ELSEVIER

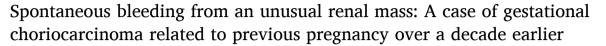
Contents lists available at ScienceDirect

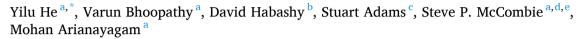
Urology Case Reports

journal homepage: http://www.elsevier.com/locate/eucr



Oncology





- ^a Nepean Urology Research Group, Nepean Hospital, Kingswood, NSW, Australia
- ^b Nepean Urology Research Group, Nepean Hospital, PO Box 63, Penrith, NSW, 2751, Australia
- ^c NSW Health Pathology, Nepean Hospital, Penrith, NSW, 2751, Australia
- ^d Fiona Stanley Hospital, Murdoch, WA, Australia
- ^e University of Western Australia, Crawley, WA, Australia

ARTICLE INFO

Keywords: Choriocarcinoma in female Gestational choriocarcinoma Single renal metastasis

ABSTRACT

Gestational choriocarcinoma is an uncommon trophoblastic malignancy, occurring in females after pregnancy, which is rarely encountered by urologists. It can be rapidly progressive, however metastases to other organs can occur after a prolonged latency period. We describe a rare case of solitary metastatic gestational choriocarcinoma presenting with spontaneous bleeding from a renal mass, over a decade after the associated pregnancy with a presumed sub-clinical primary tumour. This case demonstrates the importance of recognising gestational choriocarcinoma as a potential differential diagnosis of spontaneous bleeding renal mass in females of child-bearing age as a urologist given the often-aggressive nature of the disease.

Introduction

Choriocarcinoma is a rare but highly malignant trophoblastic neoplasm that often progresses very rapidly and can be classified as either gestational or non-gestational in origin. Non-gestational choriocarcinoma originates from germ cells of either the gonad or rarely extragonadal germ cells, while gestational choriocarcinoma only occurs in women and usually occurs after a complete hydatiform mole, spontaneous abortion or normal pregnancy. It most frequently originates in the uterus and is known to metastasise haematogenously and most commonly to the vagina, lung or brain. Involvement of the kidneys with choriocarcinoma is very rare. We report a rare case of a solitary metastatic renal gestational choriocarcinoma occurring in the kidney without an obvious original primary tumour, presenting with spontaneous retroperitoneal haemorrhage over a decade after the associated pregnancy.

Case presentation

A 37-year-old female presented to the emergency department with acute right sided abdominal and flank pain. Abdominal examination

revealed a tender right upper quadrant and right flank. An initial diagnosis of ectopic pregnancy was suspected based on a beta HCG of 2526IU/L and an abdominal and pelvic ultrasound that demonstrated no intrauterine gestational sac and a right retroperitoneal collection. She was otherwise in good health, apart from a long history of renal angiomyolipoma that had been managed conservatively over 10 years, and she previously had two normal pregnancies five and 11 years ago. A diagnostic laparoscopy was performed that demonstrated no evidence of ectopic pregnancy, however retroperitoneal bruising near the ascending colon was noted. A renal ultrasound subsequently showed a subscapular haemorrhage with a heterogenous mass in the lower pole of the right kidney (Fig. 1). Given her long history of angiomyolipomata, it was considered that this mass may be an angiomyolipoma, and her presentation may have been due to spontaneous bleeding from this during an early pregnancy. She was managed conservatively, however over the course of her follow up her beta HCG continued to increase rapidly to 17000 IU/L over the course of four weeks.

At this stage a multiphase CT was arranged that showed a 5 cm heterogenous enhancing cortical mass in the lower pole of the right kidney, with an associated large subcapsular haematoma but no evidence of metastatic disease (Fig. 2). Given the absence of fat within the

E-mail address: lulu_rose21@hotmail.com (Y. He).

https://doi.org/10.1016/j.eucr.2021.101614

Received 22 January 2021; Received in revised form 15 February 2021; Accepted 18 February 2021 Available online 17 March 2021

 $^{^{\}ast}$ Corresponding author.

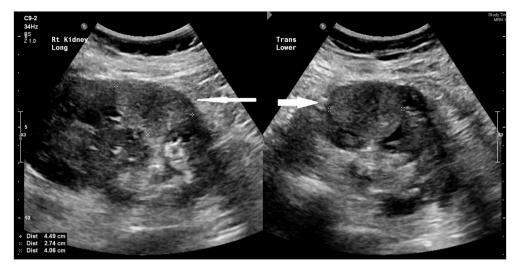


Fig. 1. Renal ultrasound showed a heterogenous mass arising from the lower pole of the right kidney (Solid arrows).

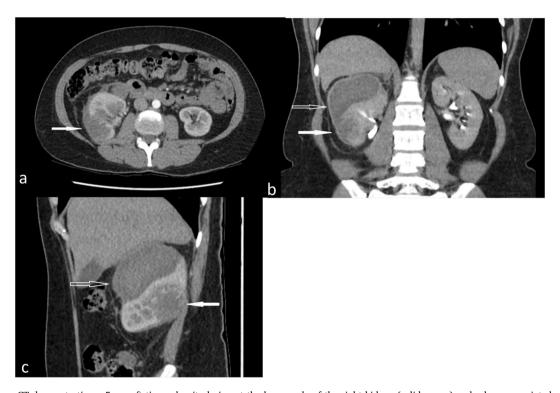


Fig. 2. Multiphase CT demonstrating a 5cm soft tissue density lesion at the lower pole of the right kidney (solid arrow) and a large associated subcapsular haematoma (hollow arrow).

lesion, it was suspected to be a primary renal cell carcinoma and so she proceeded to an urgent laparoscopic right radical nephrectomy.

