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Case Report

Surgery Section

Mixed Germ Cell Tumour in an Infertile Male Having Unilateral Cryptorchidism: A Rare Case Report

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ABSTRACT

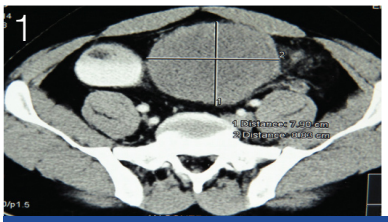
Mixed germ cell tumours with multiple components occur more frequently than the pure varieties of germ cell tumours. Embryonal carcinoma and teratoma together form the most common components of the mixed germ cell tumour but the yolk sac tumour is usually seen as a minor component in patients presenting with mixed germ cell tumour. We report a rare case of 27-year-old Hepatitis C positive male presenting with pain in left lower abdomen with associated history of same sided undescended testis and infertility. Right sided testis lying in scrotal sac appeared normal on ultrasonography but patient was azoospermic. He had raised levels of serum markers, alpha feto protein and beta HCG. Examination showed a large mass in left lower abdomen involving the sigmoid colon with the absence of left testis in left scrotum which was confirmed on CT scan. Excision of the mass was done and histopathology examination revealed it as a malignant mixed germ cell tumour composed predominantly of a yolk sac tumour, with minor component as seminoma and embryonal carcinoma in an undescended testis. Following this, the level of serum markers came down. The patient is now undergoing adjuvant chemotherapy and is doing well.

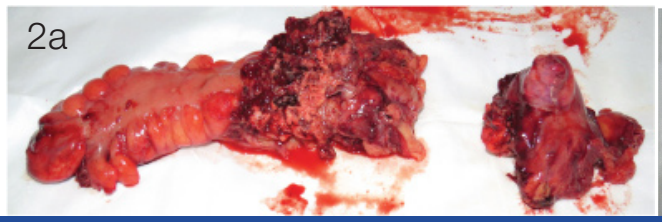
Keywords: Azoospermia, Cryptorchid testis, Infertility, Malignant mixed germ cell tumour, Yolk sac tumour

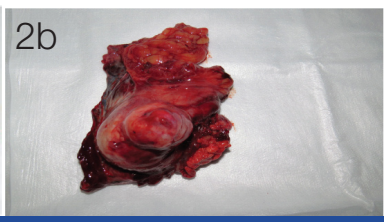
CASE REPORT

A 27-year-old HCV positive male presented with pain in left iliac fossa for 2 weeks with a history of left sided undescended testis since birth. On examination a mass was palpable in left lower abdomen along with empty scrotum on the same side. Left scrotum was not well developed. On ultrasonography a space occupying lesion was seen in left lower abdomen but left testis was not visualized and right testis lying in the scrotal sac appeared to be normal without any microcalcification. CT Scan confirmed a soft tissue mass 10cm x 8cm x 8cm lying superior to the bladder in midline & towards the left side [Table/Fig-1]. It was involving the sigmoid colon. A few subcentimetric sized lymph nodes were seen in preaortic, paraaortic, and bilateral inguinal regions. There was no involvement of the pelvic bones. CT upper abdomen and chest was normal. Semen analysis showed azoospermia. The level of alpha feto protein was 10550 ng/ml, beta HCG was 3.4 mIu/ml and LDH was 398 U/L.

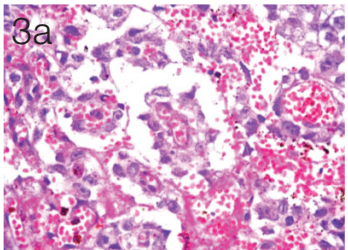
Tumour mass was excised along with part of the involved colon [Table/Fig-2a&b] and a primary colocolonic anastomosis was done. Repeated serum markers after 2 weeks showed alpha feto protein level to be 1947 ng/ml, beta HCG was 1.7mIu/ml and LDH was 249 U/L. On gross description the tumour measured 6cm x 4.5cm x 3.5cm which was adherent to the sigmoid colon. The cut surface of this mass was greyish white in colour and was irregular. The colon had an area measuring 1.5cm x 2cm greyish brown area identified 2cm from one end. Histopathological examination revealed it as a malignant mixed germ cell tumour of the undescended testis composed predominantly of yolk sac tumour [Table/Fig-3a&b] with foci of seminoma [Table/Fig-4] and embryonal cell carcinoma [Table/Fig-5]. The intestine wall was congested with areas of necrosis with acute and chronic inflammatory cells although there was no tumour infiltration. Post operative chemotherapy with BEP regimen was started and the patient is doing well presently.

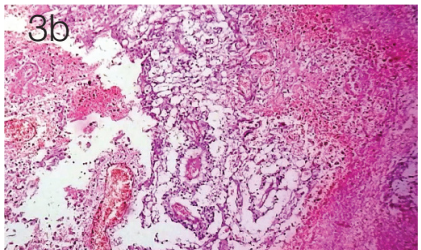


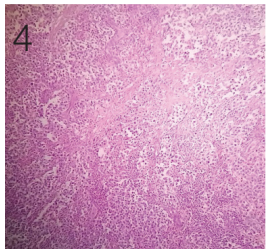


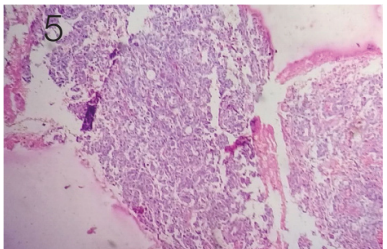


[Table/Fig-1]: CT Scan section showing large mass in the pelvis lying left superolaterally to the urinary bladder. **[Table/Fig-2a]:** Gross specimen after resection containing tumour mass and the involved sigmoid colon. **[Table/Fig-2b]:** Gross specimen after resection containing tumour mass separated from the sigmoid colon.









