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Chylous ascites following retroperitoneal lymphadenectomy in a patient with recurrent dysgerminoma of ovary: A case report

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ABSTRACT

Chylous ascites is an uncommon condition of accumulation of milky fluid rich in lymph and chylomicrons in the peritoneal cavity. Post-surgical complications following dissection near the base of the mesentery, retroperitoneum, or near the cisterna chyli, malignancies (e.g., pancreatic adenocarcinomas, lymphoma, gastric carcinoma), cirrhosis, and trauma are the prime causes of chylous ascites. Here we report a rare case of chylous ascites following clearance of isolated paraaortic nodal recurrence in a 28-year-old female with dysgerminoma of ovary. The patient developed chylous ascites on the fifth day following surgery, which was confirmed by an increased drain fluid triglyceride level. She was managed conservatively with dietary modification including a high-protein and carbohydrate but low-fat-based diet mainly containing medium-chain fatty acids. Subsequently, she recovered from chylous ascites on the sixteenth day, completed second line chemotherapy, and is now doing well.

1. Introduction

Chyle is milky fluid due to the presence of lymph and chylomicrons containing long-chain triglycerides. After absorption from the intestine, it is circulated via lymphatic channels all the way to the cisterna chyli, which are located anterior to the first and second lumbar vertebrae (Shyr et al., 2020 May 1). Chyle leakage may occur following lymphatic injury from trauma or surgery in the chest, abdomen, or neck resulting in chylous ascites (Smoke and Delegge, 2008). Chylous ascites is an uncommon condition due to accumulation of peritoneal fluid rich in lymph and triglycerides (Yeo et al., 2008), and conservative management is the mainstay of treatment (Göçmen et al., 2014).

Surgical manipulation and dissection near the base of the mesentery, retroperitoneum, or the cisterna chyli may lead to chylous ascites, but dysgerminoma of the ovary is a rare association (Yeo et al., 2008). Dysgerminoma, a malignant ovarian germ cell tumor (MOGCT), comprises up to one-third of MOGCTs. It can occur in all age groups, but with the peak incidence in adolescents and young adults (Kilic et al., 2021).

Here, we report a case of chylous ascites following nodal clearance of an isolated *para*-aortic nodal recurrence in a case of dysgerminoma ovary.

2. Case presentation

A 28-year-old female P2L2, body mass index of 19 Kg per square metres, with no significant medical and family history had undergone a total abdominal hysterectomy-bilateral salphingo-ophorectomy-infracolic omentectomy for a left ovarian mass 28 months earlier at an external institute. Her preoperative Alpha Feto-Protein (AFP) was elevated (1408 IU/mL) but the rest of the tumor markers were normal. The final pathology reviewed was International Federation of Gynaecology and Obstetrics (FIGO) Stage IIB dysgerminoma of the left ovary. Following surgery, Computed Tomography (CT) of the abdomen was done, which showed a left *para*-aortic conglomerated mass measuring 37x35 mm abutting renal vessels and aorta likely nodal metastasis (Fig. 1-a). However, the decision was made to go ahead with chemotherapy by the treating team, counselling the patient and her family

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members that the chemotherapy would take care of it. She did well with four cycles of chemotherapy, i.e., Etoposide-Platinum (EP) regimen. No radiological imaging was done after treatment completion, and she was advised for regular post-treatment follow-up. However, she was asymptomatic and did not return for follow-up for 23 months. After nearly two years, she came to seek medical consultation with the treating team when she noticed a painless mass in the epigastric region.

She was referred and presented to the gynaecological outpatient department of B. P. Koirala Institute of Health Sciences (BPKIHS) on 29 November 2021, with a three-month history of a mass in the epigastric region. CT abdomen showed an enhancing multi-cystic retroperitoneal soft tissue mass measuring 14.5x12 cm with an intraperitoneal component in the left lumbar region encasing the left upper ureter, causing proximal mild hydroureteronephrosis, and displacing the left kidney superiorly and also abutting and displacing a broad area of the abdominal aorta, pancreas, and multiple bowel loops, likely *para*-aortic nodal recurrence (Fig. 1-b, c). The rest of the metastatic workup was normal. Fine needle aspiration cytology from the mass suggested metastatic adenocarcinoma. Her tumor markers were as follows: CA125 = 43.37 U/mL, AFP = 1000 IU/mL, LDH = 419 U/mL.

