Radiology Case Reports 15 (2020) 156–160

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journal homepage: www.elsevier.com/locate/radcr

Case Report

Benign histiocyte-rich pseudotumor in post treatment mediastinal Hodgkin's lymphoma

Yash Dilip Nilak, MBBS, DA, MRCP[a,*], *Anjumara Nilak, MBBS, DMRE, DNB(Rad), MMED, FRCR*[b], *Salim Shafeek, MD FRCP FRCP(Ed) FRCPath(Haem)*[c,d], *Dr. Zbigniew Rudzki*[e,f], *Joanne Chapman, BM, FRCPath, PGCE*[e]

[a] *Department of Medicine, Worcester Royal Hospital, Worcester WR5 1DD, UK*
[b] *Department of Radiology, Worcester Royal Hospital Worcester, WR5 1DD, UK*
[c] *Worcestershire Acute Hospitals NHS trust, Worcester, WR5 1DD, UK*
[d] *Hon Sen Lecturer in Clinical Haematology, University of Birmingham, Birmingham, UK*
[e] *Heartlands Hospital, Birmingham, B9 5SS, UK*
[f] *Institute of Cancer and Genomic Sciences, University of Birmingham, Birmingham, UK*

ARTICLE INFO

Article history:
Received 12 August 2019
Revised 11 October 2019
Accepted 14 October 2019

Keywords:
Nodular sclerosing Hodgkin's lymphoma
Tumor necrosis
Histiocyte-rich pseudotumor
PET

ABSTRACT

Benign xanthomatous pseudotumors are rare, mass forming lesions composed of lipid laden histiocytes and tumor necrosis following chemotherapy.

We present a rare case of young 36 year old male with primary mediastinal Hodgkin's lymphoma who developed xanthomatous pseudotumor mimicking relapse at the site of original disease on positron emission tomography. This scenario places emphasis on histologic confirmation of suspected treatment failure or relapse.

Introduction

Xanthomatous pseudotumors have been described in literature, with differing nomenclature in multiple anatomical locations including gastrointestinal tract, spleen, liver, breast, and mediastinum. The findings of this case should prompt the radiologist to remember and raise differential of benign pseudotumors in post chemotherapy ^{18}F-fluorodeoxyglucose (18FDG)-positron emission tomography (PET)/CT studies and stress importance of obtaining histopathological confirmation.

Case report

A 36 year old male presented with 6 month history of shortness of breath on exertion, cough, and weight loss.

* Corresponding author.
E-mail addresses: yash.nilak@nhs.net (Y.D. Nilak), anjumara.nilak@nhs.net (A. Nilak), salimshafeek@nhs.net (S. Shafeek), zbigniew.rudzki@heartofengland.nhs.uk (Dr.Z. Rudzki), Joanne.chapman@heartofengland.nhs.uk (J. Chapman).
https://doi.org/10.1016/j.radcr.2019.10.018

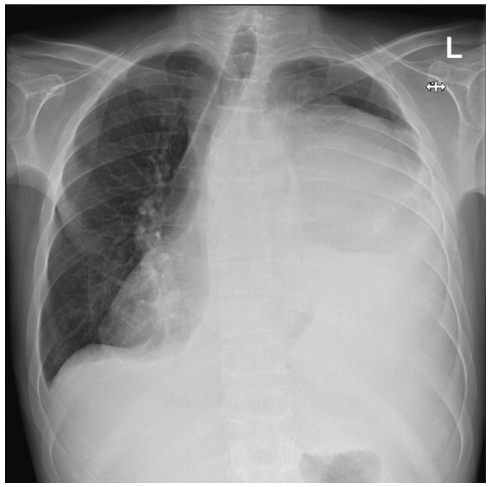

Fig. 1 – X-ray chest reveals near complete opacification of left lung field with cardiomediastinal shift to the right

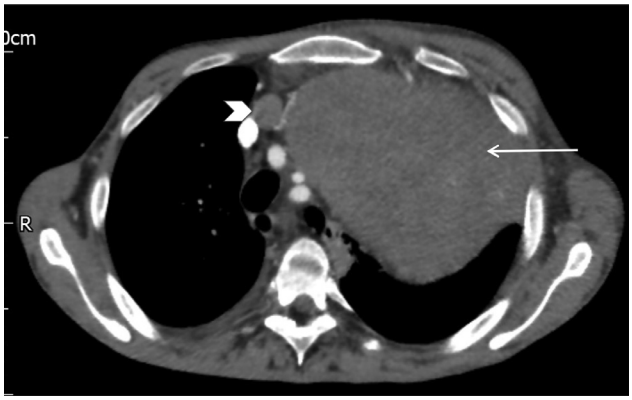

Fig. 2 – An isolated retrosternal node (white chevron) seen to the right of the mass, abutting the medial aspect of superior vena cava

