Metastatic seminoma presenting as flank pain

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Abstract

Seminoma is the most common single histological sub-type of testicular carcinoma. Patients usually present with a painless lump and stage I disease. We describe a case of an incidental metastatic seminoma in a 28-year-old man post-renal trauma with a dramatically elevated β-human chorionic gonadotropin (βHCG). His βHCG level has returned to normal post-orchidectomy and chemotherapy.

Introduction

Seminoma is the most common testicular tumour and is highly curable. It affects post-pubertal men and its highest incidence is seen in Denmark (11.5 per 100 000).[1] As with all testicular tumours, it usually presents as a painless unilateral testicular mass and risk factors include undescended testis, family history, Caucasian race, mumps, presence of a contralateral tumour and testicular intra-epithelial neoplasia. There are 3 histological sub-types of a pure seminoma (85%): classic, anaplastic, and spermacytic. For prognosis they are divided into good and intermediate, with no patients classified as poor prognosis. Seminomas can be associated with elevated human chorionic gonadotropin (HCG) and lactate dehydrogenase (LDH), but elevation of alpha feta protein (AFP), however, excludes the diagnosis of a pure seminoma.

Case report

A 28-year-old man presented to the emergency room with a 6-week history of right-sided abdominal and flank pain after a fall onto his lower back. This was not relieved with anti-inflammatories. He had no lower urinary tract or gastrointestinal symptoms; however, he had a significant history of weight loss over the previous 4 months. He was a smoker of 15 pack-years and had no relevant medical, surgical or family history. On examination his observations were normal and he had a tender mass/swelling in the right lumbar and upper quadrant regions. The external genitalia were normal, except for a small right testicle. His white cell count and creatinine were elevated at 15.7 G/L and 111 μmol/L, respectively. Alkaline phosphatase was normal. A computed tomography (CT) abdomen with intra-venous contrast revealed a 13.5 × 11-cm multi-loculated urinoma surrounding the right kidney with significant right hydroureteronephrosis down to the distal ureter, where a soft tissue mass was noted in the pelvis (Fig. 1).

The urinoma was drained percutaneously under ultrasound guidance and the drain was placed on free drainage. He then proceeded to a rigid cystoscopy and right flexible ureteroscopy. We were unable to pass the ureteroscope past the distal ureter due to extrinsic compression, but a 6 to 24-mm JJ stent could be inserted over a wire. A repeat CT scan, including the thorax, abdomen and pelvis with oral and intra-venous contrast, demonstrated a reduction in the size of the urinoma and a 6 × 7-cm lobulated soft tissue mass in the right lateral pelvis displacing the ureter posteriorly and compressing it resulting in significant hydroureteronephrosis (Fig. 2). There was no paraaortic lymphadenopathy and the other abdominal viscera were normal.

Despite the normal scrotal examination, an ultrasound was arranged and serum tumour markers assayed. His ultrasound revealed a multi-focal abnormality in the right testes with decreased echogenicity and calcification. His serum AFP was 0.9 ng/mL, βHCG 32.975 IU/L and LDH 244 U/L. We then proceeded to perform a radical inguinal orchidectomy, the histology of which was reported as a 8-mm pure seminoma with intra-epithelial germ cell neoplasia (IGCN) on a background of marked fibrosis and testicular atrophy.

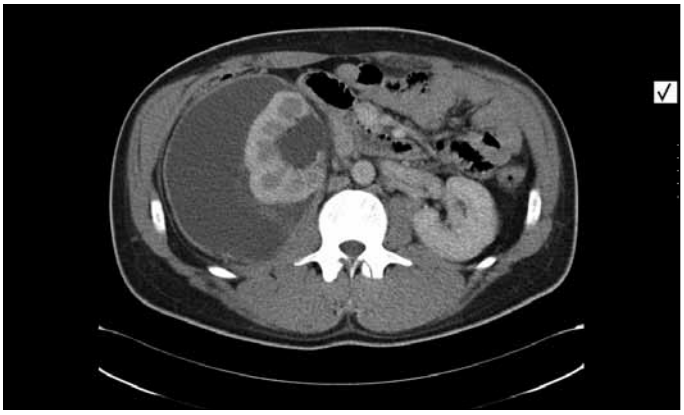

Fig.1. A computed tomography scan demonstrating large multi-loculated urinoma surrounding the right kidney.

