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An Unusual Case of Unilateral Malignant Leydig Cell Tumour of the Testis

Thivi Vasilakaki[a] Lardas Michalis[b] Evangelia Skafida[a]
Elissavet Arkoumani[a] Eleftheria Delliou[a]
Xanthippi Grammatoglou[a] Prodromos Kontovourkis[a]
Virginia Papamichail[a] Konstantinos Stamatiou[b]

Departments of [a]Pathology and [b]Urology, 'Tzaneion' General Hospital of Piraeus, Zanni and Afentouli 1, Piraeus, Greece

Key Words
Leydig cell tumour · Malignancy · Mitotic activity · Nuclear atypia · Testis · Tumour size

Abstract
Leydig cell tumour is a benign testicular non-germ cell tumour, and malignant transformation is rare. We report a case of a 35-year-old man who came to our hospital with a painless left testicular mass measuring 1.2 × 1 cm. Histological evaluation of the tumour showed features of a malignant Leydig cell tumour but no infiltration beyond the capsule or metastasis. The small size of the tumour was remarkable.

Introduction

Leydig cell tumours are rare stromal tumours of the testis accounting for 1–3% of testicular neoplasms. About 10% of them are malignant. They exhibit a peak incidence in the preadolescent as well as in the older (>50 years) age groups [1–4]. The diagnosis of a malignant Leydig cell tumour is not always easy since no definite histological criteria for malignancy exist.

Case Report

A 35-year-old man presented to our hospital with a 3-month history of a painless left testicular mass. He had no gynaecomastia. Blood concentrations of chorionic gonadotrophin, fetoprotein and human placental lactogen were within the reference range. Physical examination and ultrasonography revealed a testicular mass measuring 1.2 × 1 cm. A left radical orchidectomy was performed and the specimen submitted for histopathological examination. On microscopic examination, a malignant Leydig cell tumour was found. The cells displayed acidophilic cytoplasm, intranuclear inclusions and increased

Dr. Konstantinos Stamatiou
2 Salepoula street
GR–18536 Piraeus (Greece)
Tel. +30 210 452 6651, E-Mail stamatiouk@gmail.com

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mitotic activity (>3/10 HPF). Many cells with large atypical pleomorphic nuclei could also be observed (fig. 1, fig. 2). There was no angiolymphatic invasion, foci of necrosis or extension beyond the capsule of the testis. The immunohistochemical study showed that the tumour cells were positive for vimentin, melan A and inhibin, and negative for $CKAE_1$, $CKAE_3$, $S_{100}p$, P63, CEA, AFP and actin. Ki67 was expressed in 10% of the malignant cells (fig. 3).

Retroperitoneal lymph node dissection was suggested, but the patient declined further surgery. One year later, he showed no evidence of metastasis.

Discussion

Leydig cell tumours are rare and only about 3% of them are bilateral. About 15–20% of the patients already present with metastatic disease, particularly in the lymph nodes, lung and liver [5]. These tumours may be hormonally active, and gynaecomastia is seen in 30% of the cases [2, 3, 6].

Leydig cell tumours in an undescended testis may exhibit only manifestation of endocrinological disorders such as gynaecomastia, impotence and loss of libido. However, among the 480 cases of Leydig cell tumours reported in the literature, only 20 cases were associated with cryptorchidism, and there is no evidence that undescended testes are more prone to develop Leydig cell tumours [2, 5, 7]. Features associated with malignancy include a large tumour size (5.7 cm), nuclear atypia, a mitotic count of >3/10 HPF, foci of necrosis, angiolymphatic invasion, infiltrative margins, DNA aneuploidy and an increased expression of Ki67/MIB-1 and p53 [2, 3, 8–10].

Our patient was young and did not fit into either of the known age incidence peaks. Microscopic features such as the severe nuclear atypia and increased mitotic activity (>3/10 HPF) favoured the diagnosis of malignancy. However, there was no angiolymphatic invasion or extension beyond the capsule of the testis. Also, the small size of the tumour was remarkable. One year after surgery, the patient was well with no evidence of metastasis.

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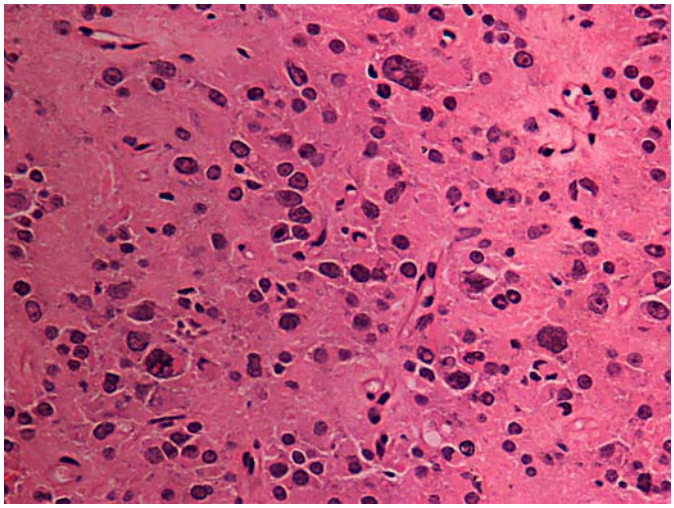

Fig. 1. Malignant cell tumour. Nuclear and cellular polymorphism and abnormal mitoses. ×400.

