured and abdomen closed in layers after placement of an intra-abdominal drain.

Post operatively this tumor mass was excised in vitro and found to be filled with yellowish creamy material. Histopathology examination confirmed it to be a case of benign mature cystic Teratoma. No evidence of immaturity or malignancy was found.
DISCUSSION:
The chief objective of outlining this case report was to highlight the rarity of a cause one can encounter while dealing with asymptomatic recurrent abdominal mass. In addition there are few peculiarities in this case which attracts attention. Retroperitoneal teratomas are uncommon germ cell tumours. Furthermore, retroperitoneal teratomas occur mainly in children and have been very rarely described in the adults [5]. Half of these cases present in children less than 10 years of age and only a fifth of them present after 30 years of age [7]. Here, we have a case of recurrent retroperitoneal teratoma in an adult forty-five-year-old gentleman.

The incidence of teratoma in females is much higher than in males [7], yet we find a male patient here which is uncommon. Serum AFP assay is a reliable method for detecting the recurrence in teratoma [8]. However, here serum AFP was not found raised, another peculiarity in this case. Other serum markers like LDH (Lactate dehydrogenase) and β-HCG (Human Chorionic Gonadotropin) were also measured but none of them came out to abnormal.

Ultrasound and computed tomography are important in its diagnosis and may show the presence of calcification, teeth or fat. Calcification on the rim of tumour or inside the tumour is seen in 53-62% of teratomas and although radiologically three quarters of patients with a benign teratoma may have calcification within it, a quarter of malignant cases may also demonstrate calcification. Computed tomography is better than Ultrasonography in defining the extent and spread of teratoma to the surrounding organs [9]. In this case, neither any calcification nor any tissue like hair or bone was found inside the tumour.

Retroperitoneal teratomas typically are found in midline or paraxial region with preponderance to the left [7]. Teratomas are usually benign if they are cystic and contain sebum or mature tissue. Both of these features were present in our case. Treatment of a teratoma consists of conservative surgical removal, a procedure which often requires finesse and delicacy, which was particularly imperative in this case because of the close proximity to important anatomic structures like external iliac vessels. With conservative removal, occasional recurrence is to be expected but it’s rare after complete removal. Our patient underwent a complete excision of tumour during abdominal surgery 18 months ago, yet it recurred, which is unusual due particularly to the short interval.

Papanicolaou and Yoder [10] advocate angiography, inferior venacavography and needle biopsy for the accurate diagnosis of these tumours, which were not feasible and not done in this case. The
prognosis is excellent for benign retroperitoneal teratoma if complete resection can be accomplished. But particular care should be taken regarding spillage as it is associated with increased risk of chemical peritonitis (estimated incidence of 0.2%) and increased risk of adhesion formation. The risks of recurrence (4%), as well as malignant degeneration (0.2-2%), should also be discussed [11].

CONCLUSION:
This case presented with a unique dilemma of a recurrent abdominal swelling in an adult patient following complete surgical resection of benign teratoma within a span of 18 months, yet unaccompanied by many characteristic features. CT features and DTPA renogram report warranted repeat surgical intervention. Exploratory laparotomy revealed a left sided mass close to hydronephrotic left kidney and external iliac vessels which rendered the case surgically challenging but ultimately repeat complete excision was possible. Postoperative period was uneventful with a good recovery.

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