

ATYPICAL LYMPHATIC DISSEMINATION OF A TESTICULAR TUMOR FROM A CRYPTORCHID TESTICLE

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Summary.- OBJECTIVES: Atypical lymphatic spread of germ cell tumors to inguinal lymph nodes has been reported mostly related to prior surgical involvement of the inguinoscrotal region, as in orchiopexy.

METHODS: A patient with cryptorchidism and failed orchiopexy in his childhood was diagnosed with a testicular neoplasm. Inguinal orchiectomy was performed.

RESULTS: Pathological analysis showed classical seminoma spreading into a subcutaneous adjacent lymph node. Further metastatic disease was not found. Patient was treated with adjuvant chemotherapy.

CONCLUSIONS: We emphasize the need for accurate staging and a multidisciplinary approach when dealing with onco-urological patients presenting with atypical disease.

Keywords: Testicular neoplasms. Cryptorchidism. Inguinal lymph node metastasis.

Resumen.- OBJETIVOS: Existen casos descritos en la literatura de diseminación linfática atípica de un tumor testicular a ganglios inguinales, en la mayoría de las ocasiones asociados a una cirugía previa de la región inguinal o testicular, como es el caso de la orquidopexia en testículos criptorquídicos.

MÉTODOS: Presentamos el caso clínico de un paciente con testículo criptorquídico con orquidopexia previa fallida, en el que se diagnostica un tumor testicular y se realiza orquiectomía.

RESULTADOS: En el análisis histopatológico se objetiva un tumor de estirpe seminomatosa sobre testículo criptorquídico, así como una diseminación a estación linfática inguinal ipsilateral. El paciente es tratado con quimioterapia complementaria.

CONCLUSIONES: Se pretende mostrar la necesidad de establecer una correcta estadificación y el tratamiento más adecuado en los casos con este tipo de afectación linfática.

Palabras clave: Tumor testicular. Criptorquidia. Metástasis ganglionar inguinal.

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INTRODUCTION

Cryptorchidism is considered the most common alteration of male differentiation. Although it is not a pathology which is particularly serious, it has significant clinical relevance, both due to its incidence (approximately 0.8-5.8% in full-term infants) (1,2) as well as its possible complications, among them testicular cancer and infertility.

The association between cryptorchidism and testicular tumours is therefore well-known. It is estimated that the incidence of testicular cancer in cryptorchid testicles is between 3 and 48 times greater than in the general population according to the series (3), and this rate in-

creases if the testes are intra-abdominal (4). It has been shown that although orchiopexy decreases the incidence of testicular tumours, it does not eliminate the risk in these cases (5,6). The most common histopathological type, and which is most frequently associated with cryptorchidism, is seminomatous (7).

Lymphatic dissemination from the testicular tumour is to the retroperitoneal lymph nodes. Although not often, cases of lymphatic dissemination to ipsilateral inguinal lymph nodes have also been found, mainly in cases of previous surgery of the inguinal or scrotal area (8). Cryptorchidism, therefore, represents a risk factor for the development of testicular cancer and for dissemination to inguinal lymph nodes if the correction was previously performed by orchiopexy.

CLINICAL CASE

We present the case of a 37-year old male, with a history of failed right orchiopexy for an inguinal testicle in childhood, who attended the emergency department for inguinal discomfort of approximately one month's evolution. On physical examination, the left testicle appeared normal while the right inguinal testicle had an indurated region in the upper pole. The rest of the examination was unremarkable.

In view of the suspicion of a testicular tumour, it was decided to perform an ultrasound, which revealed the undescended right testicle, of normal size and regular contours, in the inguinal canal; a solid, hypoechoic, vascularised image was observed inside the testicle, suggestive of testicular neoplasia. The left testis was in the scrotal sac and did not show any alterations; no retrope-

ritoneal adenopathies or hepatic lesions were noted on ultrasound examination of the abdominal cavity.

Due to the findings, it was decided to admit the patient to hospital for right radical inguinal orchiectomy of the cryptorchid testis. During surgery, a small adenopathy visible on subcutaneous cellular tissue was observed and removed, along with skin with the scar from the previous surgery. The pre-operative tumour markers (LDH, alpha-fetoprotein and β -HCG) were normal.

During the post-operative period, an extension study was performed by thoracoabdominal tomography, in which neither adenopathies of a significant size or space occupying lesions were noted at any level, except for a small pseudonodular image at the base of the left lung which appeared to correspond to a scar.