[Table/Fig-3a]: 400X, H&E, Yolk sac component showing schiller duval bodies seen in the patient's specimen sent for histopathology examination after excision of the tumour mass.
[Table/Fig-3b]: 100X, H&E, Yolk sac component showing schiller duval bodies.
[Table/Fig-4]: 100X, H&E, Seminoma component showing dispersed population of cells along with connective tissue and lymphocytes.
[Table/Fig-5]: 100X, H&E, Embryonal cell component showing sheets of undifferentiated pleomorphic cells seen in the patient's specimen sent for histopathology examination.

DISCUSSION

Cryptorchidism is a developmental defect in which the testes fail to descend completely into the scrotum. Cryptorchidism is most often unilateral, but in a small number of patients it may be bilateral. Around 90% of patients with untreated bilateral cryptorchidism ultimately develop azoospermia as against the reported 0.4 to 0.5% incidence in the general population. About 49% of men with persistent unilateral cryptorchidism have a normal sperm count as compared to 71% after orchidopexy. Reduced paternity rates post treatment, have been found for bilateral, but not unilateral cryptorchidism [1-4]. In our case, inspite of unilateral cryptorchidism, the patient had azoospermia.

Though cryptorchidism is a certain risk factor for testicular tumours, the proportion of testicular cancer attributed to cryptorchidism is just about 5% [5]. In unilateral cryptorchidism most tumours occur in the affected testis [6]. In 8–15% of cases these tumour occur in the contralateral scrotal testis [6]. A higher risk of malignancy is seen in bilateral cryptorchidism than unilateral [7]. The risk of developing germ cell tumour is usually greater if the testis is located intra-abdominally. In our patient the cryptorchid testis presented as an intra abdominal mass which turned out to be malignant and involved the sigmoid colon.

Testicular tumours are classified into germ cell tumours and non-germinal tumours. Tumours of germ cell origin comprise about 95% of all testicular cancer. Germ cell tumours are further divided into seminomas which occur in approximately 40% of the population and non-seminomatous tumours which may be seen in a pure or mixed form [8]. Mixed germ cell tumours are much more common than any of the pure histologic forms. They represent 32%-60% of all germ cell tumours with most common admixtures being embryonal carcinoma and teratoma [9]. Yolk sac tumour is usually present as a minor component, being overshadowed by other components, such as embryonal carcinoma [9]. But on the contrary our patient had an unusual mixed variety of germ cell tumour in which the intra abdominal testicular tumour was found to be a malignant mixed germ cell tumour consisting of yolk sac tumour as the majority of the part along with seminoma and embryonal carcinoma as minor components. The exact incidence of occurrence of this type of histopathogenesis is not known as it is rare entity. Only few such cases have been reported to the best of our knowledge [10-12]. This mass further involved the wall of sigmoid colon though no tumour infiltration was seen however areas of necrosis with acute and chronic inflammatory cells were present.

In infancy and childhood the pure yolk sac tumour forms is one of the most common testicular tumours [13,14]. However, its occurence is extremely rare in postpubertal age group [10]. In adults it is often found as a mixed variety leading to increase in levels of AFP, as in our patient [10]. Beta HCG and AFP are the serum tumour markers of testicular neoplasia, which play an important role in the diagnosis, management and prognosis. In case of a metastatic disease, the AFP level does not return to normal after orchidectomy [9,15]. In our patient the serum marker alpha feto protein was highly raised i.e. 10550.0 ng/ml indicating predominantly a yolk sac tumour but as beta HCG was also raised i.e. 3.4 mlu/ml it was indicative of a mixed germ cell tumour. After the removal of the tumour mass the levels of serum tumour markers came down significantly. All testicular germ cell tumours usually spread via lymphatic route except pure choriocarcinoma in which vascular spread is seen [9]. In our patient no significant lymphadenopathy was present. Tumour mass was excised along with part of the involved sigmoid colon. Colocolic anastomosis was performed and the patient recovered well. Postoperative levels of alpha feto protein came out to be 1947 ng/ml and that of beta HCG was 1.7 mlu/ml. Although yolk sac tumour is an aggressive tumour, no distant spread was seen in our patient. Adjuvant chemotherapy with bleomycin, etoposide and cisplatin regimen was started and the patient is recovering well.

CONCLUSION

Yolk sac tumour as a whole or as a predominant component of mixed germ cell tumour is a rare entity in adults, but it should always be kept as a possible diagnosis. Owing to its aggressive nature, it should be promptly diagnosed after complete evaluation with adequate treatment and follow up. Azoospermia although an unusual manifestation of unilateral cryptorchidism, is essential to be assessed in every presenting case with the same complaint.

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