CASE REPORT

SIADH caused by immature ovarian teratoma with gliomatosis peritonei: A case report

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INTRODUCTION

Immature ovarian teratoma causing SIADH with gliomatosis peritonei (GP) is a rare clinical entity. Immature ovarian teratoma comprises of less than 1% of all ovarian tumours and is made of all 3 germ cell layers (ectoderm, endoderm and mesoderm). This is the only tumour that is graded according to immature neural elements and determines the prognosis as well as treatment plan. GP is defined as benign peritoneal deposits of matured glial tissues. SIADH is a constellation of hyponatremia, hypotonicity and increased sodium loss in the urine. We report a case of a 30-year-old female with a right immature ovarian teratoma with SIADH and GP. To the best of our knowledge, this is the first immature ovarian teratoma with SIADH and GP ever published.

CASE PRESENTATION

A 30-year-old, para 1 female presented with a lower abdominal pain and distension for one week duration. Physical examination revealed a huge mobile right lower abdominal mass that crosses the umbilicus and was firm in consistency measuring 20 x 20cm. Contrast-enhanced computerized tomography scan(CECT) of the abdomen and pelvis showed a large, well demarcated, encapsulated lobulated heterogeneous enhancing solid cystic mass with coarse calcification measuring 8.9cm x 15cm x 15cm (APXWXCC) with clear plane with surrounding organs (Figure 1A and 1B). Her tumour markers that were raised were AFP which was 19.6 IU/ml, CA 125 which was 223 U/ml and CA 19-9 which was 197 U/ml. Her LDH was 253 U/L with sodium of 125, hypotonic serum osmolarity and high urine sodium.

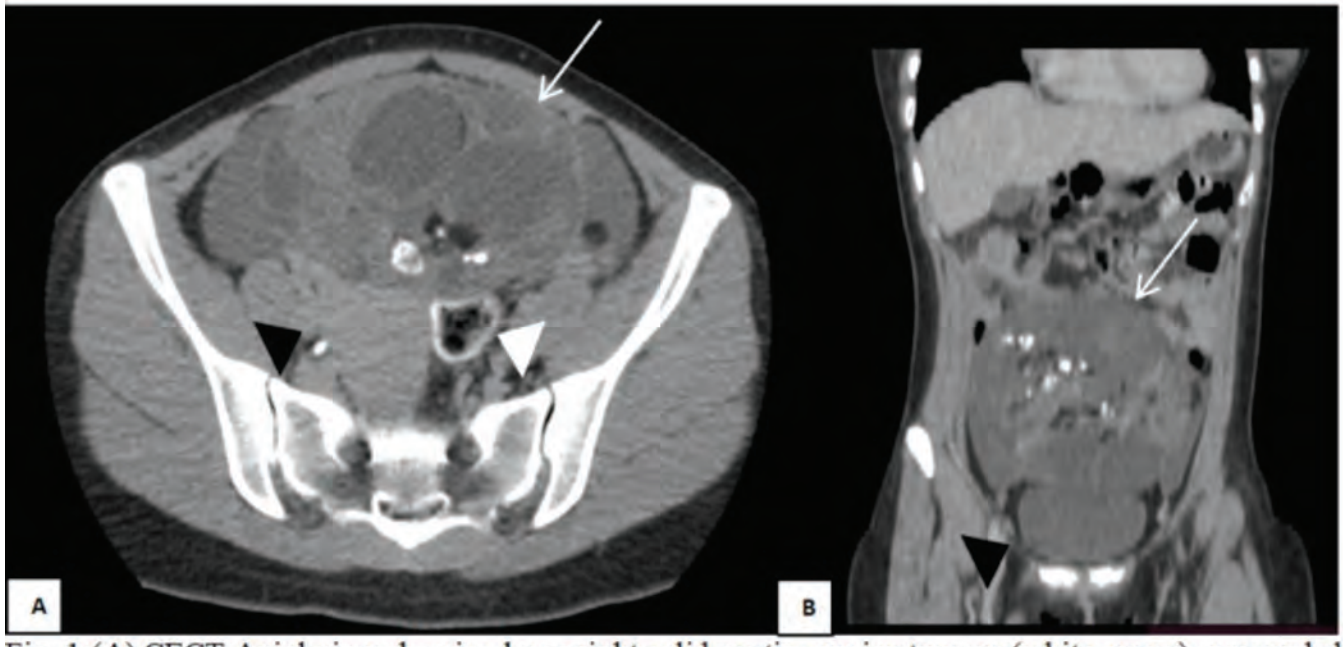

Fig. 1:(A) CECT Axial view showing large right solid-cystic ovarian tumour (white arrow) surrounded by uterus (black arrow head) and sigmoid colon (white arrow head) posteriorly. (B) CECT Coronal view showing a large right solid-cystic ovarian tumour displacing the bladder inferiorly.

