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Case Report

Bilateral synchronous testicular germ cell tumours in a patient with bilateral cryptorchidism

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Bilateral testicular tumours are rare, and 80% of bilateral tumours are metachronous. The incidence of testicular tumours is high in cryptorchidism. Synchronous bilateral testicular tumours are rare, and bilateral synchronous testicular tumours in bilateral cryptorchidism extremely rare, probably not reported previously.

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The incidence of testicular germ cell tumours in the general population is 0.005%.[1] Patients with a history of testicular germ cell tumour in one testis have a 1 - 5% risk of developing a second germ cell tumour in the contralateral testis.[1] Synchronous bilateral testicular tumour is exceedingly rare, with only a few cases reported in the literature.[1-4] Bilateral synchronous testicular tumours in bilateral cryptorchidism have not been reported previously. We report a case of bilateral synchronous testicular tumours in both intra-abdominal testes.

Case report

A 30-year-old man presented to the surgical outpatient department of our hospital. The testes had been absent on both sides of the scrotum since birth. He complained of heaviness and mild pain in both flanks, but gave no history of anorexia or weight loss. He had been married for 5 years, with a history of normal intercourse but no children even with unprotected sex. On examination the abdomen was normal, secondary sexual characteristics were normally developed and the penis was well developed. The scrotal sacs were underdeveloped and empty. The results of haematological investigations, including kidney and liver function, were normal.

An ultrasound scan of the abdomen revealed bilateral enlarged intra-abdominal testes in the pelvic region. The left testis was situated just beneath the deep inguinal ring anterior to the external iliac vessels, and measured 6.1×3.2 cm. The right testis was located in the pelvis anterior to the right internal iliac vessels, and measured 7.3×4.6 cm. The parenchyma was hypo-echoic with clusters of fine high-echo foci microcalcification. The margins of the testes were well defined. Colour Doppler imaging showed bilateral enlarged hypo-echoic testes with a normal colour flow pattern. The parenchyma was hypo-echoic with tiny echogenic foci, and the margins of the lesions were well defined. A computed tomography scan of the abdomen (Fig. 1) revealed two mass lesions, one measuring 6.5×5.2 cm in the left iliac fossa and the other measuring 7.4×5.3 cm in the right side of the pelvis posterior to the urinary bladder.

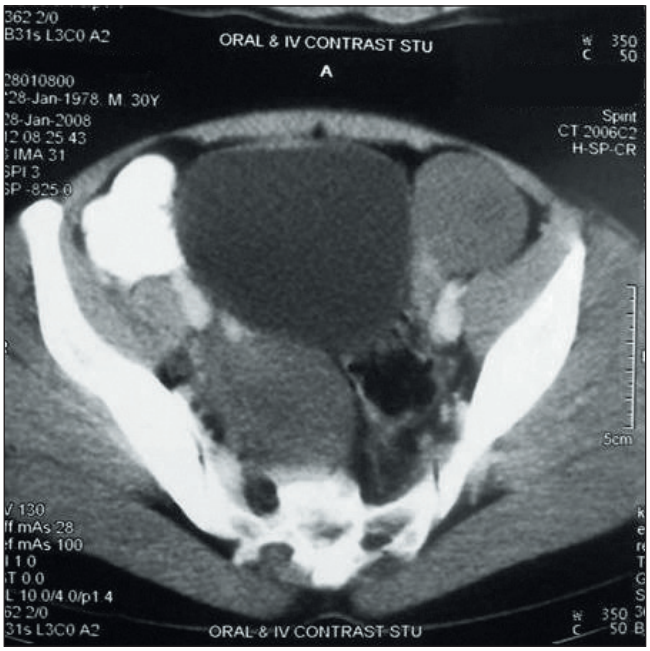

Fig. 1. CT scan showing enlarged testis in the left iliac fossa and pelvis.

The serum lactate dehydrogenase (LDH) level was 1 257 U/l, the serum α-fetoprotein (AFP) level 2.02 ng/ml and the serum beta-human chorionic gonadotrophin (β-hCG) level 0.38 mIU/ml. A seminogram revealed no spermatozoa, and a chest radiograph was normal. Laparotomy was performed and both testes were excised. Both testes were seen to be enlarged (Fig. 2), and the cut surfaces showed greyish-white homogeneous areas without any haemorrhage or necrosis. Histopathological examination of both testes showed the tumours to be poorly differentiated seminomas (Fig. 3). Postoperatively the patient was given PEB (cisplatin, etoposide and bleomycin) chemotherapy for 4 cycles. A follow-up scan of the abdomen after 8 months did not reveal any nodes or residual tumour. The serum LDH level returned to the normal range.

