MEDICAL SCIENCE

A rare case of acute pulmonary embolism in a postoperative case of dysgerminoma in a young adolescent girl: Lesson learnt

Kamlesh Chaudhari[1], **Apoorva Dave**[2], **Deepti Shrivatsava**[1], **Tanvi Chaurasia**[3]

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Authors' Affiliation:
[1]Professor, Department of Obstetrics and Gynaecology, Jawaharlal Nehru Medical College, Datta Meghe Institute of Higher Education and Research (Deemed to be University), Wardha, Maharashtra, India
[2]Assistant Professor, Department of Obstetrics and Gynaecology, Jawaharlal Nehru Medical College, Datta Meghe Institute of Higher Education and Research (Deemed to be University), Wardha, Maharashtra, India
[3]Junior Resident, Department of Obstetrics and Gynecology, Datta Meghe Institute of Medical Sciences, Wardha, Maharashtra, India

ORCID List

Kamlesh Chaudhari	0000-0002-1673-7467
Apoorva Dave	0000-0001-8400-4241
Deepti Shrivatsava	0000-0003-2058-9476
Tanvi Chaurasia	0000-0002-7570-8236

Contact List

Kamlesh Chaudhari	dr.kamleshchaudhari@gmail.com
Apoorva Dave	drapoorvadave@gmail.com
Deepti Shrivatsava	deepti_shrivastava69@yahoo.com
Tanvi Chaurasia	tanvichaurasia001@gmail.com

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ABSTRACT

We present a rare case of pulmonary angiography evidence of pulmonary embolism in an operated case of dysgerminoma in a young adult. Malignant germ cell tumor of the ovary constitutes less than 10% of total ovarian tumors. In young girls malignant primitive germ cell tumour that is most prevalent is dysgerminoma, which has low potential to invade and spread and is treatable when diagnosed early. Thromboembolism in childhood and adolescent age group is not so common. Here, we discuss an unusual form of ovarian dysgerminoma diagnosed intraoperatively on frozen section with post-operative pulmonary embolism with coagulation parameters being normal post operatively. According to our knowledge, this is rarest case where post-operatively patient developed acute pulmonary embolism. We have also reviewed various articles on dysgerminoma and thromboembolism.

Keywords: Dysgerminoma, ovarian germ cell tumors, pulmonary thromboembolism

1. INTRODUCTION

A cancerous tumour that develops from the ovarian primitive germ cells is called an ovarian dysgerminoma (OD). World Health Organization (WHO) defines them as tumors comprising of primordial type of germ cells without specific differentiation pattern (Kaur, 2020). Approximately 0.9 to 2% of all ovarian cancers are ovarian dysgerminoma and is nearly 50% of ovarian cancerous germ cell tumours (33–37%) (Zogbi et al., 2018). In a United State based survey done from the year 1973 to 2002, total 1262 cases of malignant germ cell tumor of the ovary were registered, the incidence of dysgerminoma of the ovary was 0.109 per 100,000 women-years after age adjustment (Smith et al., 2006). Dysgerminoma is the ovarian counterpart of testicular seminoma. Germ cell tumors can be noted at all the age group females, but it is mostly seen in children and adolescents mostly till first 30 years of life. It is rare to

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find these tumors in the age group of less than five or in the post-menopausal female (Gordon et al., 1981; Andela et al., 2022). In the literature the incidence of dysgerminoma ranges in age from 7 months to 70 years (Shaaban et al., 2014). Approximately 10% of all the cancers developed in women younger than 20 years are dysgerminoma (Susnerwala et al., 1991). There are only two reports in the literature that showed thromboembolism in association with germ cell tumor (Oh et al., 2012). We encountered an interesting operated case of dysgerminoma which developed pulmonary thromboembolism post operatively. Along with this we have reviewed available literature on dysgerminoma and thromboembolism in the present study.

Aims and objectives

To describe a rare instance of acute pulmonary embolism in an operated case of dysgerminoma

To review available literature on ovarian dysgerminoma

2. MATERIAL AND METHODS

In this article we present an unusual instance of dysgerminoma which we lost sadly due to postop venous thromboembolism (VTE). And also, we shall review various articles on ovarian dysgerminoma. Review was undertaken by searching the database using Pubmed, Scopus, EMBASE, Web of science, Science direct etc. various studies describing dysgerminoma of ovary were included in the present study. We searched studies till Jan 2023. The search strategy included key words such as ovarian dysgerminoma, metastasis, tumor markers, thromboembolism etc.