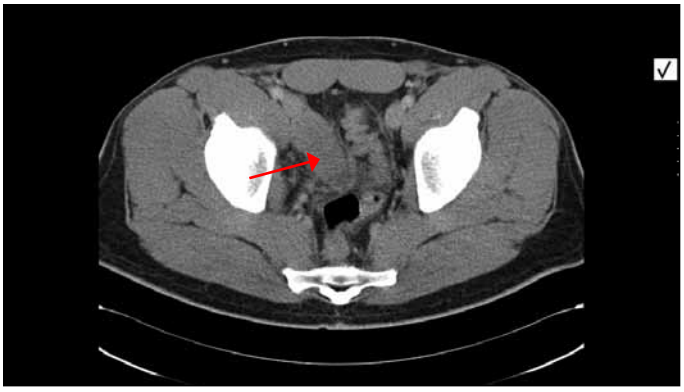

Fig. 2. A computed tomography scan demonstrating soft tissue mass in the right sided pelvic mass compressing the ureter.

The pathological stage of the primary tumour was pT1 with clinical stage T1N0M1aS2 (stage IIIA), and he was classified in the good prognosis group as per International Germ Cell Cancer Collaborative Group (IGCCCG) staging system.

On postoperative day 5, the βHCG had dropped to 17.222 IU/L and then fell further to 15.558 IU/L on day 7. A medical oncology referral was requested and the patient was assessed in the fertility clinic for sperm banking. He was unfortunately found to be azospermic. He underwent 3 cycles of bleomycin, etoposide and cisplatin (BEP) and 1 of EP (to reduce risk of bleomycin-induced pulmonary toxicity), with twice-weekly tumour markers. His βHCG was 13530 IU/L on day 1 of his first cycle of BEP. On day 1 of cycle 2, his βHCG had decreased to 553 IU/L. The βHCG had normalized prior to his third cycle of BEP. A follow-up CT at 3 months revealed resolution of his right-sided pelvic mass and his JJ stent was removed post-completion of his chemotherapy (Fig. 3). He is currently well and remains in remission 1 year later; his βHCG level has reduced to <1 IU/L (Fig. 4).

Discussion

Seminomas account for 55% of germ cell tumours[2] and typically present in men between the ages of 15 and 35. About 80% present with stage I disease, which is highly curable.[3] Our case is an unusual presentation of an incidental pure seminoma, with a pelvic lymph node metastasis causing hydronephrosis and no history of scrotal surgery. Other rare presentations of seminomas include cutaneous and perineal metastases, in addition to paraneoplastic syndromes including polycythaemia.[4-6] Hardey and colleagues reported a case of metastatic seminoma presenting as an incidental renal mass.[7]

βHCG is produced by the syncytiotrophoblasts and it has a half-life of 24 to 36 hours. Choriocarcinoma is frequently associated with a markedly elevated βHCG, but this marker is only elevated in 10% to 20% of seminomas, and it is uncommon to find levels above 500 IU/L.[8,9] βHCG level is thought to be related to tumour burden expression in metastases.[10] It was previously associated with a poor prognosis; however, several studies have demonstrated no impact on survival.[11,12] AFP is produced by yolk sac cells and is elevated in 50% to 70% of non-seminomatous germ cell tumours (NSGCTs). It is not elevated in seminomas and its elevation excludes a diagnosis of pure seminoma despite pathology, and subsequently patients must be treated as NSGCTs (or mixed tumours). LDH is not specific, but it is raised in 80% of patients with advanced disease.[13] Serum markers should be checked preoperatively and the European Association of Urology (EAU) guidelines[14] include levels as part of the TNM staging system. Levels should also be recorded at day 5 and 7 postoperatively and during follow-up to assess response to treatment and residual disease. Radiological staging is

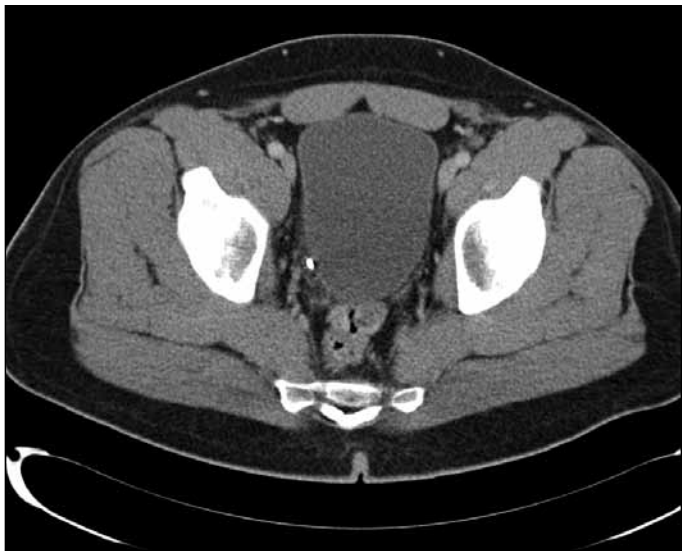

Fig. 3. A computed tomography scan demonstrating resolution of the soft tissue mass in the right pelvis, post-chemotherapy. Right JJ stent in situ in the right ureter.

