Case Report:
A Rare association of Trousseau’s Syndrome with Ovarian Serous Cystadenocarcinoma

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Citation

Open Access Archives
http://cogprints.org/view/subjects/OJHAS.html
http://openmed.nic.in/view/subjects/ojhas.html

Submitted: Sep 6, 2015; Accepted: Sep 23, 2015; Published: Oct 15, 2015

Abstract: Trousseau’s syndrome is defined as a migratory thrombophlebitis found typically in patients with an underlying malignancy. This is often associated with GIT malignancies. Limited literature is available showing its association with ovarian malignancies and there is no available report showing occurrence of thrombosis in Internal jugular, subclavian & axillary veins with serous cystadenocarcinoma of ovary.

Key Words: Internal jugular vein thrombosis, Trousseau’s Syndrome, Serous cystadenocarcinoma.

Introduction:
Trousseau’s syndrome is defined as migratory thrombophlebitis found typically in patients with an underlying malignancy. Its association with ovarian malignancy was first reported by Womack and Castelenno in 1952 with papillary cystadenocarcinoma of ovary. Literature search did not reveal any case report of Trousseau’s syndrome with serous cystadenocarcinoma of ovary. Moreover, occurrence of thrombosis in Internal Jugular Vein, subclavian & axillary veins are rare and we found only one case report of internal jugular thrombosis with ovarian malignancy. In our case, thrombosis involved Internal Jugular vein, subclavian & axillary vein which were detected first and further evaluation revealed associated ovarian malignancy.

Case Report
A 33 yrs old female presented with complaints of cough, chest pain & breathing difficulty. Respiratory examination showed bilateral crepts & decreased air entry on right side. PA view of chest showed bilateral pleural effusion. Pleural fluid aspiration was done. Pleural fluid showed exudative fluid with lymphocytosis with no evidence of malignant cells. She was put on ATT. But symptoms were not relieved and she complained of irregular fever, swelling on the left side of neck which she noticed after vomiting. Swelling was soft in consistency.

She was advised ultrasonography for neck swelling which revealed non compressible dilated left internal jugular vein with thrombosis within it, extending distally to involve left subclavian, axillary and external jugular vein with no flow on colour Doppler study (Fig-1).

Further scanning of the abdomen and pelvis showed a large predominantly solid heterogeneous right ovarian mass of size 7.0x4.5cms with few cystic areas within it and mild ascites. Doppler study of the mass showed increased vascularity with spectral wave pattern suggestive of malignancy. Rest of the abdominal organs were normal. Radiological diagnosis of internal jugular vein thrombosis with ovarian malignancy was made.

Gynaecological evaluation revealed firm suprapubic mass. PV examination could not assess size of uterus & both fornices felt hard.

Patient was advised CECT abdomen which showed predominantly solid, mildly enhancing right ovarian mass (Fig-2) with mild ascites, bilateral pleural effusion & multiple enlarged retroperitoneal lymph nodes.

Fig 1: HRUSG of neck showing thrombus
vein to the junction of external and internal jugular vein. This is important to know the prognosis. The rate of incidence of pulmonary embolism is 0.5% for isolated internal jugular thrombosis and 2.4% for combined Internal Jugular and subclavian / axillary vein thrombosis. Mortality rates at 1, 3 and 12 months have been reported to be 14%, 33% and 42% respectively.[5] 

Once the diagnosis of Trousseau's syndrome is made, most patients must continue to receive heparin therapy for the rest of their lives to prevent new thrombotic events. However in our case, there was dramatic resolution of thrombus after surgery and chemotherapy with no recurrence of thrombosis. However in our case, there was dramatic resolution of thrombus after surgery and chemotherapy with no recurrence of thrombosis without continuing antithrombotic agents even after one year of follow up. Thrombotic episodes may cease in a rare patient in whom a localized tumor can be resected.[6] 

Of all the modalities to diagnose Internal Jugular vein thrombosis, USG, CT and MRI were considered to be better because of their non-invasive nature and as they can also give additional information about the cause such as extrinsic compression or invasion by malignancy of surrounding structures. USG with Doppler is the recommended imaging modality as it is useful for diagnosis as well as for monitoring the progression of thrombus non-invasively.

**Conclusion**

This rare and potentially fatal condition of Trousseau’s syndrome, illustrating the occurrence of internal jugular vein thrombosis in association with serous adenocarcinoma of the ovary, successfully treated by resection of ovarian mass followed by chemotherapy, sets an example that such a case can be managed successfully without causing mortality, if recognized early.

**References**


**Discussion:**

Armand Trousseau[1] was the first to describe the association between migratory thrombophlebitis and visceral malignancy in1865. He said thrombophlebitis is often the initial sign of underlying malignancy.

Interestingly, Trousseau himself subsequently presented with the syndrome he described and died of gastric carcinoma. Callender N and Rapaport concluded that Trousseau’s syndrome result due to the expression of tissue factor on the tumour cells and also due to the exposure of cells or vesicles shed from the tumour to the circulating blood directly or indirectly.[2]

Malignancies most commonly associated with Trousseau’s syndrome include those of the pancreas, lung, prostate, stomach, colon and ovary. Rarely, it can be the first clinical diagnosis when a patient presents with a neck swelling as reported by Lam WM and colleagues in a case of metastatic adenocarcinoma of unknown primary.[3] Our case presented with neck swelling which was later diagnosed as having malignancy in the form of Serous Adenocarcinoma of Ovary. In a similar case reported by Ball and colleagues, the patient had advanced ovarian malignancy, coagulopathy and insertion of a central line into the internal jugular vein and hence the cause of internal jugular vein thrombosis was not clear.[4]

Internal Jugular vein thrombosis refers to an intraluminal thrombus occurring anywhere from intracranial internal jugular vein to the junction of the internal jugular and Subclavian vein to form the brachiocephalic vein.[5] In the present case the thrombus extended from the left Subclavian

**FNAC from neck swelling showed predominantly red cells with fair number of degenerated cells. CA-125 was significantly raised (2845 IU). She went for bilateral salpingo-oophorectomy. Peroperative findings revealed right Ovarian Tumour involving the POD & Rectum posteriorly. Post-operatively she was given LMW heparin, early ambulation & calf exercises. Histopathological examination of excised sample showed Poorly Differentiated Serous cystadenocarcinoma with involvement of surface of ovary by the tumour and rupture of capsule.**

After that she was referred to oncology dept where she was put on chemotherapy with BEP regimen (Bleomycin, Etoposide, and Cisplatin) which she tolerated without any side-effects. All other haematological parameters were normal. Repeat Doppler examination showed resolution of thrombus. She completed 6 cycles of chemotherapy with mild side effects. Now she is doing well with no evidence of further fresh thrombosis.

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