3. CASE REPORT

A 20-year-old unmarried, nulligravida girl referred to our casualty from a private nursing home in view of lump and pain abdomen for 7 days with outside ultrasound suggestive of query malignant ovarian mass and CA 125 being 439U/l on per abdominal examination abdomen was distended (Left>right) up to 24-week size of the uterus occupying left hypo gastric region and left iliac fossa. On admission her general condition was fair with bilateral non pitting pedal oedema present and pallor absent and her pulse rate was 118 bpm for which Tablet Evabredin 5mg twice daily given as advised by physician preoperatively. Her TSH was raised 5.89 for which Tablet Thyroxin 25 mcg before breakfast started. Her Preop investigations revealed Hb 10.2gm%, TLC 8700, Platelet 4lakhs, APTT: 30.7 sec, PT: 13.6, INR 1.15 and her LFT and KFT were within normal limits. Her tumor markers such as beta hCG: 25.70, CA 19-9 39.1, AFP was raised to 7176 AND LDH was also high 1342 (Table 1). And her USG findings were suggestive of A well-defined heterogeneous mass of size 19x10x16cms noted in pelvis. It shows high grade vascularity on Doppler study. It is seen separately from both ovary and uterus, likely query neoplastic or metastatic. Left ovary appears mildly bulky with slight irregular margin. MRI findings were suggestive of heterogeneously enhancing solid, cystic lesion in the pelvis with above mentioned extensions and mass effect most likely to be malignant etiology germ cell tumor with left ovarian cyst and Ascites as depicted (Figure 1). She was planned for exploratory laparotomy.

Table 1 Pre-operative investigations

Test	Report
Hb	10.2gm%
TLC	8700/mm3
Platelet	4 lakhs
APTT	30.7 sec
PT	13.6
INR	1.15
LFT and KFT	WNL
Beta hCG	25.70
CA 19-9	39.1
AFP	7176
LDH	1342
Ultrasound	A well-defined heterogeneous mass of size 19x10x16cms noted in pelvis. It shows high grade vascularity on Doppler study. It is

	seen separately from both ovary and uterus, likely query neoplastic or metastatic. Left ovary appears mildly bulky with slight irregular margin.
MRI	Heterogeneously enhancing solid, cystic lesion in the pelvis with above mentioned extensions and mass effect most likely to be malignant etiology germ cell tumor with left ovarian cyst and ascites.

