Case Report:
Ovarian Growing Teratoma Syndrome Presenting as Sciatica.

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Abstract: Growing Teratoma Syndrome (GTS) is an unusual sequela of nondysgerminomatous germ cell tumours. Here, we report the case of a lady with GTS presenting in a unique manner, as sciatica. The predisposing factors for development of GTS are advanced nature of primary tumor, mature elements in primary tumor and incomplete primary surgery. Prompt and timely excision of the tumour results in complete excision and symptomatic relief, in addition to confirmation of diagnosis and exclusion of malignancy.

Key Words: Growing Teratoma Syndrome; Chemotherapeutic Retroconversion; Sciatic; Dermoid.

Case Report:
A 25 years old lady was referred to our department with histopathological diagnosis of immature teratoma of the ovary. She had undergone suboptimal surgery elsewhere, including sub-total hysterectomy and ovarian tumor biopsy one month back. Tumor markers’ profile revealed an elevated LDH value of 1232 IU and CA-125 of 125 IU, with normal levels of AFP and hCG. As she had acute deep vein thrombosis of left leg at presentation, we decided to give her one cycle of neo-adjuvant chemotherapy with BEP (Bleomycin, Etoposide, Cisplatin) regime prior to surgery along with thrombolytic therapy. Following one cycle, she underwent optimal cytoreductive surgery, with bilateral ovariectomy, total omentectomy, excision of cervical stump and diaphragmatic deposit excision, with no gross residual disease. Final histopathology revealed grade 1 immature teratoma bilateral ovaries with immature neuroepithelial elements, with metastasis along with mature glial elements in omentum and diaphragmatic deposits, indicating stage IIIc disease. She received two adjuvant cycles of BEP regime uneventfully with falling values of LDH. During her third adjuvant cycle, she developed severe late blood transfusion reaction on the tenth day of chemotherapy, and further chemotherapy withheld. At time of termination of therapy, LDH had normalised at 117 IU, and there was no clinical or radiological evidence of disease. Patient was put on follow up.

Fourteen months after termination of chemotherapy, the patient reported with low back ache along with sciatica like pain along the back of her left thigh increasing on walking. An abdominopelvic scan showed a cystic lesion of 8cm x 8cm resembling a lymphocyst, with normal tumor markers. She was advised symptomatic management with NSAID’s and follow up after three months. Two successive three monthly scans revealed the same mass with no increase in size or tumor markers. However, her sciatic pain progressively increased in duration and in intensity from an initial NRS-11 of around 2 to a score of 8, now extending down to the foot. In view of the debilitating nature of her symptom, surgery was contemplated, and a computerised tomogram of abdomen and pelvis obtained. CT scan showed a heterogenous enhancing soft tissue mass lesion with specks if calcification in the left pararectal region abutting the rectosigmoid and the left psoas muscle, in close proximity to bowel loops and compressing the left internal iliac vessels (Figure 1). Exploratory laparotomy revealed a hard irregular well circumscribed mass of 8cm x 8cm x 6 cm impacted in the left pararectal space retroperitoneally displacing the left ureter laterally and compressing the left internal iliac vessels just below the bifurcation of the common iliacs (Figure 2). Due to the bony nature of the mass and difficulty in handling with conventional instruments, a bone holder was used to grip the mass to deliver it out (Figures 3,4). The mass was completely excised intact, causing a small tear in the internal iliac vein which was sutured appropriately (Figure 5). Intraoperative
frozen section examination showed only mature cartilaginous elements with no immature elements. She was discharged on the sixth post operative day. The final histopathology report read “tissues derived from all three germ cell layers with islands of mature cartilage predominantly, and no immature or malignant elements” (Figure 6). At the time of this report, patient is in the third post operative week and recuperating well, with complete relief of sacral pain (NRS-0).

