

Case report

Open Access

Metastatic appendiceal adenocarcinoma presenting late as epididymo-orchitis: a case report and review of literature

Shashank Kulkarni¹, Andrew Coup², John B Kershaw² and Noor PN Buchholz*³

Address: ¹Dept. of Urology, United Lincolnshire Hospitals, Lincoln/ UK, ²Dept. of Pathology, United Lincolnshire Hospitals, Lincoln/ UK and ³Dept. of Urology, St. Bartholomew's & The Royal London Hospitals, London/ UK

Email: Shashank Kulkarni - shashank_kulkarni@hotmail.com; Andrew Coup - andrew.coup@ulh.nhs.uk; John B Kershaw - john.kershaw@nlg.nhs.uk; Noor PN Buchholz* - nielspeter@yahoo.com

* Corresponding author

Published: 04 March 2004

Received: 04 October 2003

BMC Urology 2004, 4:1

Accepted: 04 March 2004

This article is available from: <http://www.biomedcentral.com/1471-2490/4/1>

© 2004 Kulkarni et al; licensee BioMed Central Ltd. This is an Open Access article: verbatim copying and redistribution of this article are permitted in all media for any purpose, provided this notice is preserved along with the article's original URL.

Abstract

Background: Whereas testicular metastases are in themselves a rare entity, testicular secondaries from an appendiceal carcinoma have not yet been described. The case also illustrates the diagnostic dilemma of a tumour presenting as epididymo-orchitis.

Case presentation: The authors present a case of an appendiceal carcinoma that, two years after radical therapy, manifested as a secondary in the testis. It was misdiagnosed as an epididymo-orchitis and was only revealed through histology.

Conclusions: Practitioners need to remember that long-standing testicular inflammation may result form secondary tumours. Even "exotic" primary tumours in the medical history of the patient must give rise to an increased suspicion threshold.

Background

Rarely, a testicular mass, whether painful or painless, represents a metastasis. In a series of 85 testicular tumours, only 10% were secondaries. Less than half of those actually represented the initial presentation of a tumour [1]. On the other hand, only 0.68% of solid tumours in an autopsy series of 738 patients metastasised into the testes [2]. Although carcinoma of the appendix spreads fast and has usually a poor prognosis [3], it has not been reported to spread into the scrotum. Therefore, we like to present this case of a late metastasis of a radically treated carcinoma of the appendix that presented as an epididymo-orchitis and was only finally diagnosed by histology.

Case presentation

A 72-year-old gentleman underwent appendectomy under the clinical picture of an acute appendicitis two years ago. Unexpectedly, histology revealed a mucinous adenocarcinoma of the appendix (figure 1). Subsequently, a right hemicolectomy was performed. Histology confirmed a poorly differentiated adenocarcinoma of the appendix (Dukes C1) pT3 pG3 pN1 cM0. The patient underwent adjuvant chemotherapy. At follow-up 18 months post-operatively, abdominal computer tomography (CT) did not show any evidence of recurrence.

Another six months later, the patient presented in the urology outpatient clinic with an inflammatory scrotal swelling which persisted in spite of antibiotic treatments by the general practitioner for by now eight weeks.

