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original article

Caecal leiomyoma detected by whole-body MRI in a patient with Hodgkin lymphoma: first case report

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SUMMARY: **Caecal leiomyoma detected by whole-body MRI in a patient with Hodgkin lymphoma: first case report.**

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Introduction. *Hodgkin Lymphoma (HL) is one of the most curable malignant diseases. Modern treatments, like the combined radiochemotherapy and stem cell transplantation, have increased the number of malignant disease survivors. However, HL survivors are at risk of long-term effects, including the development of solid tumors. Secondary neoplasms are a major cause of late morbidity and mortality following treatment for HL.*

Case report. *We report the case of a male patient, treated for HL by chemotherapy, who developed a large leiomyoma of the cecum one year after the treatment. A whole-body Magnetic Resonance (WB-MRI) scan performed during the follow-up allowed the detection of this incidental caecal mass that was absent in a Computed Tomography (CT) scan performed immediately after the treatment. After a CT-guided biopsy, the lesion was surgically removed and the diagnosis of caecal leiomyoma was obtained.*

Discussion. *To our knowledge, this is the first case report, according to the scientific literature, of caecal leiomyoma developing after chemotherapy in a HL survivor. Leiomyoma is a rare benign tumor that usually appears as a solitary small mass with a nodular growth and a benign course.*

Conclusion. *This case shows that WB-MRI allows detecting relevant incidental findings during the oncologic follow-up, avoiding both radiation exposure and contrast agent administration. Furthermore, leiomyoma should be considered in the differential diagnosis between the caecal masses with high growth rate.*

KEY WORDS: Hodgkin lymphoma - Magnetic Resonance Imaging - Caecal leiomyoma - Laparoscopic surgery.

Introduction

Hodgkin Lymphoma (HL), which predominantly occurs in young and middle-aged individuals, is one of the most curable malignant diseases in adults. Modern treatments, including immunochemotherapy integrated with targeted radiotherapy techniques, have increased the number of malignant disease survivors leading to a significant improvement in the HL five-year survival rate over the past few decades (1, 2). However, HL survivors are at risk of long-term effects, including the development of solid tumors, also because of chemotherapy and radiotherapy (3, 4). Secondary neoplasms are a major cause of late morbidity and mortality following treatment for HL (5). Thus, late complications of cancer therapy, including secondary neoplasms development, are now an important area of concern (6). Leiomyoma is a benign soft tissue tumor that arises from smooth muscle cells; several cases have been reported in literature, but lower gastrointestinal tract location, and especially in the caecum, is very rare (7-10). We report the case of a 44-years-old man, affected by HL, successfully treated by chemotherapy, who developed a large and quickly growing leiomyoma of the caecum that was absent in post-treatment work-up and was detected by a whole-body Magnetic Resonance (WB-MRI) scan performed one year after treatment. The Institutional review board of our hospital approved our work and the patient provided written informed consent.