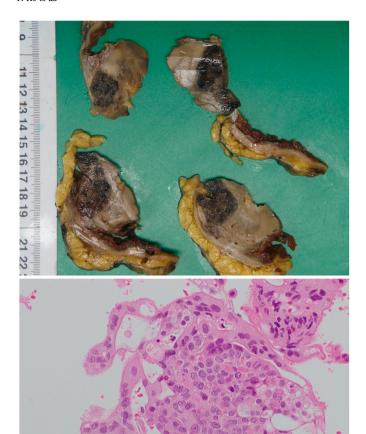
The histopathology revealed a malignant choriocarcinoma consisting of a biphasic proliferation of malignant cytotrophoblastic and syncytiotrophoblastic cells (Fig. 3). The majority of the tumour consisted of blood clot and fibrin, and the surgical margins were negative. Further genetic fingerprinting of the tumour against the DNA of her partner and children was performed. This was performed by DNA amplification of tagged microsatellite markers in a multiplex reaction analysed by laser-induced capillary electrophoresis. These genotyping studies revealed that this tumour was a gestational choriocarcinoma related to her first pregnancy from 11 years earlier.

Unexpectedly, her beta HCG remained high following her nephrectomy. A repeat CT was arranged four weeks later that unfortunately

revealed disseminated disease involving the liver, lung and multiple subcutaneous nodules. She was subsequently treated with systemic chemotherapy with an excellent response, and subsequent imaging and serial beta HCG monitoring has not shown evidence of recurrence over the following 12 months.

Discussion

Gestational choriocarcinoma is a rare tumour that may present with metastatic disease in the absence of a primary uterine tumour after any pregnancy. Although extrauterine gestational choriocarcinoma has been described, involvement of the kidney is rare and more likely to occur in the later stages of the disease. Rarely, metastatic choriocarcinoma can present as a primary urological concern such as haematuria and flank



 $\textbf{Fig. 3.} \ \ \text{Macroscopic} \ \ \text{and} \ \ \text{microscopic} \ \ \text{appearance} \ \ \text{of} \ \ \text{renal} \ \ \text{gestational}$ choriocarcinoma.

pain(2,3). More commonly, choriocarcinoma tends to metastasise initially to other organs such as the lungs, vagina and brain. 1,3,4 Achieving the diagnosis in this case was difficult due to the unusual presentation of a single renal lesion with high beta HCG. Differential diagnosis at the time included a beta HCG-secreting renal cell carcinoma, which can occur as part of a paraneoplastic syndrome but is also a very rare entity. 5

Differentiating gestational and non-gestational choriocarcinoma using DNA fingerprinting is essential, as they have different prognoses and sensitivities to chemotherapy $^{\rm I}$. Identifying the pregnancy that was responsible for the choriocarcinoma helped to identify the long dormant

period of 11 years in our patient. A long latency with gestational choriocarcinoma has previously been reported, with a latency of up to 23 years being observed. 3,4

The aggressive nature of this disease in this patient, with spontaneous bleeding and rapidly evolving metastases, is consistent with previously reported cases of renal metastases^{2,3}. This patient demonstrated an excellent response to chemotherapy, which is similar to previously reported gestational choriocarcinoma cases^{2,3}.

Conclusion

This case demonstrates that choriocarcinoma is an important, although rare, diagnosis that should be considered in any woman with spontaneous bleeding from a renal mass, particularly in the context of an elevated beta HCG. This case is particularly unique given the misleading history of renal angiomyolipomata and the absence of evidence of the disease until over a decade after the responsible pregnancy.

Consent

Consent has been obtained from the patient to use their deidentified clinical history, pathology and imaging findings.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Declaration of competing interest

No conflict of interest is identified during the write up of this case report.

References

- Vereczkey I, Csernák E, Olasz J, Küronya Z, Szentirmay Z, Tóth E. Renal choriocarcinoma: gestational or germ cell origin? *Int J Surg Pathol*. 2012;20(6): 623–628.
- Karadeniz T, Topsakal M, Ozkaptan O, Cakir C. Bilateral renal choriocarcinoma in a postmenopausal woman. Korean J Urol. 2011;52(7):498–501.
- Li HM, Hou WC, Lai YJ, et al. Gestational choriocarcinoma with renal and pulmonary metastases lacking a primary uterine origin. *Taiwan J Obstet Gynecol*. 2016;55(6): 881–885. https://doi.org/10.1016/j.tjog.2015.08.028. Available from:.
- Sonobe H, Taguchi K, Ogawa K, Yoshioka T. Latent vaginal choriocarcinoma in a postmenopausal woman. Pathol Int. 1976;26(5):611–618.
- Mohammed Ilyas MI, Turner GDH, Cranston D. Human chorionic gonadotropinsecreting clear cell renal cell carcinoma with paraneoplastic gynaecomastia. Scand J Urol Nephrol. 2008;42(6):555–557.