CASE REPORT

Paratesticular liposarcoma and contralateral angiolipoma in a 60-year-old patient

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SUMMARY

We present the case of a 60-year-old man who was referred with a 1-year history of a slow-growing right scrotal lump. Following surgical excision together with a radical orchidectomy, the histological diagnosis was of a paratesticular dedifferentiated liposarcoma. Radiological staging showed no metastatic disease. Six months later the patient presented with a new left scrotal lump, which was felt to be separate from the testis. Owing to the previous history of a liposarcoma and indeterminate ultrasound findings, this was surgically excised without orchidectomy. Histological analysis showed this to be an angiolipoma. The patient remained on clinical and radiological surveillance. Paratesticular tumours are rare neoplasms and liposarcomas of the spermatic cord only represent 7% of these lesions. Any link between lipomatous tumours and liposarcomas remains controversial and is still under investigation. This is the first case to report a paratesticular liposarcoma and a contralateral angiolipoma in the same patient.

BACKGROUND

Referrals for scrotal pain and lumps are common in general urology clinics. Even though, in most cases, there is no significant pathology found in the testicles, this case highlights the importance of a full assessment of all scrotal structures including the cord, epididymis and scrotal skin. This case presented initially with a right-sided scrotal lump, which was found to be a rare paratesticular dedifferentiated liposarcoma requiring an orchidectomy. Six months later, the patient re-presented with a new left-sided lump separate to the testis, which was histologically a paratesticular angiolipoma.

CASE PRESENTATION

A 60-year-old man was referred with a right scrotal lump that had been present for a year before it became painful and increased in size. There were no other associated symptoms during this period.

There was no previous history of trauma or surgery to the testes. When examined, the left testis appeared to be normal but the right testis was replaced on the upper pole by an irregular mass that felt partly cystic and partly solid. Abdominal and groin examination was unremarkable.

INVESTIGATIONS

An urgent ultrasound scan (USS) of the scrotum revealed three lesions: two hypoechoic mass lesions measuring 1 cm in the epididymal region, with considerable intralesional vascularity (figure 1), and one larger 2.6×1.6 cm lesion along the body of

Figure 1 Ultrasound scan demonstrating two lesions measuring 1 cm in the epididymal region, with intralesional vascularity.

the epididymis, with no intralesional vascularity (figure 2). The two small lesions were thought to be adenomatoid lesions and the larger one a possible haematoma. The testis was separate to the growths and was normal on USS. The left testis was normal apart from a 4 mm epididymal cyst (figure 3).

Routine blood tests and testicular tumour markers were normal.

DIFFERENTIAL DIAGNOSIS

Initial differential diagnosis was an adenomatoid tumour of the epididymis and a possible encapsulated haematoma representing the hypovascular lesion on USS.

TREATMENT

The patient underwent a right scrotal exploration and the three lesions were found to be on the cord.

Figure 2 Ultrasound scan demonstrating the third 2.6×1.6 cm lesion with no intralesional vascularity.

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Figure 3 Epididymal cyst on the left side.

Two were smaller, solid, and adherent to the epididymis and cord, and the larger growth, which was initially thought to be a haematoma, was felt to be a lipoma, intraoperatively, and was excised. A radical orchidectomy was also performed.

All three lesions were assessed histologically and revealed a paratesticular multifocal dedifferentiated liposarcoma. It was staged as pT2b Grade 3 and MDM2 gene amplification was positive. Microscopically, the dedifferentiated part of the tumour was composed of the two smaller but more solid lesions on the epididymis. These were composed of fascicles of spindle cells with moderate nuclear atypia that did not infiltrate the testis or the epididymis (figure 4). The larger lipomatous tumour was composed of well-differentiated adipocytes and scattered atypical cells (figure 5). The testis was not involved.

A staging CT scan did not reveal any lymphadenopathy in the abdomen, pelvis or groin, but there was a circular structure measuring 1.6 cm in diameter in the right scrotum. Re-excision of the right spermatic cord and scrotal skin showed an area of fibrosis, haemorrhage and giant cell reaction to suture material but with no residual liposarcoma. A decision for surveillance was made.

OUTCOME AND FOLLOW-UP

The patient made a good recovery from his second operation, with no complications. Six months following his operation, the patient re-presented with a new scrotal lump on the contralateral side.

When examined, this was a growth separate to the testis. An USS showed a 13×18×39 mm hyperechoic, fatty ovoid looking lesion with some internal vascularity, arising laterally from the

Figure 4 Dedifferentiated liposarcoma with fascicles of spindle cells and moderate nuclear atypia, ×100 magnification.