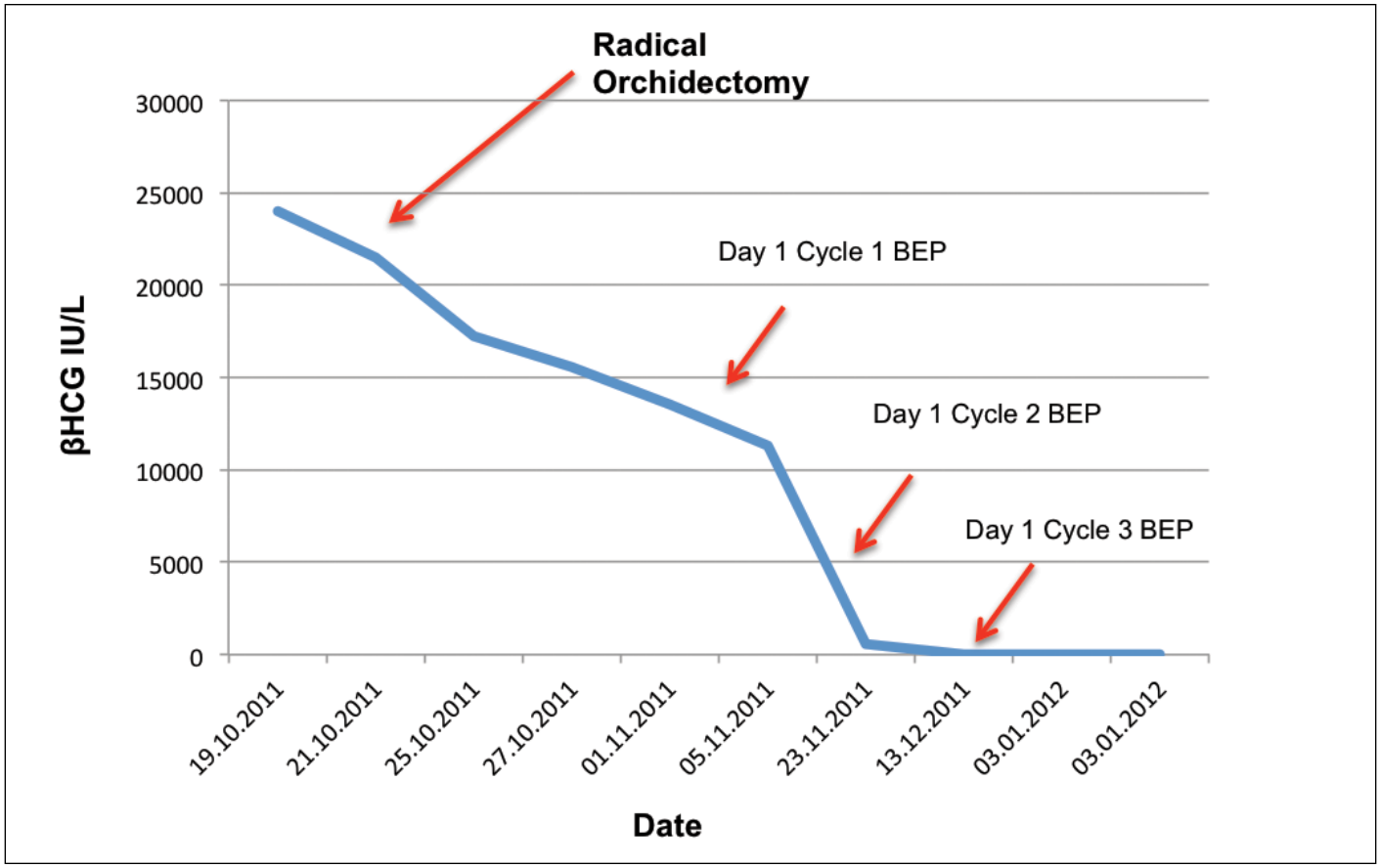

Fig. 4. Graph of serum β-Human Chorionic Gonadotropin levels pre- and postoperatively and during chemotherapy.

performed with CT imaging of the chest, abdomen, and pelvis. Patients are staged from I to III using a combination of pathological, radiological, and serum marker levels. Stage I disease has no lymph node or visceral metastases, Stage II has lymph node involvement but no visceral metastases, while Stage III has visceral metastases (except certain subgroups of Stage IIIB and IIIC which have both heavy lymph node burden and high serum tumour markers, see EAU 2011 guidelines 2011).[14]

Spontaneous regression of germ cell tumours is a well-known phenomenon. Histologically this may be visualized as a fibrotic scar in association with IGCN. Other supportive features include atrophy, calcification and lymphoplasmacytic infiltrates and prominent vascularity within the scar.[15] There is a possibility that this tumour may originally have had a non-seminomatous component that regressed or burned out, and lead to the development of a non-seminomatous metastases explaining the dramatically elevated βHCG that reduced to normal after chemotherapy.