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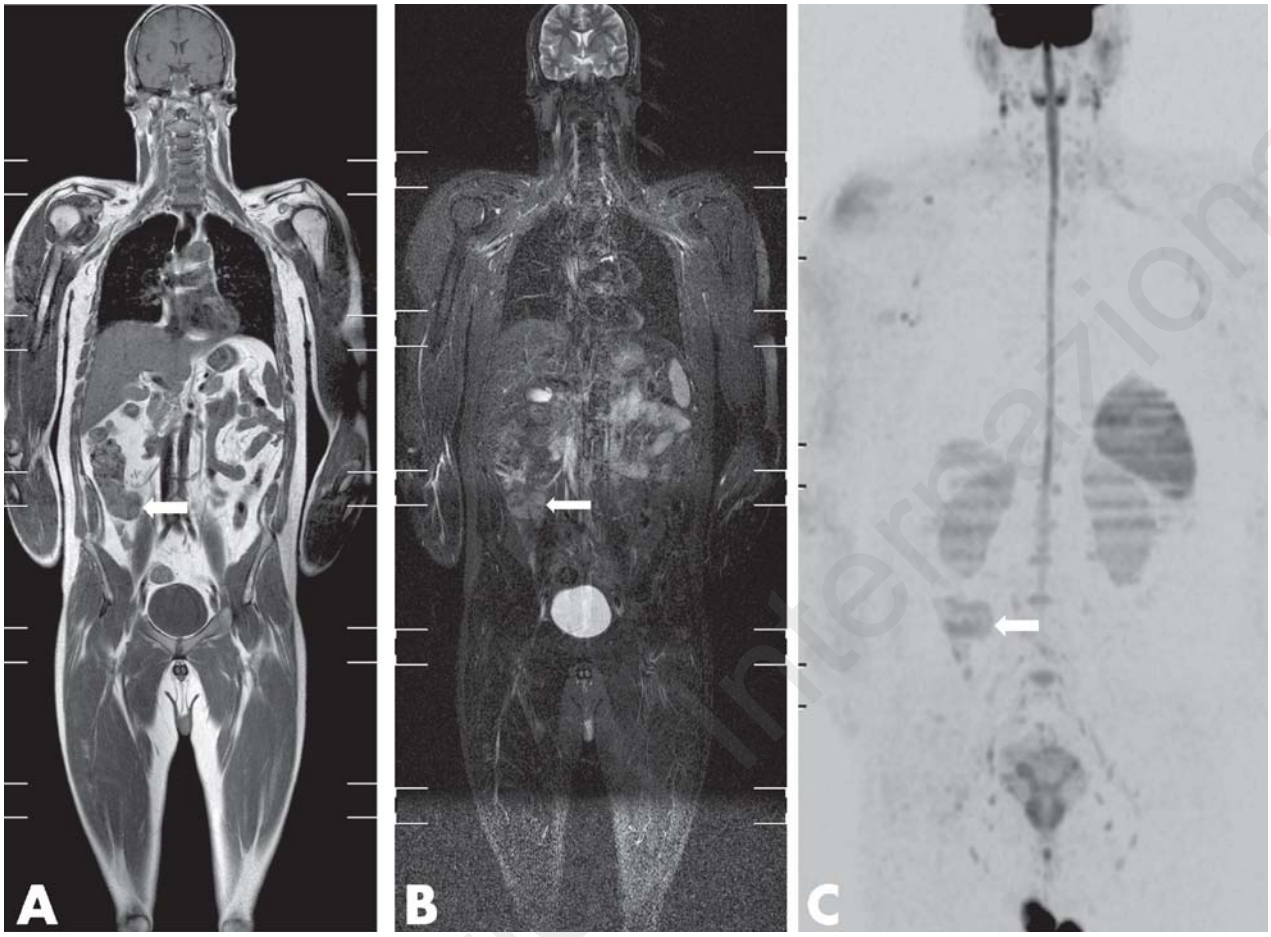

Figure 1 - Caecal mass on WB-MRI.
Coronal T1-weighted (A), coronal T2-weighted STIR (B) and MIP grey-scale inverted DWI (C) WB-MRI images show a homogeneous solid, exophytic, with low-intermediate signal intensity in T1- and T2-weighted STIR images, without high signal intensity on DWI (white arrow).