Subsequently, in December 2021, she underwent debulking surgery with excision of a conglomerated para-aortic nodal mass along with left nephrectomy and adrenalectomy. Intraoperative findings were as described: 500 mL of blood-stained peritoneal fluid was present, which was sent for cytology. A left para-aortic conglomerated lymph node mass measuring 24 cm \times 24 cm with flimsy adhesions to the transverse colon and its mesentery was present. The mass was densely adherent to the left renal vessels at the renal hilum and left kidney at its lower pole superiorly, aorta medially, left abdominal wall laterally, psoas muscle and bed of retroperitoneum posteriorly, and transverse colon and its mesentery anteriorly. The mass was extending from the level of the left renal hilum up to the left common iliac vessels encasing the left ureter completely from its origin at the kidney up to the pelvic brim. Intraoperative urosurgery consultation was sought. During the adhesiolysis, there was trauma to the left kidney with severe bleeding and hemodynamic instability, so the decision was made for nephrectomy. The left renal vein followed by the renal artery was ligated, and nephrectomy along with adrenalectomy was done (Fig. 2-a, b). Lumbar arteries, the inferior mesenteric artery, and gonadal arteries encountered were ligated at their origins. Finally, the para-aortic nodal mass was separated from the aorta by blunt dissection (Fig. 2-c). Intraoperative blood loss was 1500 mL, and the patient received four pints of Packed Red Blood Cells and one pint of Fresh Frozen Plasma. The duration of surgery was 6 h. Post-operatively, drain output was as follows: first day-40 mL, serosanguinous; second day-100 mL, serosanguinous; third day-50 mL, serous; fourth day-230 mL, serous. On the fifth POD, drain output was surprisingly chylous, 400 mL (Fig. 3), and increased to 1400 mL on the seventh day, which was confirmed by an increased drain fluid

triglyceride level (424 mg/dL). Dietician consultation was done, and the patient was started strictly on a medium-chain fatty acid, high-protein and carbohydrate-based diet; a long-chain fatty acid diet was avoided. Following this, chylous ascites were reduced significantly and became 50 mL by the fifteenth day. The drain was removed on the sixteenth day, and the patient was discharged on the seventeenth day. The histopathological feature of the *para*-aortic nodal mass was suggestive of dysgerminoma with involvement of the ureteric margin near the renal hilum. The patient completed four cycles of second line chemotherapy (Etoposide, Ifosfamide, Cisplatin - VIP regimen) in June 2022. Follow-up tumor markers and CT abdomen and chest were normal. To date, the patient is doing well with progression-free survival of 8 months.

3. Discussion

Chylous ascites is an uncommon condition resulting from abnormal accumulation of lymph in the abdominal cavity. Nowadays, its incidence has increased with more aggressive retroperitoneal surgery. Kaas et al. (Kaas et al., 2001) in their retrospective study among 1103 patients reported an incidence of 1.1% post-operative chylous ascites. Various studies have reported different incidence rates (1-7%) for this rare condition (Combe et al., 1992; Baniel et al., 1995; Evans et al., 2006). In gynaecological malignancies, post-operative chylous ascites are rare and incidence ranges from 0.17% to 4%. Moreover, the incidence is higher for para-aortic lymphadenectomy (0.32%) than for pelvic lymphadenectomy (0.077%) (Solmaz et al., 2015). Post-surgical complications following dissection near the base of the mesentery or near the cisterna chyli, malignancies (e.g., pancreatic adenocarcinomas, lymphoma, gastric carcinoma), and cirrhosis are the prime causes (Yeo et al., 2008) but we found a rare case of chylous ascites following debulking surgery of isolated para-aortic nodal recurrence in a 28-year-old female with dysgerminoma of the ovary. Several studies reported that the incidence of lymph node metastasis was the highest in dysgerminoma, with the incidence ranging from 11.3% to 28.3% (Nasioudis et al., 2020; Nasioudis et al., 2020; Kumar et al., 2008). In our patient, the 3.7 cm metastatic para-aortic nodal mass noticed on the post-operative CT scan was missed in the initial surgery and had progressively enlarged to the present size over a period of 23 months after completion of chemotherapy. And the draining tributaries to the cysterna chyli were likely to have been injured during nodal clearance in the current surgery, leading to the chylous ascites. Hence, the lymph node condition should be carefully evaluated before and during the surgery in dysgerminoma when the decision not to perform a lymphadenectomy is made (Nasioudis et al., 2020; Nasioudis et al., 2020; Kumar et al., 2008). Abdominal distension and discomfort, anorexia, indigestion, weight loss, leg edema, weakness, nausea, vomiting, dyspnea, weight gain, and early satiety are the usual non-specific clinical symptoms. Physical examination is consistent with ascites in nearly all cases. A few days to one



Fig. 1. A,b,c. Postoperative CT images (a) showing unaddressed enlarged paraaortic lymph node at initial surgery and Preoperative CT images (b,c) showing huge conglomerated paraaortic lymph node mass at second/debulking surgery.