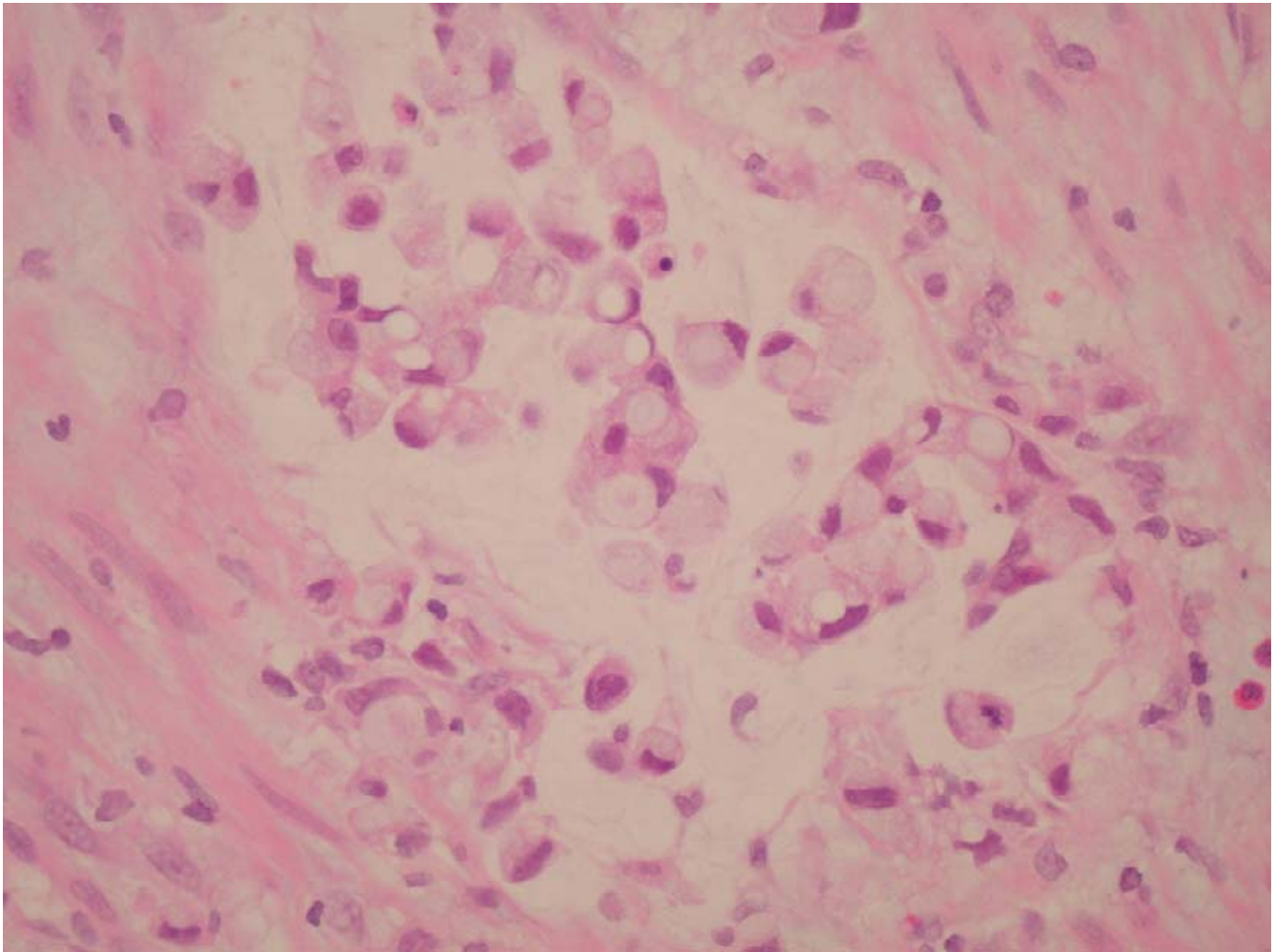


Figure 1

A representative section of the primary tumour showing adenocarcinoma cells with a signet-ring morphology and abundant extra-cellular mucin.

Tumour markers (α -fetoprotein, β -HCG, LDH) were not elevated. Scrotal and inguinal ultrasound revealed an unclear picture that could be attributed to long-standing inflammatory changes, but malignancy could also not be excluded. Therefore, the patient underwent scrotal exploration. The testis and spermatic cord were found to be severely inflamed and partially destroyed. Still during the operation, the surgeon thought this to be the result of a long-standing epididymo-orchitis. Due to the destruction of tissue and the involvement of the spermatic cord, a radical orchidectomy was performed. To our surprise, the histology revealed metastases of the appendiceal carcinoma in both, left testis and spermatic cord (figures 2 & 3). The patient was referred to the oncology department for further management.