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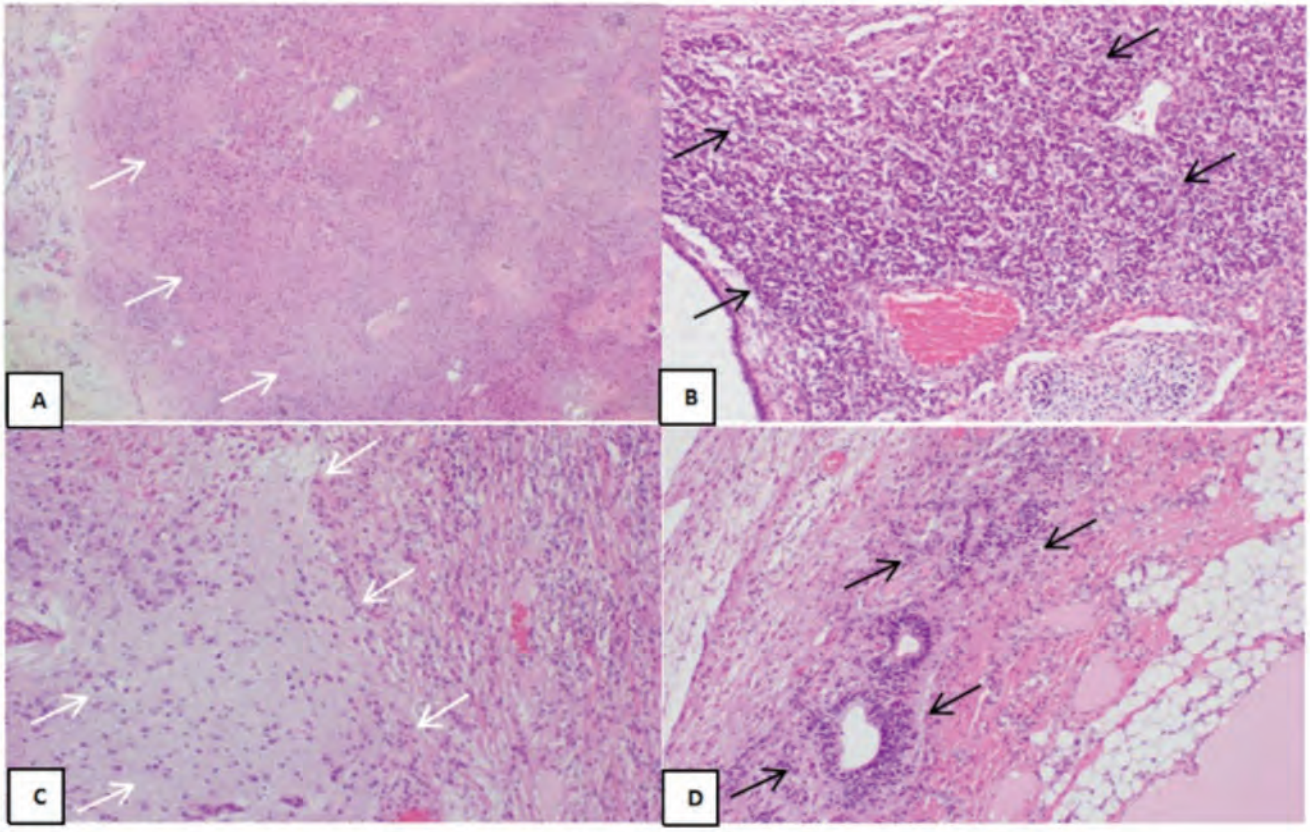

Fig. 2: (A) Peritoneal nodule HPE, 40x magnification. Compact nodules of mature glial tissue surrounded by fibrocollagenous and fibrovascular stroma (white arrows). (B) Ovarian Tumour HPE, 100x magnification. Immature neuroectodermal tissue comprises tubules and rosettes of primitive, mitotically active cells with increased nuclear:cytoplasmic ratio, hyperchromatic nuclei and scant cytoplasm (black arrows). (C) Ovarian Tumour HPE, 100x magnification. Glial tissue is seen in this field (white arrows). (D) Ovarian Tumour HPE, 100x magnification. Admixture of immature and mature teratoma components. Glial tissue, adipose tissue, and immature neuroectodermal tissue are present in this field (black arrows).