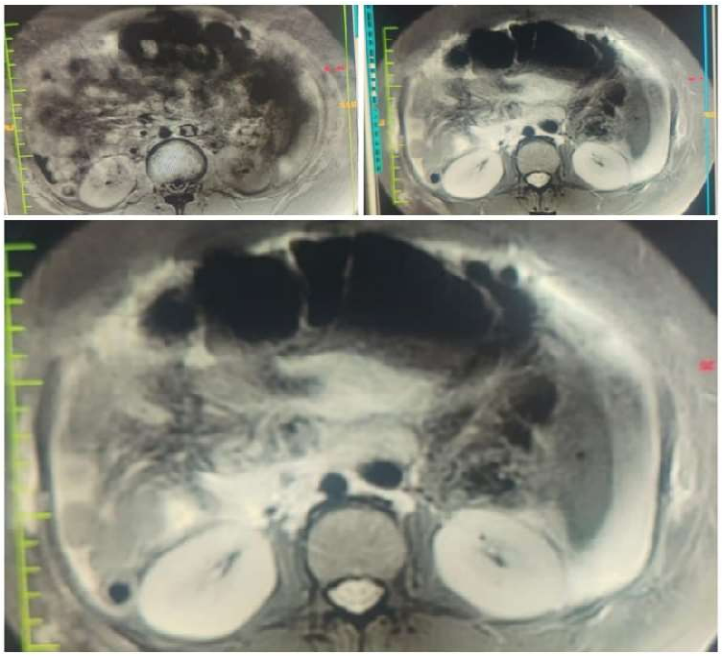

Figure 1 MRI appearance of the ovarian mass

Intra operative findings

After obtaining informed valid written consent patient was taken for exploratory laparotomy and proceed. Hemoperitoneum of about 2 litres was drained and sent for cytopathological evaluation. Solid ovarian mass of 15cm X 14 cm. X 10 cm size occupying hypo gastric region and left iliac fossa, covering postero superior region of uterus, arising from left ovary noted as depicted (Figure 2). Left fallopian tube seen with multiple para tubal cysts. Uterus, right fallopian tube and ovary were normal. Exteriorization of tumor mass was done; frozen section was sent for the evaluation which was reported as query malignant germ cell tumor or sex cord stromal tumor and left salpingo-oophorectomy was done right ovary and tubes were not removed as consent was refused by the patient and her relatives. Omental involvement was seen, partial omentectomy was done. A few deposits on right intra peritoneal surface were seen adjacent to intestinal loops as depicted (Figure 3). Intra operative surgery opinion was taken for query peritoneal and intestinal involvement which was opined as no visible metastasis noted from surgery team following which abdominal drain was placed and abdomen closed in layers. Affected ovary, left fallopian tube, omentum and necrotic tissue from tumor mass were sent for histopathological evaluation. 2 units Packed RBCs were transfused intraoperatively.

Histopathology findings

Gross appearance was as depicted (Figure 4).

Histopathological evaluation as depicted in Figure 5, 6 and 7 suggestive of dysgerminoma of ovary.

Necrotic tissue from peritoneum shows infiltration by dysgerminoma.

Omentumal section shows infiltration by dysgerminoma.

Left fallopian tube is free from invasion with Para tubal cysts.

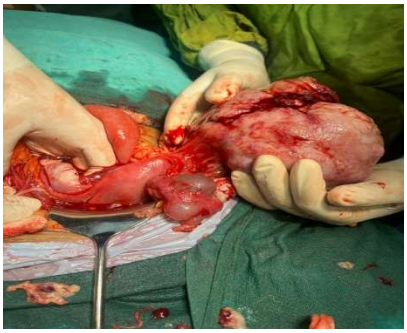

Figure 2 Dysgerminoma arising from left ovary

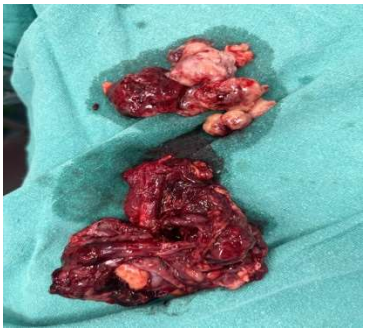

Figure 3 Omental deposits removed in exploratory laparotomy

Cytology

Cytopathological evaluation of hemoperitoneal fluid suggestive of no malignant cells.

Frozen section: Query malignant germ cell tumor or sex cord stromal tumor sertoli-leydig cell tumor