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Fig. 2. A,b,c. Intraoperative images (a,b) postoperative images (c) of specimen after debulking.



Fig. 3. Drain bag containing chylous ascites.

month, most commonly after one week, is the usual duration of presentation of chylous ascites following surgery (Soto et al., 2011). In our patient, clinical symptoms were non-specific and developed chylous ascites on the fifth day following surgery. To diagnose chylous ascites, a biochemical test is the diagnostic tool, aided by clinical suspicion. If the ascitic fluid appears milky white and a triglyceride concentration is above 200 mg/dL, this confirms the diagnosis of chylous ascites, while a level less than 50 mg/dL rules out chylous ascites (Lizaola et al., 2017 Nov). In this case, the drain fluid was milky with a very high triglyceride level of 424 mg/dL. The management of chylous ascites is conservative, which is the mainstay of treatment. Aalami et al. (Aalami et al., 2000) in their study identified 156 cases of chylous ascites that were successfully managed, of which 67% were treated conservatively and 33% were treated surgically. Dietary restriction with a high-protein, low-fat diet containing medium-chain triglycerides, fasting with total parenteral nutrition, somatostatin therapy (Octreotide), paracentesis, or continuous drainage are the conservative treatments (Kuboki et al., 2013 Mar). If the conservative management fails or the chyle loss is more than 1.5 L per day for more than 5 to 7 days, then surgical intervention is advocated (Smoke and Delegge, 2008). Surgical intervention includes laparotomy and peritoneo-venous shunt or peritoneo-atrial shunt (Zhao

et al., 2014 Jan). Our patient developed chylous ascites on the fifth day and was managed conservatively. She started on a low-fat diet containing mainly medium-chain fatty acid with high protein and carbohydrate; a long-chain fatty acid diet was avoided. Medium-chain triglycerides are directly absorbed by enterocytes and transported as free fatty acids and glycerol in the blood directly to the liver via the portal vein (Singh et al., 2019 Oct 1). A low-fat diet decreases fat absorption in the intestine and decreases lymphatic flow to cisterna chyli. However, long-chain triglycerides are broken down into monoglycerides and free fatty acids, which are absorbed and transported via intestinal lymphatics to the cisterna chyli, and thus aggravate chylous leaks (Rose et al., 2022). Octreotide (somatostatin analogue) reduces gastric, pancreatic, and intestinal secretion, inhibits intestinal motility, and attenuates splanchnic blood flow, hence decreasing intestinal fat absorption and reducing the triglyceride level in the lymphatic flow (Lee et al., 2005).

4. Conclusion

Post-operative chylous ascites is an uncommon presentation in gynaecological oncological surgery. Here, we present a case of chylous ascites that developed following isolated *para*-aortic nodal clearance in a patient of ovarian dysgerminoma. In our case, chylous ascites occurred following isolated *para*-aortic nodal clearance in a diagnosed case of dysgerminoma ovary, which is a rare presentation. Paracentesis analysis of ascitic fluid collected through an abdominal drain is the mainstay of diagnosis of chylous ascites. Low-fat, high-protein diets along with medium-chain fatty acids or total parenteral nutrition and avoidance of long-chain fatty acids are some principal ways of managing this uncommon problem as we did in our case. Hence, conservative management is the mainstay of treatment and provides better outcomes and lesser morbidities with respect to surgeries.

Author Contributions

A. N. and A. G. wrote the manuscript in consultation with R. S. and K. D., P. P., S. P. R., A. K., R. J. K., and S. K. supervised the project. R. S. reviewed the manuscript draft and revised it critically on intellectual content. All authors approved the final version of the manuscript to be published.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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References

Aalami, O.O., Allen, D.B., Organ, C.H., 2000. Chylous ascites: a collective review. Surgery. 128, 761Y778.

