Case report

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

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

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

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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.