Figure 4 Cut section of the tumor

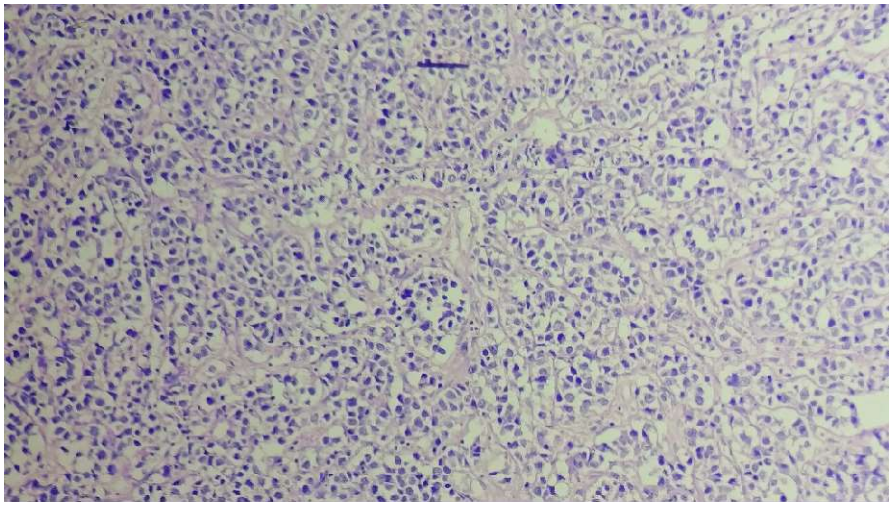

Figure 5 Ovarian dysgerminoma on histopathology

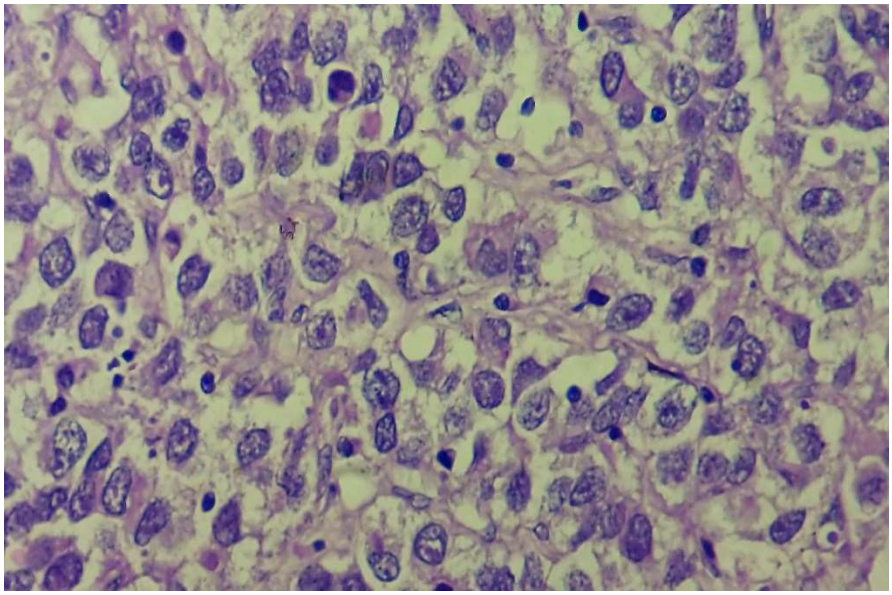

Figure 6 Histopathology of ovarian dysgerminoma

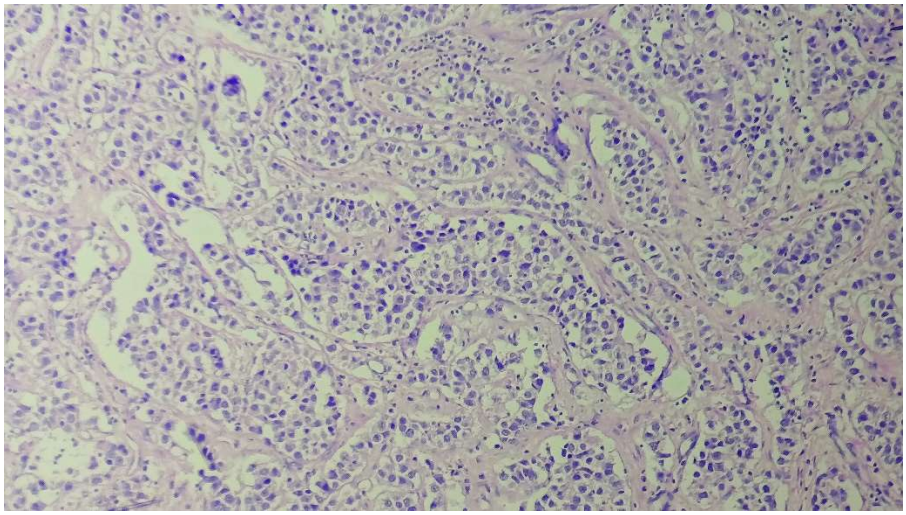

Figure 7 Omental infiltration of ovarian dysgerminoma on histopathology

Post-operative period

Post operatively 3 FFPS and 1 PRBC were transfused.

Her oocyte preservation and plan for contralateral oophorectomy followed by chemotherapy after obtaining her repeat tumor marker values was planned and discussed with the patient and her relatives. Postoperatively for first three days she was fine, post op day1 her Hb was 10.4 gm/dl, TLC was 14600/cumm and platelets was 2.64 l/cumm.

Inj. Enoxaparin 40 mg subcutaneously once daily was given for the first 7 days postoperatively. Patient was vitally stable and ambulatory till day 14.

However on day 14 post exploratory laparotomy she developed dyspnoea for which medicine opinion was taken and CTPA (Computed tomography pulmonary angiography) was done (Figure 8), which was suggestive of non-opacification of contrast in antero-medial basal segmental branch of inter lobar artery supplying the left lateral lower lobe suggestive of thrombosis of sub-segmental branch as depicted in figure 5 and coagulation profile was normal and she was shifted to MICU for better management immediately where she was started on unfractionated Heparin and higher antibiotics on D15her condition deteriorated for which she was intubated and also had developed pancytopenia and her haemoglobin dropped to 6.2gm% on post-operative day 16 and she was planned for Packed red cell transfusion but before we could start the blood transfusion her condition deteriorated further and she went into bradycardia and cardiopulmonary resuscitation as well as adrenaline and atropine given immediately despite all resuscitative measures she succumbed to death.