On examination, he was thin and cachectic with positive clubbing and mild palpable splenomegaly. No other significant clinical or positive family history. Chest X-ray (Fig. 1) revealed complete opacification of left lung with cardiomediastinal shift to the right. CT scan of chest, abdomen, and pelvis with contrast (Fig. 2) demonstrated large, hypodense, mildly enhancing mass within the left thoracic cavity with few calcifications and internal vasculature measuring 23 cms in widest dimension causing significant shift of cardiomediastinal structures to the right. No CT features to suggest pulmonary origin of the mass lesion. Associated moderate-severe bilateral pleural effusions with partial atelectasis of lower lobes and moderate sized pericardial effusion noted (Fig. 3). A second retrosternal soft tissue mass/lymph node (Fig. 2, white chevron) seen superior to the vena cava. Mild splenomegaly of 13 cms but no significant infradiaphragmatic nodal disease. CT guided biopsy

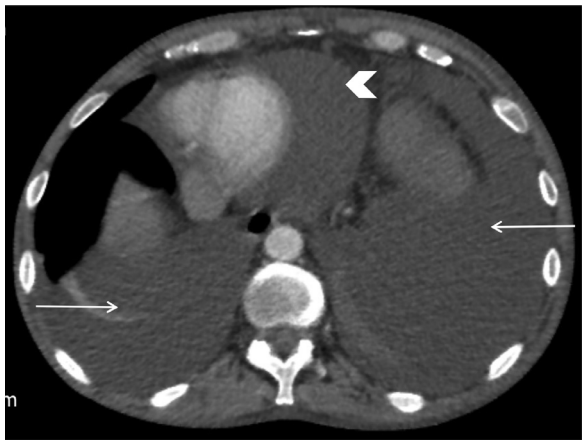

Fig. 3 – Associated moderate to severe bilateral pleural (white arrows) and moderate sized pericardial effusions (white chevron) with heart displaced to the left

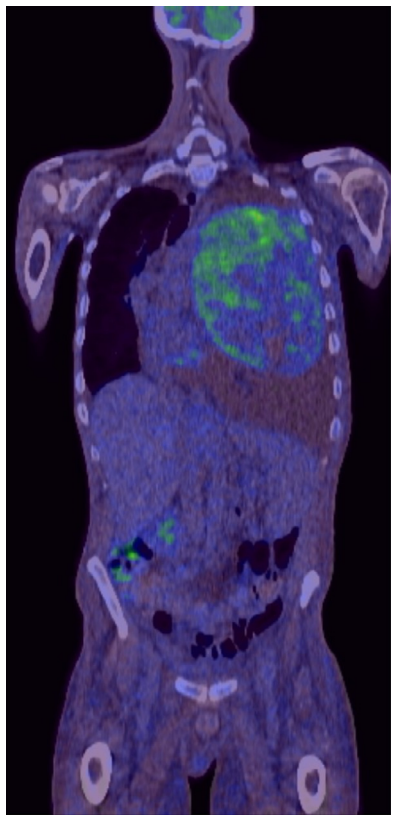

Fig. 4 – Initial ^{18}FDG PET staging scan (4 weeks after CT study) shows variable uptake within the mediastinal mass consistent with bulky stage 1 disease

a week later—confirmed classic nodular sclerosing Hodgkin's lymphoma.

