Bilateral fibrosarcoma of the epididymis

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SYNOPSIS The tenth case of primary fibrosarcoma of the epididymis is reported. Contrary to most previous cases, excision biopsy was followed by a lengthy tumour-free period, and a unique feature was bilateral epididymal recurrence.

Primary malignant tumours of the epididymis are rare by any standards, but perhaps one of the least common is fibrosarcoma. To date, only nine cases have been documented. A further single case is described.

Case Report

In 1954 a 40-year-old man sought medical advice following three weeks' swelling and discomfort in the left inguinal region. This was originally diagnosed as a cyst of the distal cord, but during the following three months the swelling enlarged and it became obvious that surgical intervention was necessary. At operation a mass adherent to the left epididymis was excised, but the epididymis and testis were preserved. The pathological diagnosis was myxosarcoma. No further treatment was undertaken, but the patient was observed for a year without evidence of recurrence.

In November 1970 he was admitted to hospital with a myocardial infarction, from which he recovered uneventfully. The admitting physician noted a hard, craggy mass at the upper pole of the left testis, but this finding was overshadowed by his more pressing cardiac problems and the matter was subsequently overlooked.

In January 1973 the patient, now aged 59, himself became aware of an intrascrotal mass. Clinical examination by a surgeon gave the impression of extensive tumour involvement of the left testis with spread to the right scrotal contents. The inguinal lymph nodes on both sides were palpable but not enlarged. Surgical exploration was arranged as a matter of urgency.

OPERATION

A radical left orchiectomy was performed. The testis was adherent to the scrotum, but complete resection was possible. Exploration of the right testis was deferred for two months, when radical orchiectomy again took place. The pathological diagnosis in each case was epididymal fibrosarcoma. Initially the operation wounds healed well, but in August 1973 a subcutaneous recurrence from the pubic region was excised. One year later the patient remains well with no clinical or radiological evidence of remaining local or distant neoplastic disease.

PATHOLOGY

The 1954 specimen weighed 90 g (the dimensions were not recorded). Histologically (fig 1) it consists of spindle-shaped and angular cells with a lightly eosinophilic cytoplasm often drawn out into irregular processes. Staining with Heidenhain's iron haematoxylin fails to demonstrate cytoplasmic striations. The nuclei vary considerably in size, shape, and staining properties; some are spherical and almost water-clear with a prominent eosinophilic nucleolus and a delicate chromatin network, but most have irregular contours and coarsely clumped chromatin and exhibit excessive basophilia. Mitotic figures are difficult to find and, when present, appear normal. Collagen fibres are distributed throughout the tumour in a haphazard way, giving it a rather open texture, but in a few areas the tissue is more compact, with broad seams of collagen running in a storiform pattern. Throughout the mass there are numerous thin-walled blood vessels, and occasional small scattered areas of haemorrhage are present. At the periphery there is a rather tenuous capsule consisting of relatively acellular compressed fibrous tissue. Normal epididymal structures are not

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Fig 1 *Original lesion (Haematoxylin and eosin $\times 304$).*

Fig 2 *Recurrence in left epididymis (H and E $\times 304$).*

present. Best Carmine and Alcian Blue stains fail to demonstrate mucin.

The final diagnosis was myxosarcoma, probably of low malignant potential.

The left orchiectomy specimen (6 Feb. 1973) consisted of the testis, epididymis, and 6 cm of spermatic cord. The epididymis was completely replaced by a firm, white mass, measuring $9 \times 5 \times 5$ cm, compressing the testis into a flattened, crescentic structure. Microscopically (fig 2) the tumour presents a much more aggressive appearance. The loose and open texture seen in the previous specimen is lost. The tissue is highly cellular, the cells being compact and spindle-shaped. Mitotic activity, aneuploidy, and other bizarre nuclear abnormalities are marked; giant nuclei are common. Hyaline necrotic areas are both larger and more numerous. Collagen production is less marked but the storiform pattern is more obvious. As before, there is a thin fibrous capsule but in areas this is transgressed by tumour. Identifiable epididymal tissue is absent, but there is no invasion of the testis.

When the 1954 sections were reviewed, the essential identity of the two tumours was recognized and the diagnosis of fibrosarcoma was applied.