- Baniel, J., Foster, R.S., Rowland, R.G., Bihrle, D.JP., 1995. Complications of postchemotherapy retroperitoneal lymph node dissection. J. Urol. 153, 976e980.
- Combe, J., Buniet, J.M., Douge, C., Bernard, Y., Camelot, G., 1992. Chylothorax and chylous ascites following surgery of an inflammatory aortic aneurysm. Case report with review of the literatüre. J. Mal. Vasc. 17, 151e156.
- Evans, J.G., Spiess, P.E., Kamat, A.M., Wood, C.G., Hernandez, M., Pettaway, C.A., et al., 2006. Chylous ascites after post-chemotherapy retroperitoneal lymph node dissection: review of the M. D. Anderson experience, J. Urol. 176.
- Göcmen A, Avcı ME, Sanlıkan F, Ucar MG. Four Cases of Chylous Ascites following Robotic Gynecologic Oncological Surgery. Case Rep Obstet Gynecol. 2014; 2014: Article ID 953965, 3 pages. https://doi.org/10.1155/2014/953965.
- Kaas, R., Rustman, L.D., Zoetmulder, F.A., 2001. Chylous ascites after oncological abdominal surgery: incidence and treatment. Eur. J. Surg. Oncol. 27, 187e189.
- Kilic, C., Cakir, C., Yuksel, D., Kilic, F., Kavikcioglu, F., Koc, S., et al., 2021. Ovarian Dysgerminoma: A Tertiary Center Experience. J. Adolesc. Young Adult Oncol. 10, 303-308
- Kuboki, S., Shimizu, H., Yoshidome, H., Ohtsuka, M., Kato, A., Yoshitomi, H., et al., 2013 Mar. Chylous ascites after hepatopancreatobiliary surgery, Br J Surg, 100 (4), 522-527.
- Kumar, S., Shah, J.P., Bryant, C.S., Imudia, A.N., Cote, M.L., Ali-Fehmi, R., et al., 2008. The prevalence and prognostic impact of lymph node metastasis in malignant germ cell tumors of the ovary. Gynecol Oncol 110, 125–132. Lee, P.H., Lin, C.L., Lai, P.C., Yang, C.W., 2005. Octreotide therapy for chylous ascites in
- achronic dialysis patient. Nephrology (Carlton) 10, 344-347.
- Lizaola, B., Bonder, A., Trivedi, H.D., Tapper, E.B., Cardenas, A., 2017 Nov. Review article: the diagnostic approach and current management of chylous ascites. Aliment Pharmacol Ther. 46 (9), 816-824.

- Nasioudis, D., Ko, E.M., Haggerty, A.F., Cory, L., Giuntoli, R.L., Burger, R.A., et al., 2020. Performance of lymphadenectomy for apparent early stage malignant ovarian germ cell tumors in the era of platinum-based chemotherapy. Gynecol Oncol 157, 613-618
- Nasioudis, D., Mastroyannis, S.A., Latif, N.A., Ko, E.M., 2020. Trends in the surgical management of malignant ovarian germcell tumors. Gynecol Oncol. 157, 89-93.
- Rose, K.M., Huelster, H.L., Roberts, E.C., Manley, B.J., Gilbert, S.M., Sexton, W.J., 2022. Contemporary Management of Chylous Ascites after Retroperitoneal Surgery : Development of an Evidence-Based Treatment Algorithm. 208 (July), 53-61.
- Shyr, B.U., Shyr, B.S., Chen, S.C., Shyr, Y.M., Wang, S.E., 2020 May 1. Chyle leakage after robotic and open pancreaticoduodenectomy. J Hepatobiliary Pancreat Sci. 27 (5), 273-279.
- Singh, H., Pandit, N., Krishnamurthy, G., Gupta, R., Verma, G.R., Singh, R., 2019 Oct 1. Management of chylous ascites following pancreaticobiliary surgery. JGH Open. 3 (5), 425-428.
- Smoke, A., Delegge, M.H., 2008. Chyle leaks: Consensus on management? Vol. 23, 529-532. Nutrition in Clinical Practice.
- Solmaz U, Turan V, Mat E, Dereli ML, Ekin A, Peker N, et al. Chylous ascites following retroperitoneal lymphadenectomy in gynecologic malignancies: Incidence, risk factors and management. Int J Surg. 2015 Apr 1;16(Part A):88-93.
- Soto, E., Soto, C., Nezhat, F.R., Gretz, H.F., Chuang, L., 2011. Chylous ascites following robotic lymph node dissection on a patient with metastatic cervical carcinoma. J Gynecol Oncol. 22 (1), 61-63.
- Yeo, M.S.W., Tan, L.G.L., Chang, S.K.Y., 2008. Postoperative chylous ascites: An institutional experience over two years. Surg Pract. 12 (4), 133-136.
- Zhao, Y., Hu, W., Hou, X., Zhou, Q., 2014 Jan. Chylous Ascites After Laparoscopic Lymph Node Dissection in Gynecologic Malignancies. J Minim Invasive Gynecol. 21 (1), 90-96.