The initial 2-deoxy-2-18Ffluoro-D-glucose (18FDG) PET scan (Fig. 4) revealed variable uptake within the mediastinal mass with an Standardized Uptake Value (SUV) max of 7.9 and SUV max of 7.5 in adjacent 2 cms superior

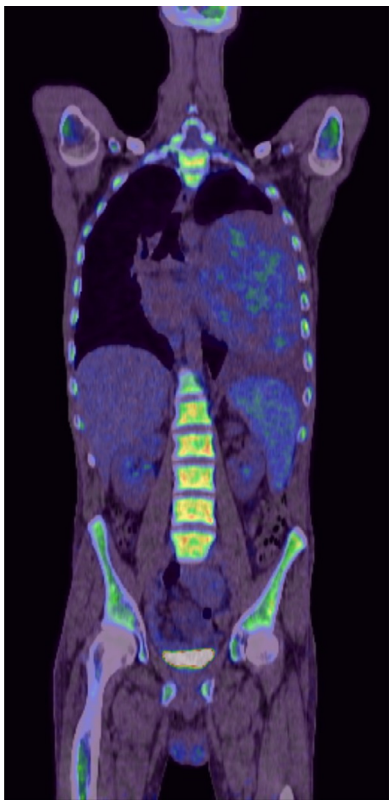

Fig. 5 – PET study post 2 cycles of chemotherapy showed reduction in size and SUV of the mediastinal mass consistent with partial response

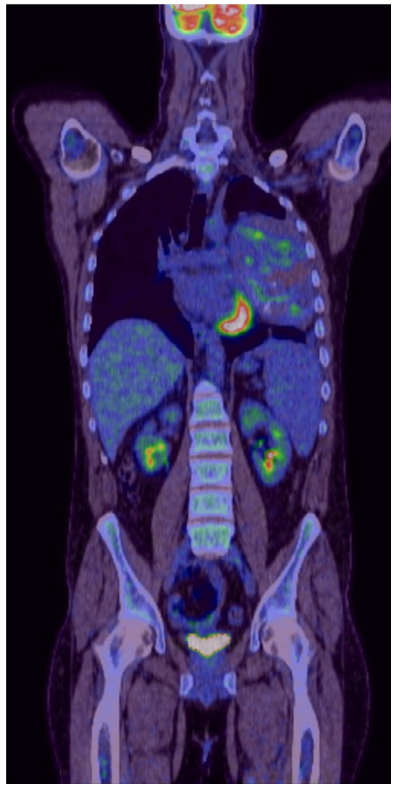

Fig. 6 – PET post 4 cycles of escalated, intensive chemotherapy shows mild further reduction in dimensions of mediastinal mass. However there is increase in SUV from previous PET, suggesting Deauville 4 response

mediastinal node. Moderate sized pleural and pericardial effusions showed no uptake and no further sites of FDG avid disease identified. PET was consistent with bulky stage I disease. Patient was started on Adriamycin, Bleomycin, Vinblastine, and Dacarbazine chemotherapy regime.

Second PET (Fig. 5) to assess initial response after 2 cycles of Adriamycin, Bleomycin, Vinblastine, and Dacarbazine demonstrated fractional reduction in size of the intrathoracic mass and increase in internal punctate calcifications. The FDG avidity reduced from SUV max 7.9 to 4.7 consistent with partial metabolic response.

Associated diffuse intense increased bone marrow and mild increased splenic uptake was ascribed to recent chemotherapy. Patient was offered escalated, intensive BEACOPP (Bleomycin + Etoposide + Cyclophosphamide + Doxorubicin + Vincristine) 4 cycles, due to poor response on interim PET-CT. Further PET post 4 cycles (Fig. 6), revealed mild further reduction in dimensions of mediastinal mass, now measuring 16 cms in maximum diameter. There was however heterogenous activity within the residual mass with SUV of 5.5 (previously 4.7), greater than background liveractivity, suggesting Deauville 4 response. On basis of PET findings, patient was offered salvage schedule with Gemcitabine + Vinorelbine + Bendamustine combination. Patient was consented for autograft. Prior to autograft, repeat 18FDG-PET in 3 month interval showed minimal increase in

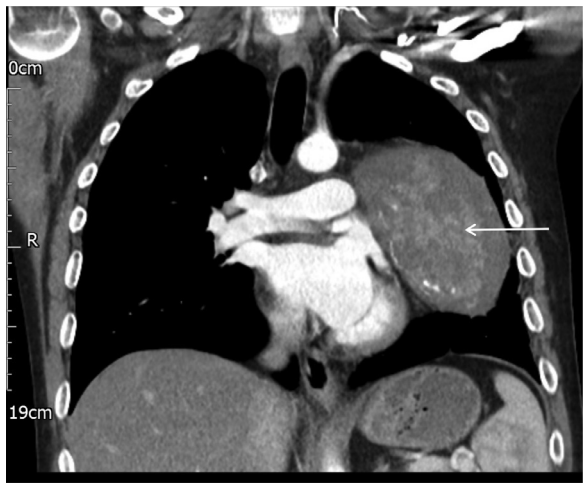

Fig. 7 – Enhanced computed tomography prior to excisional biopsy demonstrates residual mass with internal calcifications (white arrow)

heterogeneous FDG uptake within the residual mediastinal mass with SUV max of 6.6 (previously 5.5) consistent with Deauville stage 4. In the light of these findings, autograft was withheld till definitive tissue diagnosis was obtained. Multidisciplinary team decided on repeat biopsy which