Conclusions

Metastases to the testis are extremely rare. To our knowledge and after extensive literature review, only some 200 cases have been reported worldwide. Amongst these, the commonest ones are metastatic carcinoma of the prostate (34.6%), lung (17.3%), malignant melanoma (8.2%), colon (7.7%), and kidney (5.8%) [4]. In single cases, the organs of origin of the carcinoma were stomach, pancreas, penis, bladder, rectum, thyroid, ureter, bile duct, and liver. Occasionally, sarcomas and neuroblastomas have reportedly spread into the testis [5-8]. To our knowledge, although 7.7% of secondaries are from the colon, the appendix as original tumour-bearing organ has not yet been reported.

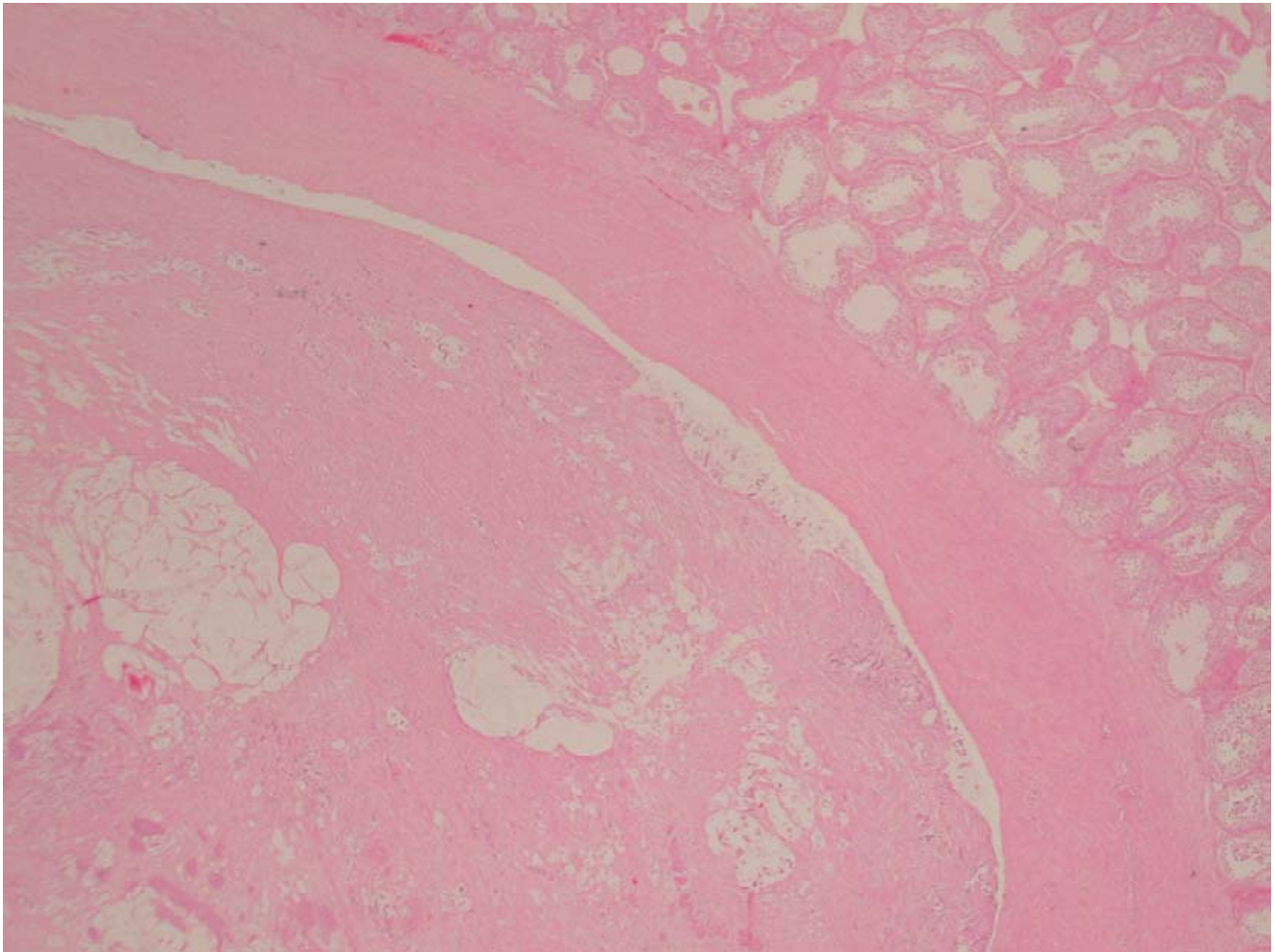


Figure 2
Metastatic tumour. At lower power, mucin lakes can be seen within fibrous tissue. Seminiferous tubules can be seen at the top right.

Adenocarcinoma of the appendix has generally a poor prognosis (5-year survival 50%) due to an early spread of disease that, in turn, is partially due to the low threshold of suspicion and difficulties of diagnosis prior to surgery [3]. It will mostly present and be diagnosed as an acute or chronic appendicitis, as it did in our case some two years ago. In spite of a relatively quick radical therapy in the form of hemicolectomy and adjuvant chemotherapy the patient relapsed with a distant metastasis into a rather unusual organ. The pathway of spread may have been haematogenously or lymphogenously. More likely though, in our case there may have been a continuous growth from the abdomen near the internal inguinal ring through the spermatic cord into the testis [9], as suggested by the histological involvement of the spermatic cord.

Clinically relevant is that the tumour did present and was treated for some time as an epididymo-orchitis. Indeed, the clinical picture, and the presence of inflammation and pain do not help to distinguish a benign from a malignant lesion [1]. Nor will the ultrasound be able to erase all doubt. We all learn that a persistent epididymitis can represent a tumour. In practice, this may need a reminder from time to time. In unclear testicular masses, even under the clinical picture of an epididymo-orchitis, a surgical exploration and/or resection is indicated after initial but short antibiotic treatment has proven unsuccessful.

Authors' contributions

SK collected the necessary data, reviewed the literature and wrote a first draft of the manuscript. AC and JBK

