



Symptomatic spinal cord compression in spinal epidural metastasis of a testicular germ cell tumor: a case report with review of the literature

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Background: Metastatic germ cell tumors particularly in the bone are rare entities. They occur in young men and classified in the group of poor prognosis. Bone metastases are responsible for a significant functional handicap and a deep alteration in the quality of life of young patients. Metastatic germ cell tumors are rare entities with a high cure rate due to their extreme chemosensitivity. To our knowledge, epidural metastasis of this tumor has not yet been reported in the English medical literature. Tumors of the breast, lung and prostate are the most notorious for metastasizing in the epidural-spinal space.

Case Description: We report a clinical case of spinal cord compression secondary to metastasis in the spinal epidural space of a testicular germ cell tumor in a 23-year-old. Patient whose evolution was marked by a decrease in tumor markers and a clear clinical improvement with disappearance of lower back pain and horsetail syndrome after starting the treatment with decompressive radiotherapy and chemotherapy based on bleomycin cisplatin and etoposide.

Conclusions: Through this, we want to emphasize that an epidural metastasis of this tumor is possible, and that it must be part of the diagnoses to be evoked in the face of metastatic spinal cord compression, which potentially allows to avoid a significant functional handicap and a profound alteration in the quality life of young patients.

Keywords: Germline tumor; bone metastases; epidural compression; case report

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Introduction

Testicular cancers represent 1 to 2% of cancers in men and 3.5% off urogenital cancers (1).

Germ cell tumors of the testicle are divided into two large subgroups whose therapeutic management is relatively different. Pure seminomas represent 55% of these germ cell tumors and non-seminomatous germ cell tumors 45% grouping under the same entity different tissue components such as embryonic carcinoma, choriocarcinoma, vitelline tumor and teratoma. There are great variations in the distribution of these different components within the testicular tumor. The extension assessment will make it

possible to specify the possible secondary locations of this cancer, whether it is its extension by the lymphatic route, primarily involving the retroperitoneal para-aortic nodes or visceral metastatic.

Metastatic germ cell tumors particularly in the bone are rare entities and classified in the group of poor prognosis. Bone metastases are responsible for severe complications, such as fracture, spinal cord compression and pain requiring the use of a local strategy (surgery or radiotherapy). The presence of bone metastases has a negative impact on the quality of life of patients and on the overall prognosis of the disease.



Figure 1 Cross section of a spinal cord MRI showing spinal cord compression at the level of the fifth lumbar vertebra. MRI, magnetic resonance imaging.

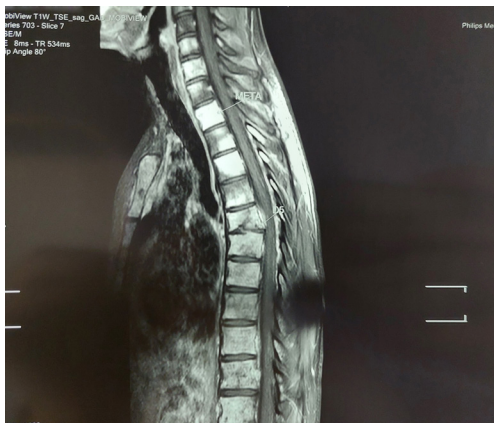


Figure 2 Sagittal section of a spinal cord MRI showing spinal cord compression at the level of the fifth dorsal vertebra. MRI, magnetic resonance imaging.

We report a clinical case of spinal cord compression secondary to metastasis in the spinal epidural space of a testicular germ cell tumor in a 23-year-old patient. We present the following case in accordance with the CARE reporting checklist (available at <https://atm.amegroups.com/article/view/10.21037/atm-2022-51/rc>).

Case presentation

A 23-year-old patient, WHO 1, ASA 1, without notable pathological history, who consulted for a feeling of testicular heaviness with unstated weight loss without other associated signs.

The clinical examination found a conscious patient,

stable on the hemodynamic and respiratory plan, on the urogenital examination: a large painless bursa approximately 10 cm in the long axis, with respect for the epididymo-testicular groove. The lymph node areas, in particular the inguinal area, were free. The Rest of the clinical examination was unremarkable. The testicular ultrasound found a heterogeneous testicular mass measuring 16 cm × 12 cm, the other testicle was without abnormality. The patient underwent a unilateral left inguinal orchiectomy. The post-operative consequences were simple.

The anatomopathological examination of the operative specimen was in favor of a vitelline tumor of 10 cm long axis associated with a mature teratoma infiltrating the epididymis and coming into contact with the albuginea, with the presence of vascular invasion, the slice of spermatic cord section was healthy. The tumor was classified as pT2 NxMx.

Postoperatively, our patient underwent a further extension assessment based on a thoraco-abdomino-pelvic scanner which revealed secondary pulmonary and hepatic lesions.