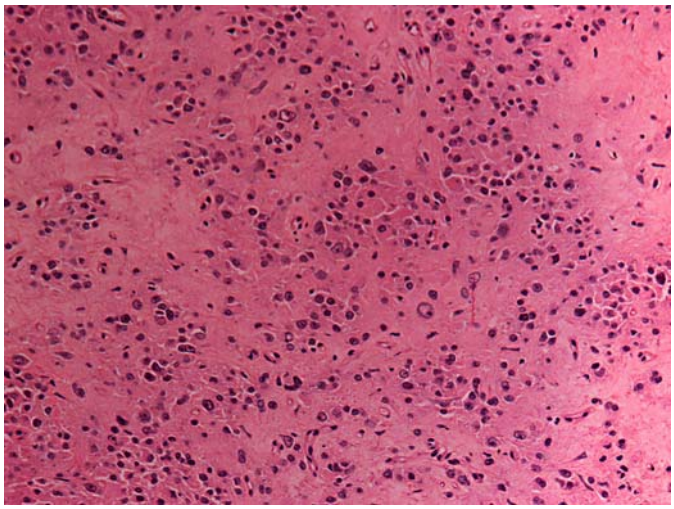

Fig. 2. Malignant Leydig cell tumour. ×200.

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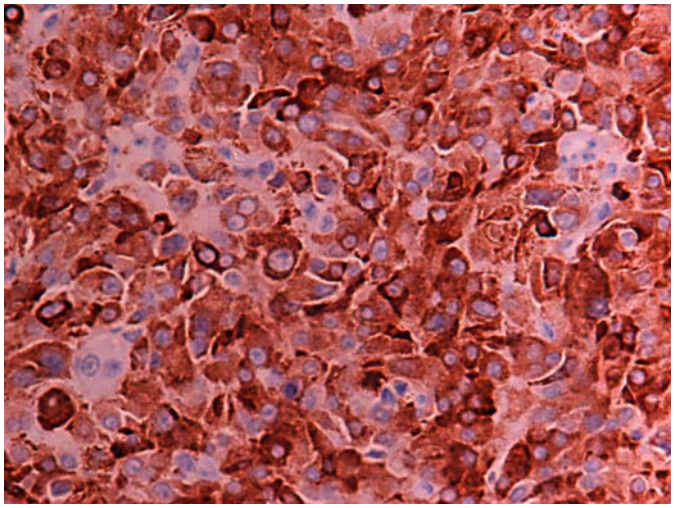

Fig. 3. Immunohistochemical staining for inhibin A. ×400.

References

1 Fink RS, Mann MS, Hopewell JP, Ginsburg J: Benign Leydig cell tumor and germ cell carcinoma in situ in a young man with gynaecomastia. Postgrad Med J 1984;60:66–69.

2 Efthimiou I, Mamoulakis Ch, Papageorgiou G, Kazoulis S, Prevedorou D, Kontogiorgos G, Christoulakis I: Unilateral malignant leydig cell tumor of testis in a patient with contralateral cryptorchidism. Urol J 2009;6:60–62.

3 Agrawal V, Sharma M, Bhatnagar D, Saxena S: Leydig cell tumor: an unusual presentation. Indian J Pathol Microbiol 2009;52:395–396.

4 Gruceyra Betriu G, Tegido Sánchez A, Duatre Ojeda JM, García De La Torre JP, De La Morena Gallego JM, Martínez Silva V, Leiva Galvis O: Leydig cell tumor: report of 8 cases and review of the literature. Actas Urol Esp 2002;26:36–40.

5 Albers P, Albrecht W, Algaba F, Bokemeyer C, Cohn-Cedermark G, Fizazi K, Horwich A, Laguna MP: Guidelines on testicular cancer. Available at http://www.uroweb.org/gls/pdf/Testicular%20Cancer%202010.pdf.

6 Carmignan L, Salvion R, Gadda F, Colecchia M, Gazzano G, Torelli T, Colpi GM, Pizzocaro G: Long-term followup and clinical characteristics of testicular Leydig cell tumor: experience with 24 cases. J Urol 2006;176:2040–2043.

7 Skakkebaek NE, Holm M, Hoei-Hanson C, Jørgensen N, Rajpert-De Myts E: Association between testicular dysgenesis syndrome (TDS) and testicular neoplasia: evidence from 20 adult patients with signs of maldevelopment of the testis. APMIS 2003;111:1–9.

8 Cheville JC, Sebo TJ, Lager DJ, Bostwick DG, Farrow GM: Leydig cell tumor of the testis: a clinicopathologic, DNA content, and MIB-1 comparison of nonmetastasizing and metastasizing tumors. Am J Surg Pathol 1998;22:1361–1367.

9 Sign V, Chandhary H, Srivastava A: Leydig cell tumor of the testis – a case report. Indian J Vrol 2004;20:166.

10 Tichoo SK, Tamboli P, Warner NE, Amin MB: Testicular and paratesticular tumors; in Weidner N, Cote RJ, Suster S, Weiss LM (eds): Modern Surgical Pathology. Philadelphia, Saunders, Elsevier Science, 2003, pp 1215–1256.