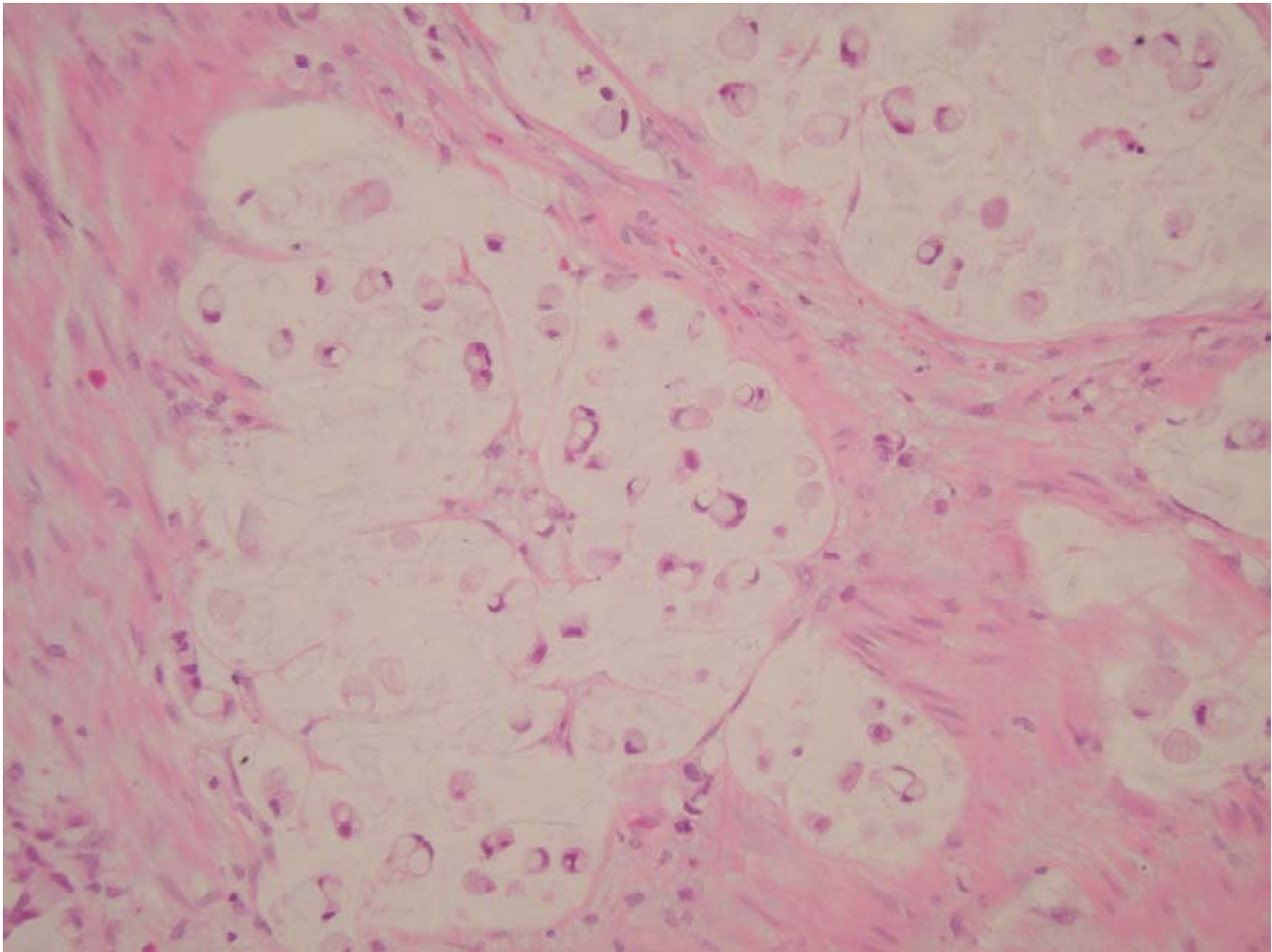


Figure 3

At higher power, the metastatic tumour also has a signet-ring cell morphology similar to that of the primary tumour in the appendix.

provided expert pathology input and histology slides. NPNB reviewed the literature, corrected, finalised and submitted the manuscript.

Acknowledgement

Written consent was obtained from the patient or his relatives for publication of the study.

References

- Lioe T, Biggard JD: **Tumours of the spermatic chord and paratesticular tissue. A clinicopathological study.** *Br J Urol* 1993, **71**:600-606.
- Garcia-Gonzalez R, Pinto J, Val-Bernal JF: **Testicular metastases from solid tumors: an autopsy study.** *Ann Diag Pathol* 2000, **4**:59-64.
- Amadio M, Lucarelli L, Bellone M: **Cancer of the appendix.** *Minerva Chir* 1991, **46**:1067-1070.
- Patel SR, Richardson RL, Kvols L: **Metastatic cancer to the testes: a report of 20 cases and review of the literature.** *J Urol* 1989, **142**:1003-1005.
- Brayan NP, Jackson A, Raftery AT: **Carcinoma of the sigmoid colon presenting as a scrotal swelling.** *Postgrad Med J* 1997, **73**:47-48.
- Rosser CJ, Gerrad E: **Metastatic carcinoma of the pancreas to the testicle.** *Am J Clin Oncol* 1999, **22**:619-620.
- Singh M, Samartunga H, Wright C, Guandalini I: **Prostatic carcinoma metastasising to the testis – an unusual pattern of spread.** *Br J Urol* 1995, **75**:803-804.
- Dutt N, Bates AW, Baithun SI: **Secondary neoplasms of the male genital tract with different patterns of involvements in adults and children.** *Histopathology* 2000, **37**:323-331.
- Hanash KE, Carney JA, Kelalis DP: **Metastatic tumours to testicles: routes of metastasis.** *J Urol* 1969, **102**:465.

Pre-publication history

The pre-publication history for this paper can be accessed here:

<http://www.biomedcentral.com/1471-2490/4/1/prepub>