The result of the anatomical pathology study of the testicular sample was testicular seminoma infiltrating the rete testis, sparing the tunica albuginea and epididymis and with lymphatic invasion (pT2), and non-tumour testicular parenchyma which showed intratubular germ cell neoplasia and histological alterations compatible with cryptorchidism. The surgical margin (spermatic cord) was tumour-free. In the excised lymph node, a granulomatous histiocytic reaction was found with isolated tumour cells in the sinuses and in one of the lymphatic vascular spaces; an immunohistochemical study was performed in which the atypical cells were positive for CD117 (c-kit) and placental alkaline phosphatase (PALP).

The case was presented at a joint session of the urological tumour committee, where it was decided to administer complementary treatment with polychemotherapy.

DISCUSSION

Cryptorchidism is one to the most widely known risk factors for the development of testicular cancer, so that almost 10% of testicular tumours appear on an undescended testis. It has been postulated that the increase in temperature in the inguinal canal or in the abdomen with respect to the scrotal sac may be the cause of both infertility and malignant transformation of these types of testicles (4). Thus, in childhood it is recommended to treat the condition by surgical descent of the testicle to the scrotum, which, although it does not eliminate the risk of degeneration, decreases its incidence (5) and permits better examination and early detection in the event that a tumour appears. In adult males, particularly after puberty, orchiectomy is recommended (9), due to the frequent functional non-viability and its stronger association with neoplastic disease, which has a peak incidence between 20 and 35 years of age. Certain authors though, believe that orchiopexy with adequate follow-up is also possible, provided there is no testicular atrophy and it is not located intraabdominally (10). All histological types

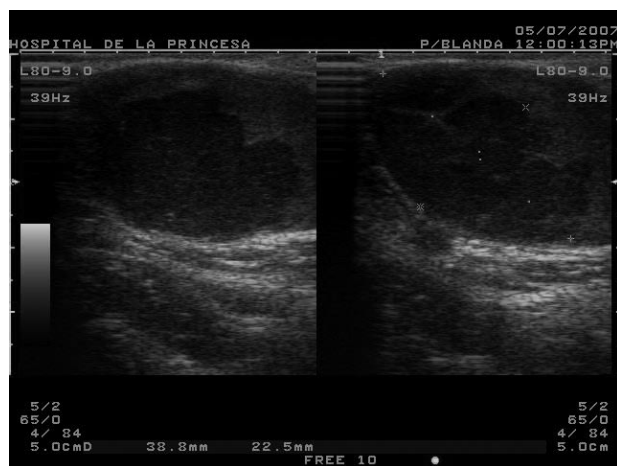


FIGURE 1. Ultrasound image where a hypoechoic mass can be seen in the parenchyma of the cryptorchid testicle.

may be found, although the most common is seminoma, and its prognosis will depend on both the histology and the initial stage (7). In developed countries, the presentation of an undescended testicle in an adult male is rare, as surgical correction during childhood is elective.

Lymphatic dissemination from the testicular tumour is to the retroperitoneal lymph nodes, although cases of atypical lymphatic dissemination to the inguinal lymph nodes have been documented. On most occasions, this atypical dissemination is associated with previous history of a testicular biopsy or prior surgery on the scrotal or inguinal area, as is the case of orchiopexy to correct cryptorchidism, although cases have been described in which this history of prior surgery is not found (11-13). Some of these cases present with tumour infiltration of the spermatic cord, which may facilitate dissemination to the inguinal region if tumour remnants remain after the surgery. Retrograde contralateral inguinal lymph node involvement from retroperitoneal masses has also been described (13).

The problem that arises as regards the correct staging and need for adjuvant treatment after orchiectomy in atypical lymphatic dissemination of testicular tumours has still not been resolved. The presence of inguinal lymph node involvement without evidence of disease of retroperitoneal lymph nodes indicates the appearance of new drainage pathways after testicular surgery, but it is logical to suggest the possibility that the same tumour cells that have migrated via this new pathway due to invasion of the lymph vessels, may have travelled to the anatomical lymph station. This gives us two therapeutic possibilities, both correct in a stage IIA: localised treatment with radiotherapy on the inguinal area, in which there is evidence of disease, or systemic treatment with polychemotherapy (etoposide, cisplatin, bleomycin), presupposing possible simultaneous involvement of the retroperitoneal lymph nodes, since the area that the radiotherapy should include would be too widespread in this case. In our case, the patient presented stage pT2 added to a poor prognostic factor as is invasion of the rete testis, which together with the possibility of retroperitoneal involvement led us to decide on polychemotherapy treatment (14).

CONCLUSION

We have tried to transmit the need to have a multidisciplinary approach to these cases, so that agreement can be reached on what would be the most suitable individualised treatment when we find ourselves with a testicular tumour with tumour invasion of the inguinal lymph nodes and without evidence of retroperitoneal lymphatic disease.