Testicular lymphatic drainage relate to the testicular embryological descent. The lymphatic vessels of the testes follow the gonadal vessels ascending through the spermatic cord, ending at the para-aortic and para-caval nodes at the renal hilum. Testicular tumours metastasize via the para-aortic nodes, which bypasses the pelvic node pathway. Disease may then spread downwards towards the aortic bifurcation. Aberrant lymphatic drainage of the testes may occur following surgery (e.g., orchidopexy).[16] Our patient was very unusual as his pelvic lymph nodes were affected without no evidence of para-aortic nodal involvement.

Conclusion

We report a case of incidental metastatic seminoma. This case also had the unusual feature of pelvic lymph node metastasis in the absence of para-aortic lymphadenopathy. It is important to remember that testicular cancer can present in an atypical fashion, and that young men presenting with isolated pelvic lymphadenopathy may harbor occult testicular cancer.

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References

1. Parkin DM, Muir CS. Incidence in Five Continents. Comparability and quality of data. *IARC Sci Publ* 1992;(120):45-173.
2. Rusner C, Trabert B, Katalinic A, et al. Incidence patterns and trends of malignant gonadal and extragonadal germ cell tumours in Germany, 1998-2008. *Cancer Epidemiol* 2013;37:370-3. http://dx.doi.org/10.1016/j.canep.2013.04.003
3. Boujelbene N, Cosinschi A, Boujelbene N, et al. Pure seminoma: A review and update. *Radiat Oncol* 2011;6:90. http://dx.doi.org/10.1186/1748-717X-6-90
4. Tazi H, Badraoui M, Qasmi S, et al. Metastatic pure testicular seminoma of the skin. *Prog Urol* 2011;21:76-8. http://dx.doi.org/10.1016/j.purol.2010.02.006
5. Hosono TY, Kuratsukuri K, Nitta Y, et al. A case of primary extragonadal seminoma arising in the perineum. *Urol Int* 2006;76:364-7. http://dx.doi.org/10.1159/000092065
6. Kaito K, Otsubo H, Usui N, et al. Secondary polycythemia as a paraneoplastic syndrome of testicular seminoma. *Ann Hematol* 2004;83:55-7. http://dx.doi.org/10.1007/s00277-003-0745-7
7. Hadley DA, Cannon GH, Bishoff, JT. A solitary seminoma renal metastasis presenting as an incidental renal mass. *Urology* 2010;75:245-6. http://dx.doi.org/10.1016/j.urology.2009.05.068
8. Djeffal C, Demailly M, Tillou X, et al. Place of serum HCG assay in the follow-up of non-HCG-secreting testicular seminomas. *Prog Urol* 2008;18:654-6. http://dx.doi.org/10.1016/j.purol.2008.04.024
9. Weissbach L, Bussar-Maatz R. HCG-positive seminoma. *Eur Urol* 1993;23(Suppl 2):29-32.
10. Hartmann, M, Pottek, T, Bussar-Matz, R, et al. Elevated human chorionic gonadotropin concentrations in the testicular vein and in peripheral blood in seminoma patients. An analysis of various parameters. *Eur Urol* 1997;31:408-13.
11. Weissbach L, Bussar-Maatz R, Lohrs U, et al. Prognostic factors in seminomas with special respect to HCG: results of a prospective multicenter study. Seminoma Study Group. *Eur Urol* 1999;36:601-8.
12. Bruns F, Raub M, Schaefer U, et al. No predictive value of beta- hCG in patients with stage I seminoma—results of a long-term follow-up study after adjuvant radiotherapy. *Anticancer Res* 2005;25:1543-6.
13. Peyret C. Testicular tumours. Summary of onco-urological recommendations. *Prog Urol* 1993;2:60- 4.
14. Rouprêt M, Zigeuner R, Palou J, et al. European guidelines for the diagnosis and management of upper urinary tract urothelial cell carcinomas: 2011 update. *Eur Urol* 2011;59:584-94. http://dx.doi.org/10.1016/j.eururo.2010.12.042. Epub 2011 Jan 14.
15. Balzer BL, Ulbright TM. Spontaneous regression of testicular germ cell tumors: an analysis of 42 cases. *Am J Surg Pathol* 2006;30:858-65. http://dx.doi.org/10.1097/01.pas.0000209831.24230.56
16. Lundon D, Kelly B, Rowaiye B, et al. Nodal presentation of seminoma. *Ir Med J* 2011;104:121-2.

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