Discussion
GTS is defined as the continuing clinical or radiological enlargement of existing tumor or development of new tumor with normal tumor markers in a patient with NSGCT after or during chemotherapy, and the final histopathology of the resected tumor revealing no NSGCT other than mature teratoma.[1] Our patient was initially taken up for surgery with provisional diagnosis of symptomatic recurrence as tumour markers are less sensitive in immature teratoma. However on final histopathology report she was found to fulfil all the criteria for the diagnosis of growing teratoma syndrome.

GTS has been reported to occur in testicular NSGCTs at the rate of 1.9-7.6%, and is now being increasingly reported in ovarian germ cell tumors as well.[2] Though the phenomenon
of GTS was first described by Logothetis in non seminomatous testicular germ cells, a similar phenomenon of conversion of immature ovarian teratoma into mature teratoma was described by Di Saisa et al, which the latter termed “chemotherapeutic retroconversion.”[3] Current literature shows that the two phenomena are one and the same.[4] Some authors state that GTS occurs due to selective destruction of the immature elements by chemotherapy leaving behind only the mature elements, while others argue that chemotherapy alters the cell kinetics of the malignant cells.[5] Hong et al proposed that chemotherapy prolongs the survival of the patient, allowing time for “spontaneous differentiation” of the immature cells into mature cells. We are more in favour of the third theory in our case, assuming that there might have been peritoneal dissemination and micrometastases at the time of the first suboptimal surgery when the tumour was also biopsied. Zagame et al suggested that possible predictors of GTS may be the presence of mature elements in the primary tumor, predominance of immature neuromepithelial tissue and stage III disease with peritoneal spread or incomplete removal with residual disease prechemotherapy.[6] In this case, all the above mentioned factors, with the exception of incomplete cytoreduction, were present.

Timing of diagnosis of GTS has varied from as early as during the early courses of chemotherapy to as late as 12 years post-chemotherapy.[7] Djordjevic et al observed that that most ovarian GTS tend to appear for the first time within 2 years of the initial primary and remain confined almost exclusively to the pelvis, abdomen, and the retroperitoneum and do not venture to distant systemic sites.[8] The latters’ observations hold good in our case too. GTS is associated with a 3% risk of malignant transformation, into immature teratoma, sarcoma, adenocarcinoma, squamous cell carcinoma, or primitive neuroectodermal tumor.[9,10] GTS is also known to cause pressure effects on adjacent organs and neurovascular structures in 12% cases due to rapid growth.[9] However, extensive literature search yielded no other reports of GTS causing sciatica.

GTS requires complete surgical excision both to prevent and to exclude malignancy, in addition to relieving mechanical symptoms. Callaghan et al recommend that surgery be planned as early as possible once GTS is suspected, before the tumor becomes difficult or potentially impossible to operate upon because of operative risks to adjacent organs.[11] In the present case also, we encountered a minor vascular injury due to close proximity of the tumor to vessels. To enable diagnosis at the earliest, Spiess et al recommend regular imaging and monitoring of tumor markers after chemotherapy for the primary tumor.[7] Most reports indicate that no further treatment is needed. However, Djordjevic reported the exceptional case of a young girl in whom mature teratomas recurred at 1, 6, and 19 years after optimal surgery and chemotherapy for immature teratoma. Possibly in the same light, Hsieh et al have recommended close follow-up with tumor markers and imaging studies as stabilization of GTS can take as long as 10 years after diagnosis.[12] The patient in our case is currently asymptomatic, and has been advised three monthly follow up with LDH and abdomino-pelvic ultrasonogram.

**Conclusion**

Though a rare complication of non dysgerminomatous germ cell tumours, the GTS needs special mention in view of its occurrence mimicking recurrence, the debilitating nature of symptoms and the possibility of a late malignant transformation. The above stated factors also necessitate surgical excision, which if performed early in the course of the tumor, can avoid further surgical damage. Post excision, the condition also warrants close follow up with tumor markers and imaging.

**References**