Tumor markers assayed postoperatively: alphafoetoprotein 8,937.39 ng/mL lacticodehydrogenase 1,369 IU/L, chorionic gonadotropic hormone beta less than 1.20 mIU/mL. One month after the operation, the patient developed a ponytail syndrome associated with stepping. An additional medullary MRI revealed a tumor proliferation next to the fifth lumbar vertebra with epidural extension responsible for spinal cord compression, in addition to bone metastases involving the axial skeleton, the iliac wings with epiduritis opposite the fifth dorsal vertebra (*Figures 1,2*).

In total, this is a 23-year-old patient followed for a mixed non-seminomatous germ cell tumor in the liver, lungs and secondary bone with epidural extension responsible for spinal cord compression, non-seminomatous germ cell tumor with a poor prognosis : high tumor markers + extra-pulmonary metastases.

Cryopreservation could not be performed due to the cauda equina syndrome responsible for impotence. A BEP-type polychemotherapy was started quickly: bleomycin-based protocol 30 mg, cisplatin 20 mg/m² and etoposide 100 mg/m² for 5 days + D8: bleomycin 30 mg + D15 bleomycin 30 mg. Decompressive radiotherapy was performed on the two metastatic sites at the dorsal and lumbar level. The patient received 20 GY divided into 4 GY per fraction spread over 5 days (*Figures 3,4*).

Currently, the patient is still under treatment, he received 3 courses of chemotherapy: 5 days of bleomycin, etoposide and cisplatin, on the eighth day a course of bleomycin, on the fifteenth day a course of bleomycin, with decompressive

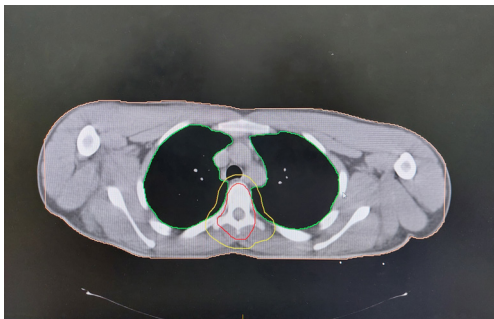


Figure 3 Radiotherapy contour in cross section covering the target volume of a dorsal metastasis.

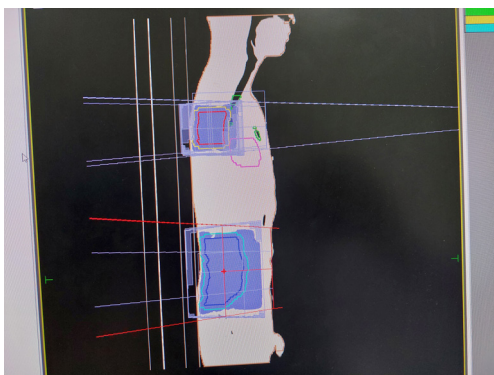


Figure 4 Illustration which shows the distribution of doses at the level of the target volumes on the sagittal plane in three-dimensional radiotherapy.

radiotherapy at the rate of 20 GY divided into 4 GY per fraction and spread over 5 days. The evolution was marked by a decrease in tumor markers: Alpha Foetoprotein: 403.32 ng/mL, Lacticodehydrogenase: 271 IU/L, Chorionic gonadotropic hormone Beta: less than 1.20 mIU/L, and a clear clinical improvement with disappearance of lower back pain and horsetail syndrome.

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Discussion

Testicular cancers represent 1% to 2% of cancers in men

and 3.5% of urogenital cancers (1). Whether they are seminomatous (TGS) or non-seminomatous (TGNS), they represent the first cancer in young men with an incidence in France estimated at around 7 new cases per 100,000 inhabitants per year (2). They mainly concern Caucasian men aged 15 to 45 years (3). Metastatic germ cell tumors are rare entities with a high cure rate due to their extreme chemosensitivity. Germ cell tumors of the testis account for 90% to 95% of testicular cancers. Among these germ cell tumors, three tumor types have a high metastatic capacity: seminoma, embryonic carcinoma and choriocarcinoma (4). In order of frequency, metastases of non seminomatous germ tumors are localized in the lungs, liver, system central nervous system and bone (5). Bone metastases are rare, occurring only in 5% of patients (4,6) and rarely isolated, often associated with lymph node and/or visceral metastases, patients with bone metastases are classified in the poor prognosis group with a low rate 45–55% long-term survival with standard-dose chemotherapy (6).

In adults, metastasis generally invades the epidural space from a vertebral site as in our case (7). Screening for bone metastases in germ cell tumors is generally not recommended, it should be noted that patients may not show symptoms and may be detected accidentally on imaging as was the case for a significant number of patients (11/19, 58%) in a retrospective study of 2,550 cases of germ cell tumors (8). To our knowledge, epidural metastasis of this tumor has not yet been reported in the English medical literature (7). Tumors of the breast, lung and prostate are the most known to metastasize in the epidural-spinal space (9,10). In a retrospective study of 969 patients with germ cell tumors, with predominantly non seminomatous histology (84%). The site of metastatic involvement has an impact on survival outcomes in patients with germ cell tumors, among patients with isolated extrapulmonary disease, those with brain metastases had the lowest survival (HR 3.24, 95% CI: 1.98–5.28, $P < 0.01$) followed by liver (HR 2.29, 95% CI: 1.56–3.35, $P < 0.01$) and bone (HR 1.97, 95% CI: 1.11–3.50, $P = 0.02$) (11). Improving the management of these patients remains a major challenge because the prognosis of these tumors remains unfavorable, with an overall 5-year survival of around only 50% in the 1990s when the IGCCCG classification was developed (12) and around 60% in more recent series (13). From 1987, 4 cycles of BEP were the standard treatment for 25 years, 4 cycles of VIP with G-CSF support being an alternative in case of contraindication to bleomycin (14). The standard treatment for metastatic germ cell tumors is multimodal (15),