Figure 5 Well-differentiated adipocytes and scattered atypical cells, ×100 magnification.

epididymis (figure 6). The patient underwent a surgical excision of the lesion with testicular preservation. A repeat staging CT scan did not reveal any metastatic disease.

Macroscopically this was fibrofatty tissue measuring 40×25×10 mm with no evidence of haemorrhage or necrosis. Microscopically there was a well-circumscribed encapsulated tumour composed of mature adipocytes with a few thin intervening fibrous septa (figure 7). There were also scattered groups of small capillary-like vessels, some containing fibrin thrombi (figure 8). There were no atypical features and the appearances were histologically consistent with a benign angioliipoma with no evidence of malignancy.

When reviewed again in outpatient clinic, the patient was well and is currently still on surveillance for the previous liposarcoma.

DISCUSSION

Liposarcomas are a rare sarcoma subtype with an incidence of approximately 1 case per 2.5 million population.¹ Liposarcoma of the spermatic cord only represents 7% of paratesticular sarcomas and just over 100 cases have been reported in the literature from 1952 to 2013.^{2–6}

They are malignant lesions derived embryologically from mesodermal tissue and can occur in any part of the body containing fatty tissue.² As they arise from the structures of the

Figure 6 Ultrasound scan demonstrating a 13×18×39 mm, fatty ovoid looking lesion with some internal vascularity.

Figure 7 Angiolipoma and scattered groups of small vessels, $\times 100$ magnification.

cord they can mimic an inguinal or scrotal tumour, but they are separate from the testis and epididymis.^{2,3} The histological type and grade of the sarcoma is significant when considering prognosis. From previous case series it was observed that 40% of patients with rhabdomyosarcoma of the spermatic cord developed metastatic disease due to a higher propensity for lymphatic and haematogenous spread.^{4,7} This is not so frequently observed in patients with liposarcoma.⁴

Two cases of liposarcoma and lipoma in the same tissue specimen have been reported in the literature, one in a paratesticular tumour and the other in an inguinal tumour.^{5,8} This, however, is the first case to report a paratesticular multifocal dedifferentiated liposarcoma and contralateral paratesticular angiolipoma in one patient.

The aetiology of liposarcomas still remains unknown. The hypothesis that there is a continuum between lipomas and well-differentiated liposarcomas has caused much debate. Even cases describing the presence of both a lipoma and a liposarcoma in a single specimen are reported rarely. In the earlier literature, a possible link between paratesticular tumours and trauma was reported but this has yet to be proven.⁹ Molecular studies, however, have found specific genes that are expressed only in liposarcomas, assisting in their differential diagnosis.^{5,8,10–12} Two studies by Boltze *et al*⁸ and Pilotti *et al*¹¹ showed that an increase in MDM2 protein expression was associated with

malignancy and dedifferentiation, supporting the fact that MDM2 is a reliable marker in distinguishing between ordinary lipoma and liposarcoma.¹²

USS provides little information on paratesticular sarcomas, as echodensity is not consistent in reported cases.^{2,4,5} The initial USS in our case characterised the two small solid tumours as adenomatoid tumours of the epididymis and the well-differentiated component of the liposarcoma as a haematoma. Adenomatoid tumour of the epididymis was a possible differential diagnosis as it is a less rare paratesticular tumour compared to sarcomas.⁷ CT and MRI assist in staging disease both locally and for metastases, but do not provide a definite diagnosis.^{5,6} Liposarcomas are rarely diagnosed preoperatively. Accurate diagnosis requires a full histological assessment.

Liposarcomas are locally aggressive tumours and a recurrence rate up to 50% has been reported.^{2,4} We re-resected the cord and scrotal skin to ensure the excision margins had no residual tumour. Initial radical orchiectomy has been performed in other cases with paratesticular sarcomas and is supported in the literature.^{2,4–6,13}

Radiotherapy has been used for local control in the literature, especially for liposarcomas, which are the most radiosensitive, though there is not enough evidence in the literature to support adjuvant radiotherapy or chemotherapy for metastatic paratesticular liposarcomas.^{4,5,13} Wide local excision with close surveillance is the treatment of choice for paratesticular liposarcomas.

Learning points

- Paratesticular tumours are rare but need to be considered when assessing a patient with a scrotal lump separate to the testis.
- Ultrasound imaging of the scrotum is still the primary investigation in any patient with scrotal pain and lump as it assesses size, echogenicity, vascularity and association with other scrotal structures.
- Paratesticular liposarcomas are rare tumours, and although they rarely metastasise, local recurrence is possible. Therefore, close surveillance is necessary.
- Further molecular studies are needed to investigate the aetiology of liposarcomas and stratify risk factors.

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Figure 8 Fibrin thrombi within the scattered groups of small capillary-like vessels, $\times 400$ magnification.

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