Case report

A 44-years-old male patient, without previous comorbidities, on October 2012 developed fever and back pain. A contrast enhanced CT scan was performed showing a left lung hilar mass that was biopsied, obtaining the diagnosis of nodular sclerosis classical HL. Systemic workup revealed diffuse nodal and extra-nodal locations of disease with multiple bone lesions. He received chemotherapy with Adriamycin-Bleomicyn-Vinblastine-Dacarbazine (ABVD) regimen. The response to chemotherapy was optimal and he completed 6 courses of chemotherapy. Post-treatment work-up, including CT, showed evidence of complete remission of disease; therefore, the patient was on follow-up through WB-MRI scans. One year after the end of treatment, a WB-MRI was performed, in order to monitor the patient and to avoid further radiation exposure. WB-MRI did not show any suspicions of relapse of lymphoma, however it revealed an incidental mass of the caecum (Figure 1), which was absent in the CT scan performed one year before. The patient underwent a contrast enhanced CT examination that confirmed the presence of an exophytic caecal mass, suspicious for a lesion developing from submucosa (Figure 2). We performed a CT-guided percutaneous biopsy of the mass and a diagnosis of leiomyoma was obtained. Based on our experience we performed a 3D laparoscopic right hemicolectomy (11-14) with intra-corporeal anastomosis: the patient was placed on the operating table in the Trendelenburg position and we used Veress needle in left subcostal region (15-17), an optical trocar in left peri-umbilical region and other three trocars in left upper and lower quadrant (5 mm) and in sovra-pubic region (12 mm) for endo-stapler. First we identified the ileum-colic vessels and then continued with the preparation of the last ileal loop and the colo-epiploic detachment. We carried a side to side intracorporeal anastomosis with endo-stapler and a continuous riassorbable suture. Operative time was 145 min and blood loss was no significant. Patient began food intake