She was first electively admitted for right salpingo-oopherectomy, omentectomy and appendicectomy with keep in view pelvic lymph node dissection. However, as her sodium was reported to be low and her surgery was postponed to the following week while optimizing her sodium by restricting her fluid as her blood results was suggestive of SIADH. When her bloods were optimized she underwent a midline laparotomy for right salpingo-oopherectomy, omentum biopsy and appendicectomy with pelvic lymph node harvest. Intra operatively we noticed multiple peritoneal nodules at anterior abdominal wall and the pelvis as well as 800 cc of straw colour ascitic fluid which we biopsied as well as aspirated to send for histopathological examination (HPE) and cytological examination. The right ovary measured 25 x 25cm intraoperatively with nodules on the surface of the right ovary while the left ovary appears to be normal. Uterus and other bowels were normal. There was also omental caking and enlarged right pelvic lymph nodes and hence we proceeded with omentectomy and right pelvic lymph node dissection.

The post-operative course was uneventful and her sodium normalized and she was discharged at post operative day 6. HPE showed grade 1 immature right ovarian teratoma which is low grade (Grade 1)with focal capsular breach (Figure 2B, 2C and 2D). Her lymph nodes showed nodal gliomatosis while her peritoneum and omentum showed gliomatosis peritonei (Figure 2A) and lastly appendix was normal. The ascitic fluid showed loose cohesive cells, reactive mesothelial cells, lymphocytes and histocytes. At follow up review of 3 months, she was well and undergoing adjuvant chemotherapy - BEP regime (Bleomycin, Etoposide and Carboplatin) planned for 4 cycles.

DISCUSSION

GP was first reported by Benirschke in 1960 while SIADH due to immature ovarian teratoma was first reported by Lam in 1996.[1-2] Based on Pubmed search engine there are only 118 cases that reported immature ovarian teratoma with GP and only 8 cases with SIADH. However, there were no reported cases on immature ovarian teratoma with SIADH and GP.

There are 2 main theories that spurred the development of GP. The first theory being via angiolymphatic spread or via capsular breach of the ovarian tumour like our case based on the histopathology.[3] The second theory is that glial foci are not genetically associated with the ovarian tumour but arises from normal cells from the pluripotent Mullerian stem cells. These cells gets differentiated into glial cells by the ovarian teratoma secreting some stimulation factors.[4]

SIADH is postulated by the immature cells of ovarian teratoma being pluripotent in nature. There have been previous publications in regards to pituitary component present in ovarian teratoma.[5] The definitive diagnosis can only be made via histopathology. Immature ovarian teratomas are germ cell tumours. For immature ovarian teratoma with gliomatosis peritonei based on the WHO grading system it is graded 0 however due to the certain areas having rare foci of immature cells it is histologically grade 1.

Salpingo-oopherectomy, omentectomy, appendicectomy, peritoneal biopsy and pelvic lymph node dissection has always been advocated as the treatment of ovarian tumours. While the mainstay of SIADH treatment is fluid restriction and surgical resection. Prognosis or overall survival of immature ovarian teratoma is not affected by presence of GP however the recurrence rates were higher in those with GP. Prognosis is based on the quantity of immature cells in the ovarian tumour and this will decide the grade of the tumour and treatment plan. Adjuvant chemotherapy with BEP is the chemotherapy of choice mainly for high grade tumours (Grade 2 and 3). However, the role of adjuvant chemotherapy is debatable in grade 1 immature ovarian tumours as some centres suggest only close observation while others advocate adjuvant chemotherapy based on the age, GP spread, and tumour burden. For our case, we suggested 4 cycles of chemotherapy with BEP regime because the patient was young, there was wide GP spread and huge tumour burden. We also discussed with the patient in regards to fertility preservation as she was still young and had a baseline lung function and ECHO in view that the chemotherapy can lead to adverse effects affecting her lung and heart.

CONCLUSION

Immature ovarian teratoma can present with SIADH. GP although rare is a differential diagnosis in the event of intraoperative wide spread peritoneal dissemination. Immature ovarian teratomas with GP have a good prognosis with equivalent overall survival with those without peritoneal disease but have a higher recurrence rate. Adjuvant chemotherapy in grade 1 tumours are uncertain and it all depends on few factors. However, in our patient we believe the surgery we performed and adjuvant chemotherapy we offered was the best option and effective method to treat her and reduce risk of recurrence.

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DECLARATION

The authors declare no potential conflict of interest with respect to the research, authorship, and publication of this article.

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