The contralateral orchiectomy specimen (22 Mar. 1974) measured $6 \times 4.5 \times 4$ cm with 6 cm of spermatic cord. The epididymis was almost completely replaced by tumour, similar to the preceding specimen but with the additional feature of a light lymphocytic infiltrate with occasional germinal centres. Again, there was no testicular invasion.

The final specimen from this patient, the recurrent tumour nodule, measured $5 \times 6 \times 4$ cm and has an identical histological pattern.

Discussion

The rarity of epididymal fibrosarcoma is noteworthy. In an effort to gain some perspective, the files of the Pathology Department at Aberdeen University Medical School were searched through the years 1954-73. In this period, and excluding the present case, 12 primary epididymal tumours are documented. They are: adenomatoid tumour (6 cases), carcinoma (4 cases), leiomyoma and leiomyosarcoma (1 case each).

Gilbert, quoted by O'Brien (1942), accepted seven cases of primary fibrosarcoma out of 91 malignant epididymal neoplasms in the literature. However, we do not know the cases which he accepted or the criteria upon which he judged them. A thorough personal review yields only eight well documented cases, and these have been accepted on the basis of the histological description of the tumour, with or without photomicrographs. Gowing and Morgan (1964) noted an epididymal fibrosarcoma in their series of paratesticular tumours but gave no details of the case; consequently, it is excluded from this discussion. The table summarizes these acceptable cases and includes the salient details of the present example.

It is apparent that the tumour may occur at any age. In most cases the left side was affected, but an interesting observation is that the three youngest patients had right-sided neoplasms. The commonest presenting complaint was of a tender intrascrotal mass of relatively short duration, but three patients had evidence of extra-epididymal spread at the time of operation. All patients were treated initially by hemicastration, and four subsequently had radio-

Author (year)	Age at Diagnosis	Side affected	Length of History	Spread at Time of Diagnosis	Radiotherapy	Eventual Outcome
Belluzzi (1951)	15 yr	Right	6 mth	None	Not given	Well after 6 mth
Falkinburg and Kay (1954)	2 yr 4 mth	Right	8 mth	None	Given	Retroperitoneal and pulmonary metastases with death after 1 mth
Halpert and Thompson (1947)	3 yr	Right	3 mth	None	Given	Metastases to peritoneum and retroperitoneal tissues with death after 10 mth
Jessen and Storm (1955)	49 yr	Left	3 mth	Spermatic cord, iliac and femoral vessels	Not stated	Local recurrence after 2 mth; survival not stated
Lazarus (1938)	44 yr	Left	6 mth	None	Not given	Well after one year
Mendoza <i>et al</i> (1969)	62 yr	Left	3 yr	Pulmonary metastases	Not stated	Not stated
O'Brien (1942)	20 yr	Left	6 mth	None	Given	Metastases to retroperitoneum and bone; death after 3 mth
Somani <i>et al</i> (1965)	60 yr	Left	1½ yr	Intra-abdominal	Given	Metastases to bone; death after 4 mth
McCormack (present report)	40 yr	Left	3 wk	None	Not given	Bilateral epididymal recurrences removed after 19 yr; incisional recurrence 5 mth later; remains well 1 yr later

Table Epididymal fibrosarcoma—analysis of published cases

therapy either prophylactically or to established metastases.

One is impressed by the viciousness of this tumour. Four patients had died of disseminated malignancy within one year, whether or not there was apparent extra-epididymal spread at the time of diagnosis. In addition, the patient of Jessen and Storm (1955) had inoperable dissemination, and the patient of Mendoza, Levin, and Escobar (1969) had pulmonary metastases, both at the time of diagnosis. It is reasonable to assume that there was a rapidly fatal outcome in those patients also. Only two patients (Belluzzi, 1951; Lazarus, 1938) were stated to be well and free of tumour at the time of reporting, but follow-up on these cases was for six months and one year only.

The longest definitely known survival concerns the present case, when 19 years passed between excision of the primary tumour and removal of bilateral epididymal recurrences. Five months later a subcutaneous recurrence in the scar was removed, and one year later the patient remains well. However, he is being kept under close medical supervision since

the experience of other authors emphasizes the aggressive nature of this neoplasm.

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