Discussion

Testicular carcinoma is the most common malignancy in men between 15 and 35 years of age. The incidence is 4.5/100 000 and has increased in the past century.[2] Patients who develop carcinoma

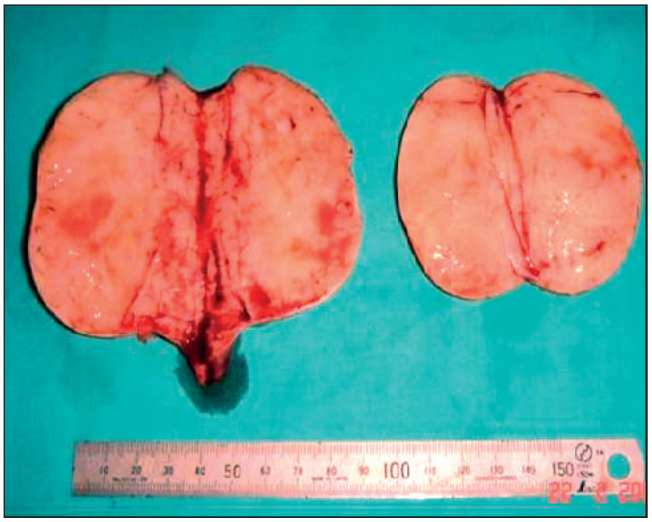

Fig. 2. Both testes are enlarged, with greyish-white homogeneous cut surfaces.

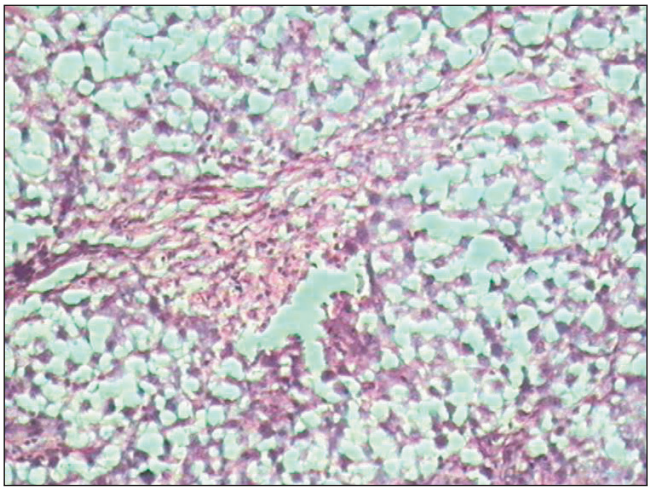

Fig. 3. Micrograph showing poorly differentiated seminoma.

in one testis have a 500 - 1 000 times increased risk of developing carcinoma in the contralateral testis.[2-5] Approximately 0.5 - 7% of patients with testicular carcinoma will develop a contralateral tumour.[2-5] Improved survival of patients with testicular carcinoma has led to an increased incidence of contralateral testicular tumour. A male with a tumour in a descended testis is estimated to have a 0.7% risk of developing a second testicular tumour on the contralateral side. The risk of a second testicular tumour increases to 15% when both the testes are undescended and is as high as 30% if the second testis is intra-abdominal.[4]

Bilateral testicular tumours occur metachronously in 80 - 85% of patients and synchronously in 15 - 20%.[2] Metachronous lesions are usually detected early by self-examination of the testis.[2] The literature suggests that the majority (two-thirds) of metachronous lesions occur within 5 years of the first diagnosis, although there are reports of second testicular tumours appearing as late as 23 and 25 years after the first diagnosis.[3]

Dieckmann *et al.* found that approximately 50% of metachronous tumours in their study had similar pathological features.[5] Seminomas accounted for 80% of cases with similar pathological features, and 20% were non-seminomatous tumours. Synchronous bilateral testicular tumours account for less than 1% cases of testicular carcinoma. Most synchronous testicular tumours are seminoma. Other tumours reported include embryonal carcinoma, teratocarcinoma and choriocarcinoma.[4]

Cryptorchidism is a known risk factor for testicular carcinoma, and the incidence of cryptorchidism in patients with bilateral testicular tumours is as high as 22%.[2] Most bilateral testicular tumours in undescended testes present as metachronous lesions, and synchronous bilateral tumours in bilateral cryptorchidism have not been reported. In a large series of 2 431 patients from the M D Anderson Cancer Center, 24 patients had bilateral tumours but none of these patients had a history of cryptorchidism.[1] Similarly, a study from Indiana University analysed computerised data on 2 088 patients with testicular carcinoma, of whom 21 had bilateral tumours but none had a history of cryptorchidism. Patients with synchronous testicular tumours should be treated on the basis of their clinical stage, pathological features and the most malignant component of the tumour.[4]

Survival in patients with bilateral testicular carcinoma does not appear to be worse than in patients with unilateral testicular carcinoma.[2-4] Follow-up is lifelong, and includes chest radiographs and measurement of the tumour markers AFP, β-hCG and LDH. Every patient treated with bilateral orchidectomy should receive a monthly intramuscular injection of 250 mg long-acting testosterone propionate.[4]

In conclusion, synchronous bilateral testicular tumours in bilateral cryptorchidism are very rare. Seminoma is the most common histological type. Principles of management are the same as those for primary germ cell tumour of the testis, and the clinical stage and histological type determine prognosis.

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