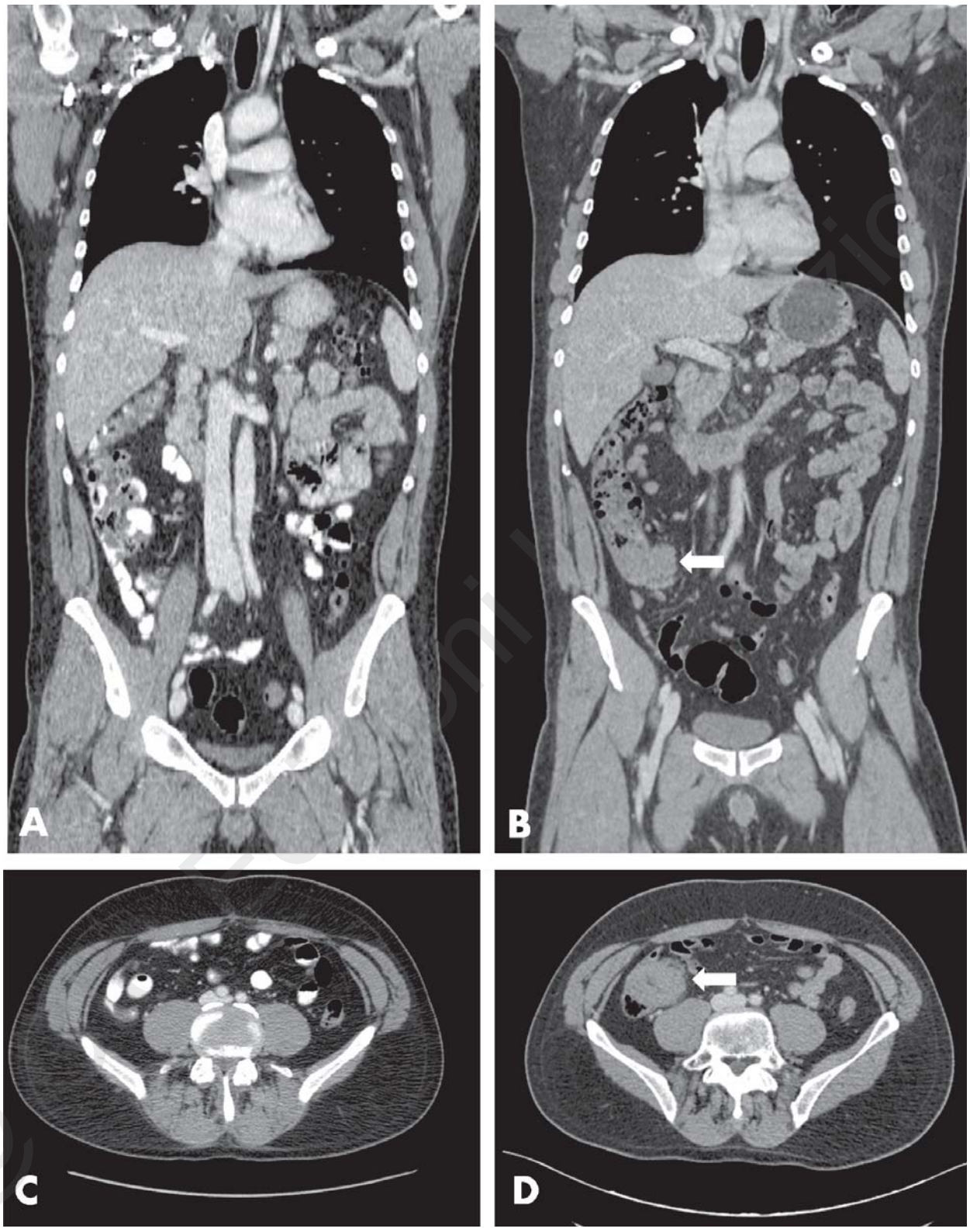

Figure 2 - Caecal mass on contrast enhanced CT.
Contrast enhanced CT performed after the WB-MRI scan (B, D) confirmed the presence of the caecal mass (white arrow) which was absent in the CT scan performed one year before (A, C). Contrast enhanced CT demonstrated a well-defined mass with slight and homogeneous enhancement after administration of intravenous contrast agent; the mucosal layer covering the mass was intact and the fat plane surrounding the lesion was preserved. All these characteristics suggested that the mass was exophytic and arisen from the submucosal layer of the caecum.