it is based on polychemotherapy and surgery of the residual masses. Orchiectomy must be systematic, even in the case of metastatic disease. It is performed as soon as the diagnosis is made but should not delay systemic treatment in the event of significant visceral tumor involvement. First-line chemotherapy for most metastatic germ cell tumors relies on a combination of chemotherapy including cisplatin and etoposide, either alone (EP) or in combination with bleomycin (BEP) or ifosfamide (VIP). The pre-therapeutic assessment must include the performance of respiratory function tests (pulmonary toxicity of bleomycin). Due to the young age of these patients, an oncofertility consultation should systematically be offered before starting treatment, although this should not delay chemotherapy in the event of extensive disease. Brain metastases and bone metastases are particularly rare, and their treatment is not well codified. An international collaborative effort is underway to try to collect more cases to define the best possible treatment strategy.

We wish to emphasize that this tumor can metastasize to the spinal epidural space and that it should be included among the differential diagnoses of the causes of metastatic spinal cord compression.

In this article, we report a clinical case of spinal cord compression secondary to metastasis in the spinal epidural space from a testicular germ cell tumor in a 23-year-old patient. Through this, we want to emphasize that an epidural metastasis of this tumor is possible, and that it must be part of the diagnoses to be evoked in the face of metastatic spinal cord compression, which potentially allows to avoid a significant functional handicap and a profound alteration in the quality life of young patients.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at <https://atm.amegroups.com/article/view/10.21037/atm-2022-51/rc>

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at <https://atm.amegroups.com/article/view/10.21037/atm-2022-51/coif>). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all

aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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References

1. Tourne M, Radulescu C, Allory Y. Testicular germ cell tumors: Histopathological and molecular features. *Bull Cancer* 2019;106:328-41.
2. Binder-Foucard F, Bossard N, Delafosse P, et al. Cancer incidence and mortality in France over the 1980-2012 period: solid tumors. *Rev Epidemiol Sante Publique* 2014;62:95-108.
3. Lavaud P, Baciarello G, Fizazi K. Management of metastatic testicular germ cell tumors. *Bull Cancer* 2019;106:896-902.
4. Neuzillet Y, Méjean A, Leuret T. Rare locations of metastases from germ cell cancers. *Prog Urol* 2008;18 Suppl 7:S388-91.
5. Bredael JJ, Vugrin D, Whitmore WF Jr. Autopsy findings in 154 patients with germ cell tumors of the testis. *Cancer* 1982;50:548-51.
6. Oechsle K, Bokemeyer C, Kollmannsberger C, et al. Bone metastases in germ cell tumor patients. *J Cancer Res Clin Oncol* 2012;138:947-52.
7. Colak A, Benli K, Berker M, et al. Epidural metastasis of testicular yolk sac tumor: an unusual cause of spinal cord compression. Case report. *Pediatr Neurosurg* 1991-1992;17:139-41.
8. Jamal-Hanjani M, Karpathakis A, Kwan A, et al. Bone metastases in germ cell tumours: lessons learnt from a

- large retrospective study. *BJU Int* 2013;112:176-81.
9. Allen JC. Management of metastatic epidural disease in children. *J Pediatr* 1984;104:241-2.
 10. Wright RL. Malignant tumors in the spinal extradural space: results of surgical treatment. *Ann Surg* 1963;157:227-231.
 11. Patel HD, Singla N, Ghandour RA, et al. Site of extranodal metastasis impacts survival in patients with testicular germ cell tumors. *Cancer* 2019;125:3947-52.
 12. Binder-Foucard F, Bossard N, Delafosse P, et al. Cancer incidence and mortality in France over the 1980-2012 period: solid tumors. *Rev Epidemiol Sante Publique* 2014;62:95-108.
 13. Kier MG, Lauritsen J, Mortensen MS, et al. Prognostic Factors and Treatment Results After Bleomycin, Etoposide, and Cisplatin in Germ Cell Cancer: A Population-based Study. *Eur Urol* 2017;71:290-8.
 14. Hinton S, Catalano PJ, Einhorn LH, et al. Cisplatin, etoposide and either bleomycin or ifosfamide in the treatment of disseminated germ cell tumors: final analysis of an intergroup trial. *Cancer* 2003;97:1869-75.
 15. Uematsu M, Kanemasa Y, Nakamura S, et al. Multimodal Treatment of Extragonadal Choriocarcinoma with Multiple Brain and Lung Metastases: A Case Report. *Case Rep Oncol* 2019;12:928-34.

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