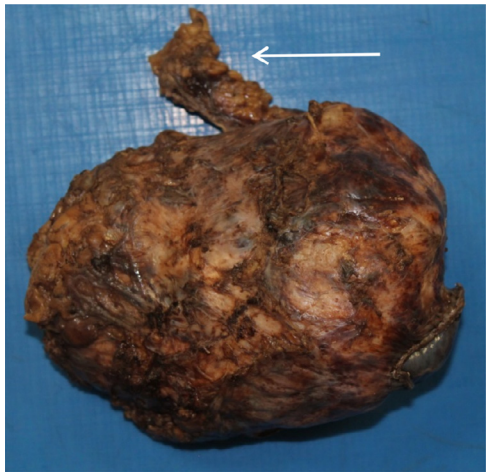

Fig. 8 – Gross excisional biopsy specimen with adherent lung wedge (white arrow)

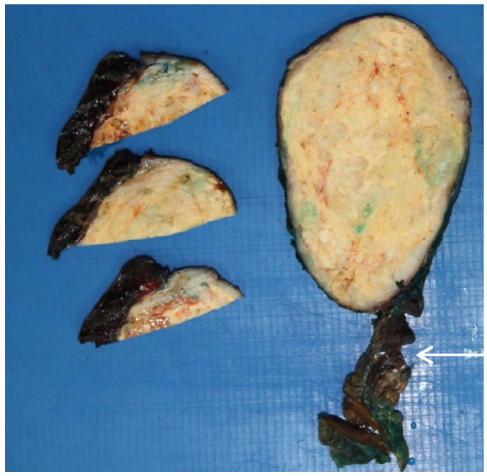

Fig. 9 – Gross excisional biopsy specimen with adherent lung wedge (white arrow)

revealed necrotic material but no active tumoral cells. Local radiotherapy was contemplated for the residual bulky mediastinal mass however due to large field and associated cardiac-pulmonary toxicities, it was declined. In patient's best interests and to confirm/exclude residual disease, which would require more aggressive therapy, left thoracotomy and complete excision of the whole mass was performed. CT chest with contrast (Fig. 7) prior to surgery revealed stable dimensions of the mediastinal mass as compared to interval CT study 4 months prior with scattered calcifications but no signs of chest wall invasion. The gross excisional specimen (Fig. 8 and 9) was 14.7 cms and weighed 593 grams. Morphologic and immunophenotypic analysis (Fig. 10) showed encapsulated necrotic mass devoid of any viable neoplastic cells, with sheets of foamy histiocytes focally within the mass. Residual thymic tissue also noted within resected pericardial fat. Final 18FDG-PET (Fig. 11), 5 months post surgery was compatible with complete metabolic response.