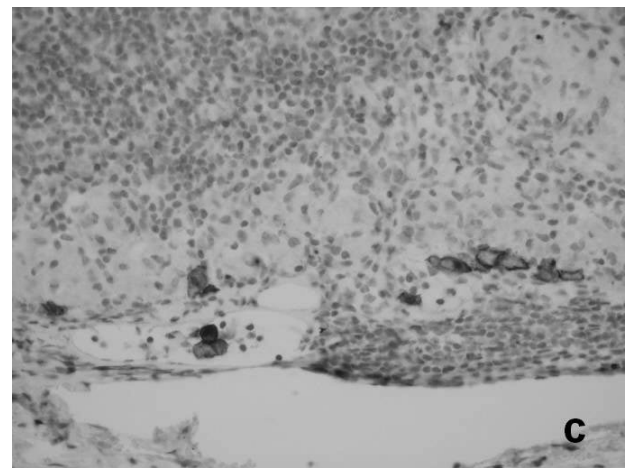
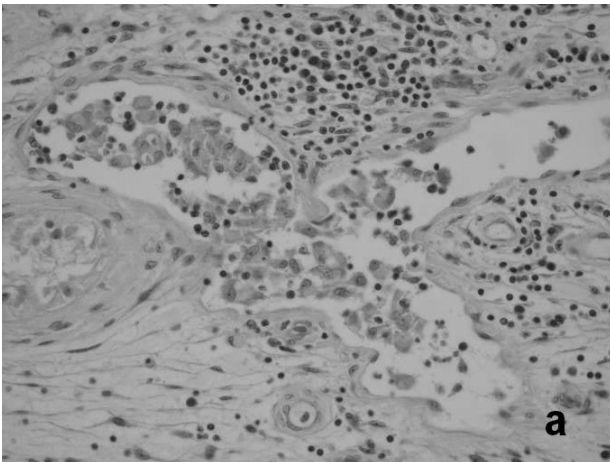


FIGURA 2. Microscopic image of: a: invasion of a lymphatic vascular space by tumour cells in the testicle; b: intratubular germ cell neoplasia (PALP+); c: tumour cells (CD117+ / PLAP+) in the inguinal lymph node, some of them inside a lymph vessel.

REFERENCES AND RECOMENDED READINGS (*of special interest, **of outstanding interest)

1. Cedron M, Duckett JW. The maldescended testicle. AUA update series 10 Lesson, 1991; 28:218.
2. Giwercman A, Bruu E, Frimodt-Moller CF, et al. Prevalence of carcinoma in situ and other histopathological abnormalities in testes of men with a history of cryptorchidism. *J Urol*, 1989; 142 (4):998.
3. Cristián Palma C, Cristóbal B, Maccioni R. Seminoma de testículo abdominal en un paciente adulto: reporte de un caso. *Actas Urol Esp*, 2007; 31 (2):160.
- *4. Batata MA, Whitmore WF Jr, Chu FCH, et al. Cryptorchidism and testicular cancer. *J Urol*, 1980; 124 (3):382.
- **5. Pettersson A, Richiardi L, Nordenskjold A, et al. Age at surgery for undescended testis and risk of testicular cancer. *N Engl J Med*, 2007; 356:1835.
6. Giwercman A, Grindsted J, Hansen B, et al. Testicular cancer risk in boys with maldescended testis: a cohort study. *J Urol*, 1987; 138 (5):1214.
- *7. Carmona Campos E, Regueiro López JC, Prieto Castro R, et al. Criptorquidia y cáncer testicular. *Actas Urol Esp*, 2000; 24 (1):49.
- **8. Gauwitz MD, Zagars GK. Treatment of seminoma arising in cryptorchid testes. *Int J Radiat Oncol Biol Phys*, 1992; 24 (1):153.
9. Alonso Domínguez FJ, Amador Sandoval BV, Fragas Valdés R, et al. Testículos no descendidos y cáncer. *Arch EspUrol*, 2005; 58 (4):365.
10. Granados Loarca EA, Esau ortega S. ¿Es necesaria la orquidectomía en los pacientes con testículo no descendido post-puberal?. *Actas Urol Esp*, 2005; 29 (10):969.
- **11. Klein FA, Whitmore WF Jr, Sogani PC, et al. Inguinal lymph node metastases from germ cell testicular tumors. *J Urol*, 1984; 131 (3):497.
12. Lanteri VJ, Choudhury M, Pontes JE, et al. Treatment of testicular tumors arising in patients with previous inguinal and/or scrotal surgery. *J Urol*, 1982; 127 (1):58.
- **13. Daugaard G, Karas V, Sommer P. Inguinal metastases from testicular cancer. *BJU Int*, 2006; 97:724.
14. Huddart RA, et al. Testicular seminoma: ESMO clinical recommendations for diagnosis, treatment and follow up. *ESMO Guidelines Working Group Ann Oncol*, 2007; 18 (2):1140.