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Case Report
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Radio-Induced Sarcoma in a Long-Term Neuroblastoma Survivor

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Abstract

We report a particular case of a long-term survivor with a 37 years interval to radio-induced sarcoma; a 4 years old girl treated in Brussels in 1975 for vertebral neuroblastoma (L1 to L4) with R2 resection and adjuvant 2D radiation therapy delivering 40Gy with complete remission, presenting after 37 years with right flank pain revealing an in radiation-field locally infiltrating mass (Paravertebral D12 to L3), biopsy revealed a NOS sarcoma, managed with Palliative chemotherapy and decompressive 3D radiation therapy, till last follow up the patient is asymptomatic and still under chemotherapy.

Keywords: Radiation induced malignancy; Second malignancy; Therapy related malignancy; Radiotherapy; Neuroblastoma

Introduction

The continuous development in radiation therapy field aims to improve local control, minimize toxicity and avoid second malignancy, this important weapon; utilized either as definitive therapy, in pre-operative, post-operative, with concomitant chemotherapy or alone is a mainstay for local control in malignant tumors. Among radio-induced malignancies, sarcoma is a rare entity with poor prognosis as the 5 year overall survival ranges between 10 and 35%. The diagnosis of radio-induced sarcoma is based on 4 criteria: antecedent of radiotherapy, a minimum 3 years interval, sarcoma sub -type different from the primitive tumor and localization in the radiation field [1,2].

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Case Presentation

A 3 years old Congolian girl, seen in pediatric department in Belgium for management of thoracic scoliosis that was considered after long history of investigation in Congo as post-polio-like scoliosis, thoracic radiography showed an intra-rachidienne process, gaseous myelography revealed a L1 to L4 intra-rachidian extra-dural tumor deviating the spinal cord, with high VMA urinary levels, with unexpected normal neurologic exam. The young child underwent a first surgery: fragmented reductive resection after which pathological analysis set the diagnosis of neuroblastoma, fifteen days later a second surgery was performed consisting on L1 to L4 laminectomy with R2 resection as the tumor was encompassing the spinal cord, than benefited from a 2D adjuvant radiotherapy delivering 40 Gy in 19 daily fractions (5 days per week) with 2 beams (anterior and posterior) using a cobalt source, radiation field extends from D6 to L3 (Figure 1). In 2012, aged of 41 years (448 months from diagnosis of neuroblastoma) the young woman consults for isolated right flank pain and sensation of local heaviness, a computed tomography showed a heterogeneous process of right paravertebral space from D12 to L3 infiltrating muscular and osseous structures, a biopsy revealed a NOS sarcoma, the diagnosis of radio-induced sarcoma was made (Figure 2). As the tumor was considered unresectable, the patient started chemotherapy; a partial response was obtained after 6 cycles of Doxorubicin and ifosfamide, than received a 2nd line chemotherapy (Etoposide and Ifosfamid) after radiological progression, after which the tumor kept progressing after 3 cycles, the tumor volume kept progressing even after 3rd line (Pazopanib) and 4th line chemotherapy (Dacarbazine). It's worth mentioning that during the entire course of chemotherapy lines the patient was PS: 1 (sensation of heaviness in the flank) without neurological disorder, but did benefited from a decompressive radiotherapy as the magnetic imaging showed a D12 epiduritis, delivering 20 Gy in 5 daily fractions with 3D conformal technic using 18 Mev linear accelerator photon beams, with good tolerance. At the last follow up, the patient was reporting moderate fatigue and still under chemotherapy.

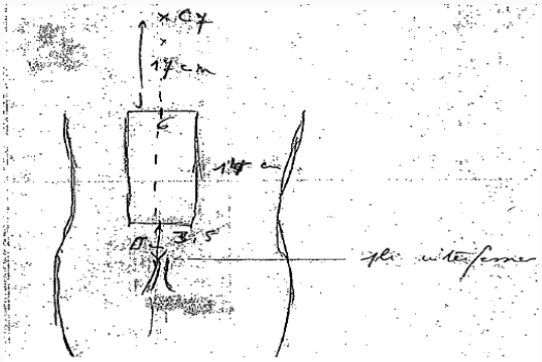

Figure 1: Drawing made the day of radiotherapy: radiation field extends from D6 to L3.

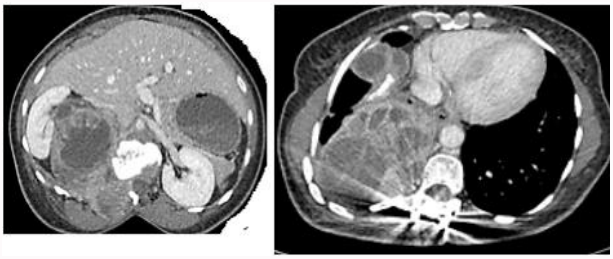

Figure 2: A voluminous heterogeneous locally infiltrative right paravertebral mass displacing abdominal organs.

Discussion

High risk neuroblastoma survivors are exposed to long term toxicities of the aggressive multimodal treatment, the childhood cancer survival study reported a 7% cumulative risk at 30 years. The risk of developing a radio-induced malignancy for high risk neuroblastoma patients is well known, in the report of CCG 3891 that included 379 children with high risk neuroblastoma authors reported 4 cases of second malignancy: an acute lymphoblastic leukemia, an acute myeloblastic leukemia, a clear cell carcinoma and a follicular thyroid carcinoma [3]. The therapy related malignancy risk is even higher in children receiving high dose alkylating agents, indeed; the study of Martin and et al. [3] including 87 children with high risk neuroblastoma managed with high dose induction chemotherapy followed by autologous steam cell rescue before local control, concluded to a significantly higher incidence of second malignancy specially hematological and recommended a long follow up for survivors. The experience of le groupe européen d'étude sur le neuroblastome trial including 69 survivors after 5 years (among 262 children with high risk neuroblastoma) reported 4 second malignancies (high grade fibroblastic osteosarcoma, carcinoma of parotid gland, rhabdomyosarcoma and anaplastic ependymoma), previous analysis established strong association between solid tumors and radiation therapy (including total body irradiation), while hematological malignancies are associated with alkylating agents [4-7]. The analysis of the experience of pediatric department of university of Chicago, including 2801 patients with neuroblastoma (among who 47% received radiotherapy) with median follow up of 74 months (67 to 455 months), reported 34 second malignancies with median latency of 139 months (10 to 432 months) [8]. In our observation we recorded one of the longer latencies in literature (37 years and 4 months), with a rare second malignancy subtype (NOS sarcoma), it's reaffirming the importance of prolonged follow up for these patients.

Conclusion

All the children treated with radiation therapy will be at risk of developing a second malignancy necessitating a prolonged risk adapted follow up, the emergent radiotherapy technics should, in addition to improving local control and minimizing side effects, avoid occurrence of second malignancy.

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