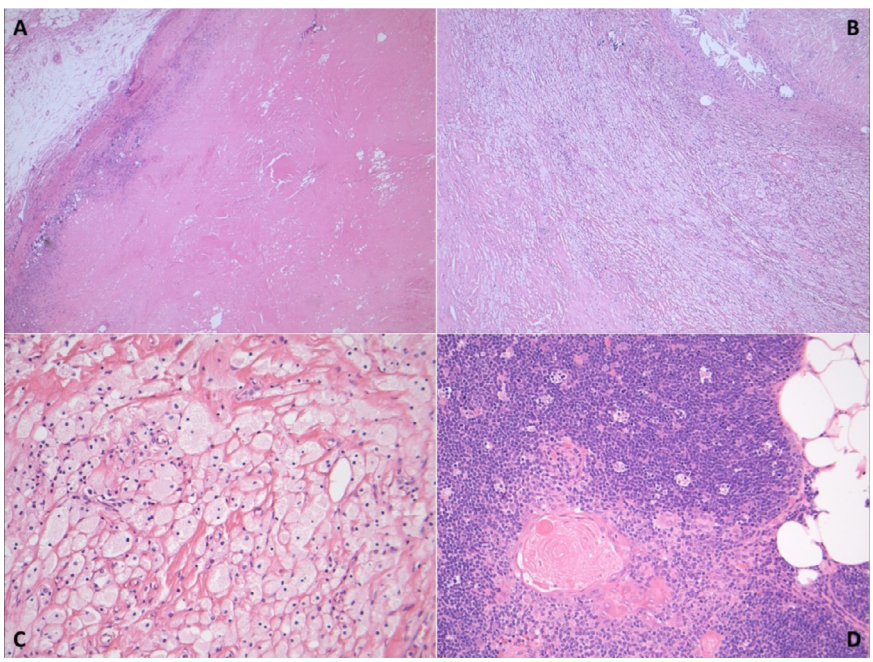

Fig. 10 – Haematoxylin and eosin stained slides. A. encapsulated necrotic mass devoid of any viable neoplastic cells (magnification—2×). B. sheets of foamy histiocytes focally seen within the mass (magnification—5×). C. details of foamy histiocytes (magnification—40×). D. normal residual thymic tissue in the adjacent pericardial fat (magnification—20×)

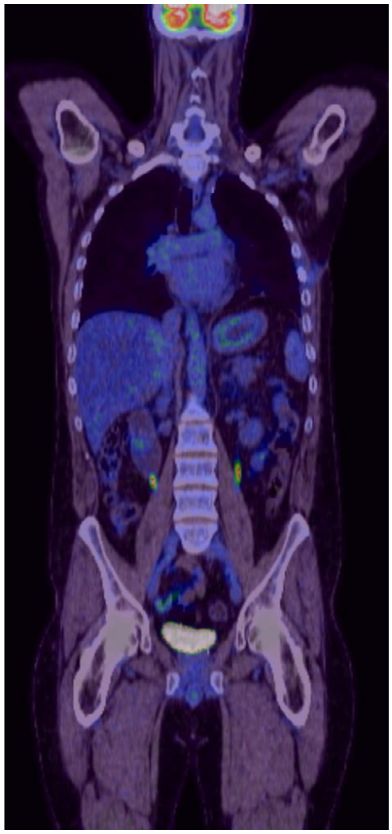

Fig. 11 – Final FDG-PET, 5 months postsurgery confirms complete metabolic response

Discussion

In patients with Hodgkin's and non-Hodgkin lymphoma, residual masses at the site of original disease after end of treatment are not uncommon [1,2] . Although cross-sectional studies, CT, and MRI are sensitive in evaluating residual masses PET is superior in distinguishing viable tumor from necrosis [3,4].

Previous literature has shown post chemotherapy mass lesions in breast [5], intestine [6], mediastinum [7], and spleen [8].

Histopathological response to chemotherapeutic agents is manifested by several changes including induction of necrosis. Post chemotherapy, first biopsy from the residual mediastinal mass in our case had revealed fibrosis and necrosis. As a response to tumor necrosis and release of chemotactic substances, circulating monocytes are recruited to the site and differentiate into histiocytes. Cascade of activation continues resulting in excessive histiocyte accumulation in response to tumor necrosis. This can explain increased uptake of FDG in our case. PET positivity at site of disease in first few months after chemo/radiotherapy completion is known, hence the imaging subcommittee of the International Harmonization Project recommended performing PET at least 3 weeks following chemotherapy and preferably 8-12 weeks after radiation therapy. [9] 18FDG-PET-CT has become the standard of care for staging patients with lymphomas. We need to have a cautious approach in patients showing radiological improvement in 1 site and new/residual disease at another site. In our case, the original and only site of disease was massive with reduction in SUV initially and then increased SUV-max, although patient was clinically completely asymptomatic. The results confounded the team. The pivotal decision to go ahead with biopsy in cases with residual masses is one of the difficult conundrums faced by clinical experts. This holds true in context of masses which are surgically not accessible/pose high complication threat as in our case wherein the mediastinal mass was closely opposed to the lung and pericardium. It is crucial never to start therapy for suspected relapse or disease progression without obtaining tissue diagnosis, as highlighted by our case. Limitations of PET-CT and postchemotherapy histiocyte-rich pseudotumor should be considered while planning follow up in patients with Hodgkin's lymphoma.

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