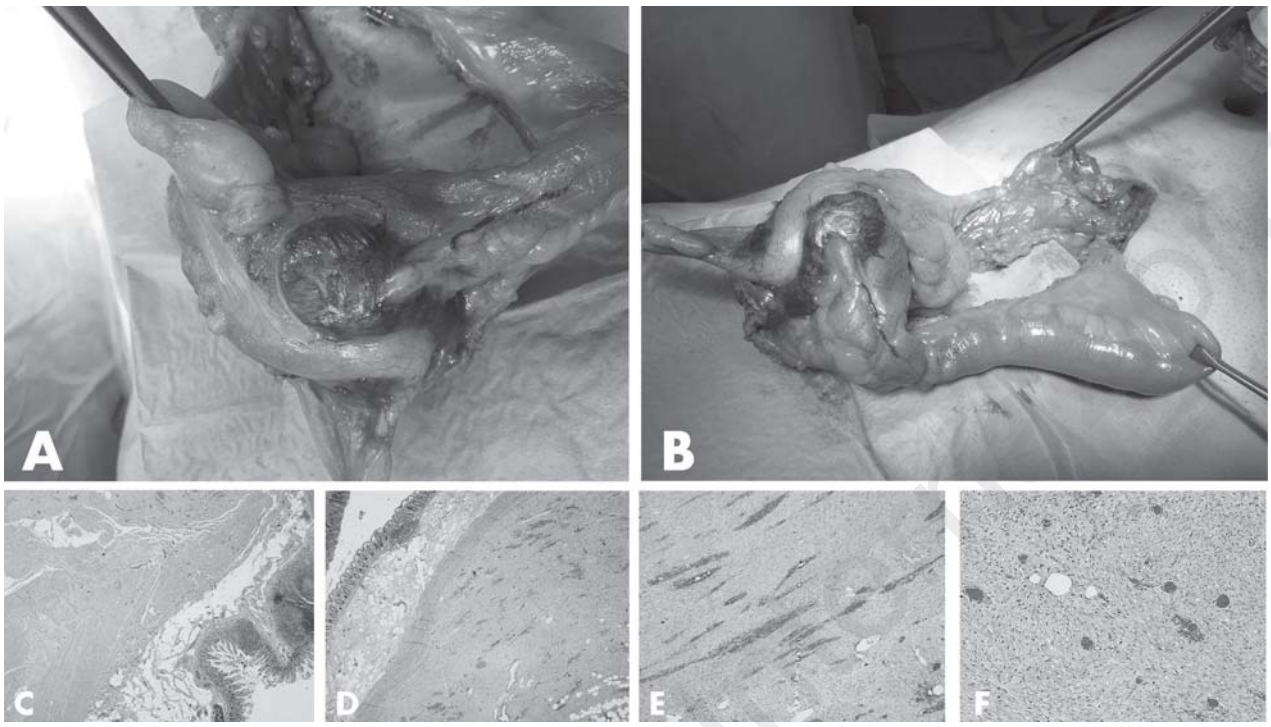

Figure 3 - Surgical specimen and histologic examination.
Right hemicolectomy with a sharply circumscribed and round mass of the caecum (A, B). The microscopic examination with Hematoxylin-Eosin Stain allowed to achieve the diagnosis of leiomyoma (C, D, E, F). 2x - overview of the tumor and its relations with the intestinal mucosa (C, D). 4x - detail of the population of the lesion with bundles of spindle-shaped cells and congested vessels (E). 10x - detail of the neoplastic population, without mitotic figures or necrotic areas, important findings for a differential diagnosis with the sarcomatous counterparts (F).

in first postoperative day with regular postoperative course. The entire lesion was removed and the histologic examination and immunohistochemical features confirmed the diagnosis of leiomyoma (Figure 3).

Discussion

According to the Surveillance, Epidemiology and End Results Program, secondary malignancies account for 16% of all cancers (6, 18), and they represent the leading cause of mortality in HL survivors (19). Several studies, recently reviewed by Lisik-Habib and coworkers (6), assessed or quantified the risk of developing secondary cancers following the successful treatment of HL. However they failed in producing consistent results, possibly due to differences in the number of patients included in the analysis, the characteristics of the study population, the length of the follow-up period, the chemotherapy schedule, the age of HL diagnosis and the dose and volume of irradiation (6). To our knowledge, this is the first case report, according to the scientific literature, of caecal leiomyoma developing after chemotherapy in a HL survivor. Leiomyoma is a rare benign tumor that usually appears as a solitary small mass with a nodular growth and a benign course; the majority of submucosal tumors, term clinically used for protuberant lesions or bumps covered with intact mucosa, are asymptomatic and incidentally diagnosed by endoscopic and radiographic examinations (20). As suggested by Japanese GIST guidelines (20), in a study regarding the endoscopic detection of submucosal lesions, it is necessary to do further evaluation with endoscopic ultrasonography, computed tomography with contrast enhancement or fine needle aspiration, when neoplastic submucosal tumors are 2-5 cm in diameter or when tumors <2 cm have clinically malignant features on endoscopy; in other cases the lesion can be followed up by endoscopy once or twice a year. In the case here reported, an endoscopic ultrasonography was not performed, due to the caecal localization of the neoplasm. The novelty of this case report is also that a rare caecal tumor was detected through unenhanced WB-MRI, which is a well-established technique for lymphoma staging (21-23) and response assessment to treatments (24, 25) and that was applied in the follow-up of our patient. WB-MRI is an imaging procedure that enables a whole body examination with high potential for application in oncologic patients using diffusion-weighted imaging (26-29). It also avoids radiation exposure in patients with high cancer risk like those with lymphoma and enables the detection of relevant incidental findings during the oncologic follow-up (30).

Conclusion

In conclusion, in this case report, a patient with HL treated by chemotherapy developed, in just a year, an incredibly quickly growing leiomyoma of the caecum, a rare benign gastrointestinal lesion, that was detected by a WB-MRI scan performed during a routine follow-up of this patient. Leiomyoma should be considered in the differential diagnosis between the caecal masses with high growth rate and between the secondary neoplasms developing in HL survivors.

Authors' contributions

All authors have made substantial contributions to the conception and design of the study; they have been involved in drafting the article and revising it critically for important intellectual content. Moreover, they have approved the final version to be published.

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Institutional review board statement

The study was reviewed and approved by the "Policlinico Paolo Giaccone Hospital" Institutional Review Board.

Informed consent statement

The patient provided informed written consent.

Conflict-of-interest statement

The authors state they have no conflict of interests.

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