Small bowel intussusception caused by a metastatic testicular germ cell tumour

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Summary
Intussusception from a testicular germ cell tumour is extremely rare. Metastatic gastrointestinal implants outgrow their blood supply leading to central necrosis. This results in erosions and ulcerations, which can be visualised as submucosal polypoid masses. These masses can then serve as a lead point for intussusception. We report a case of a 25-year-old patient with small bowel obstruction due to an intussuscepted choriocarcinoma in the absence of any other apparent retroperitoneal disease. Urologists must exclude gastrointestinal tract (GIT) involvement in testicular cancers, and similarly, surgeons need to exclude testicular tumours in young men with unexplained GIT haemorrhage, bowel obstruction or intussusception.

Keywords: germ cell tumour, choriocarcinoma, intussusception, testicular mass, orchidectomy, metastases, small bowel obstruction

Case presentation
A 25-year-old Caucasian male was referred with a six-month history of a progressively worsening swelling of the right hemiscrotum. There was no significant medical, surgical or family history. During the initial consultation, he had odd mannerisms, with random inappropriate outbursts and the patient’s mother confirmed a change in behaviour over the preceding four months. Vital signs were normal, there was no evidence of gynaecomastia, and no lymph nodes were palpated. The abdominal examination was normal. Palpation and bedside ultrasound of the right hemiscrotum revealed a mass on the upper pole of the testis. The left testis was normal. Serum alpha-fetoprotein (AFP – 41 ng/ml), beta-human chorionic gonadotropin (B-HCG – 640 929 miU/ml) and lactate dehydrogenase (LDH – 41 ng/ml), beta-human chorionic gonadotropin (B-HCG – 640 929 miU/ml) and lactate dehydrogenase (LDH – 41 ng/ml) were all elevated. The rest of the blood work was unremarkable. An urgent right radical orchidectomy was performed.

Examination revealed a lobulated tumour (85 x 70 x 60 mm) of the upper pole of the right testis confined to the testis and epididymis (pT2) with large areas of necrosis and haemorrhage. Histopathological analysis showed a mixed non-seminomatous germ cell tumour (NSGCT) with a predominant choriocarcinoma component (90%) as well as yolk sac tumour (5%) and mature teratoma (5%) components. There was evidence of angiolymphatic invasion. He had an uneventful postoperative course after the radical orchidectomy and was discharged the following day with a uneventful postoperative course after the radical orchidectomy. Chemotherapy (bleomycin, etoposide, and cisplatin) was initiated on day five post-resection. However, despite physiotherapy and early mobilisation, the patient developed Klebsiella pneumonia with rapid deterioration. Despite maximal organ support, his clinical condition deteriorated over the following days with the evolution of pancytopenia, and he died 18 days after his laparotomy.

Initial surgical margins and the excised lymph nodes were clear of disease.

Initially, there was clinical improvement post bowel resection. Chemotherapy (bleomycin, etoposide, and cisplatin) was initiated on day five post-resection. However, despite physiotherapy and early mobilisation, the patient developed Klebsiella pneumonia with rapid deterioration. Despite maximal organ support, his clinical condition deteriorated over the following days with the evolution of pancytopenia, and he died 18 days after his laparotomy.
Discussion

Testicular carcinoma, broadly classified into germ cell and non-germ cell tumours, is the most common solid organ malignancy affecting men between 15–45 years of age. It represents 1% of all male tumours and 5% of urological malignancies. While the incidence has been increasing over the years, cure rates for germ cell tumours (GCTs) are as high as 90%, and a 5-year survival rate of more than 95%.\textsuperscript{1,2} While retroperitoneal lymph nodes remain the most common site of metastases, the lungs, liver and brain are also frequently involved. Gastrointestinal tract (GIT) disease from metastatic GCT is sporadic, with clinically apparent GIT involvement occurring in less than 5% of cases.\textsuperscript{3,4} They occur in advanced disease due to either direct extension from affected retroperitoneal lymph nodes that drain the testes, haematogenous spread or peritoneal seeding.\textsuperscript{5,6} Metastatic
implants generally fail to pass through the capillary barrier and deposit into the submucosa. Here, the tumour deposits outgrow their blood supply resulting in central necrosis leading to submucosal (occasionally even mucosal) erosions and ulcerations which can be visualised as submucosal or polypoid masses on endoscopy. These masses can form a lead point for intussusception.

Only a handful of cases of intestinal intussusception caused by metastatic deposits from GCTs have been reported. Abdominal pain and melena and/or haematochezia (meena, haematemesesn and haematochezia) or bowel obstruction (abdominal pain, abdominal distension, vomiting) or perforation. Of the four reported cases, histology from three was reported as metastatic non-seminomatous GCT – two pure choriocarcinomas and one a mixed germ cell tumour (embryonal carcinoma with yolk sac tumour). In one case, the tumour was only reported as a metastatic germ cell tumour, and no distinction between the subtypes was included. This reaffirms reports from other authors that pure seminomas are least likely to metastasise to the GIT.

In the early 1970s, metastatic testicular cancer was associated with only 5% survival. With contemporary multimodal techniques involving chemotherapy, radiation and promising surgical procedures, 80% of patients will survive their disease. Based on the International Germ Cell Cancer Collaborative Group (IGCCCG), our patient falls into the poor prognostic category with a 5-year survival of 67%. As was the case with our patient, two of the other four patients died in the perioperative period or after the initiation of intense chemotherapy. However, patients with GIT metastases represent a group of patients with a much poorer prognosis. The histology of the primary testicular tumour may also contribute to the poor prognosis, as was the case in our patient. Choriocarcinomas are highly malignant lesions with a propensity for early, haematogenous metastases to distant organs, making it the GCT with the worst long-term prognosis. In addition, intussusception and surgery delayed treatment and certainly compromised our patient’s prognosis although he had a really heavy burden of metastatic disease.

**Conclusion**

Intussusception from a metastatic GCT is an infrequent finding, and we present only the fourth such case published. Nevertheless, surgeons need to be aware of this critical association. Urologists must exclude GIT involvement in testicular cancers and, similarly, surgeons need to exclude any testicular tumours in young men presenting with unexplained upper GIT haemorrhage, bowel obstruction or intussusception.

**Conflict of interest**

The authors declare no conflict of interest.

**Funding source**

The authors received no financial support for the research, authorship, and/or publication of this article.

**Informed consent**

Written informed consent was obtained from the patient’s family for the anonymised information and the accompanying images published in this article.

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