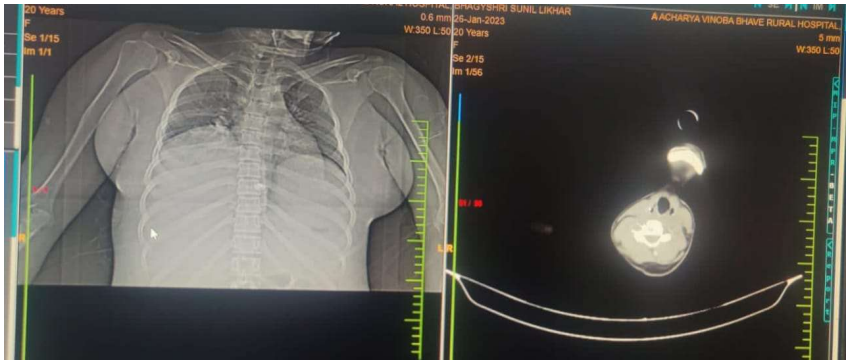

Figure 8 CTPA findings suggestive of thrombus in the artery supplying left lower lobe

4. DISCUSSION

There are various reports available on thromboembolisms in association with tumor. The frequency differs according to the type of malignancy and is reported more in association with brain malignancies, adenocarcinoma of the ovary, colon, pancreas, stomach, prostate and lung (Cyriac et al., 2009). Usually, leading cause for the thrombi is surgery, use of chemotherapeutic agent like Cisplatin and central venous catheterisation (Jafri and Protheroe, 2008). Rarely tumor embolization, primary tumor metastasis and vascular compression due to tumor mass may develop thrombus (Mitomi et al., 2011; Stergiopoulos et al., 2011; Abdel-Razeq et al., 2011; Natsuaki et al., 2009).

Thrombi linked with tumors are usually due to cisplatin-based chemotherapy as Cisplatin is considered to begin degeneration in vessel walls and also to disturb the equilibrium between dissolution of blood clots and the thrombosis, by the by leading to vascular occlusion disease (Jafri and Protheroe, 2008). There are only a few reports on thromboembolism in cases with germ cell tumor so incidence of thromboembolism in such cases is not known. Raised serum β-HCG levels may lead to thromboembolisms in cases with germ cell tumors (Cyriac et al., 2009) increased BMI and elevated levels of serum LDH also could lead to this condition (Owen, 2010; Zhou and Ding, 2009). In children and young adolescents, the probable cause for thromboembolism could be coagulation defect like thrombocythemia, deficiency of protein C and S or anti thrombin deficiency, raised homo cysteine levels and dyslipidaemia with raised BMI.

In the present study, patient had normal body mass index and coagulation profile was also within normal limits. Patient did not receive any chemotherapy based on cisplatin. In reality, our patient developed pulmonary thromboembolisms prior to chemotherapy. Our patient had elevated tumor markers such as AFP, LDH and β-HCG levels, which are linked with thromboembolisms associated with tumor. Another interesting fact is that she was given 7 days Low molecular weight heparin post operatively. Pulmonary thromboembolism may be liked with the surgical procedure as well.

Previously two studies Zganjer et al., (2006) and Mitranovici et al., (2022) have stated that if a young female comes with acute abdomen and on ultrasound if a solid mass is seen, one of the differential diagnosis could be ovarian dysgerminoma. Our case also gives history of pain abdomen from 7 days but the main presenting symptom was distension of abdomen. Khandwala et al., (2018) studied role of computed tomography in a case of ovarian torsion with dysgerminoma. But in our case MRI was done and there was no torsion noted pre op and intra operatively.

5. CONCLUSION

Our patient is the first case to be reported that we are aware of with post-operative thromboembolism in the clinical scenario. Ovarian dysgerminoma ideally, it would be a part of the differential diagnosis for a young female who presents pelvic mass that can be felt and lower quadrant pain and elevated CA125AFP and LDH. If the patient's condition had been looked into for distension of abdomen the cancers could have been found earlier in their development. The important learning aspect was post-operative PTE which was lethal in our case. Contrary to the majority of dysgerminomas, which are discovered in the early stages, our patient appeared at an advanced stage with omental metastases, increasing the likelihood of a poor outcome. The prognosis is favourable, with a decent five-year survival rate up to 85% after appropriate chemotherapy on early stages of diagnosis. The risk for VTE should not be ignored in post-operative period which may lead to mortality; hence close watch in post op period is required in such cases.

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Informed Consent
Informed Consent was obtained from the patient.

Author's contribution
All the authors contributed equally to the case report.

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Conflict of interest
The authors declare that there is no conflict of interests.

Data and materials availability
All data sets collected during this study are available upon reasonable request from the corresponding author.

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