

Available online at www.sciencedirect.com

ScienceDirect

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



Case Report

Steroid responsive inflammatory myofibroblastic tumor of the lung evaluated by FDG PET/CT imaging[☆]

James Yuheng Jiang, MD, MMed^{a,*}, Monica Comsa, MD, FRACP^{b,c}, Veronica Chi Ken Wong, MBBS, FRACP, FAANMS^{a,c}, Robert Mansberg, MBBS, FRACP, FAANMS^{a,c}

^a Departments of Nuclear Medicine and PET, Nepean Hospital, Kingswood, Derby St, NSW 2747, Australia

^b Department of Respiratory Medicine, Nepean Hospital, Kingswood, NSW, Australia

^c Faculty of Medicine and Health, University of Sydney, NSW, Australia

ARTICLE INFO

Article history: Received 9 November 2021 Accepted 21 November 2021

Keywords: PET FDG inflammatory myofibroblastic tumor inflammatory pseudotumor

ABSTRACT

A 68-year-old gentleman was referred for ¹⁸F-FDG PET/CT for a pulmonary mass in the left upper lobe which demonstrated intensely FDG-avid confluent pulmonary consolidation in the left upper lobe (SUV_{max} 15.1). Histopathologic biopsy of the left upper lobe lung mass was consistent with inflammatory myofibroblastic tumor (IMT). The patient was started on steroid treatment in conjunction with antibiotics. Follow-up FDG PET/CT 3 weeks after commence of treatment showed remarkable response of the IMTs to therapy with much less avid FDG uptake (SUV_{max} 5.4) and marked improvement in the pulmonary consolidation. Nevertheless, the patient underwent left upper lobe lobectomy due to evidence of persistent cystic disease and malignant potential associated with IMTs. Final histopathology was consistent with IMT with no evidence of malignancy.

© 2021 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license

(http://creativecommons.org/licenses/by-nc-nd/4.0/)

Background

Inflammatory myofibroblastic tumor (IMT) is a subgroup of the broad category of "inflammatory pseudotumors" and is composed of myofibroblastic mesenchymal spindle cells accompanied by an inflammatory infiltrate of plasma cells, lymphocytes, and eosinophils [1]. IMTs have been described as benign neoplasms with intermediate malignant potential as they sometimes behave clinically as malignant tumors with local recurrences or distant metastases [1, 2]. The lungs, liver and gastrointestinal tract are the most common sites for IMT [3]. Differentiation of malignant cancer and acute infection from IMT maybe challenging with ¹⁸F-FDG PET/CT [4]. We present a case of a patient with steroid responsive IMT of the lung evaluated by ¹⁸F-FDG PET/CT imaging.

^{*} Competing Interest: The authors have declared that no competing interests exist.

Corresponding author.
E-mail address: jjia8233@uni.sydney.edu.au (J.Y. Jiang).

https://doi.org/10.1016/j.radcr.2021.11.049

^{1930-0433/© 2021} The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)



Fig. 1 – (A) Maximal intensity projection (MIP), (B) axial ¹⁸F-FDG PET and (C) axial CT of a 68-year-old-gentleman referred for a pulmonary mass in the left upper lobe demonstrates intensely avid confluent pulmonary consolidation in the left upper lobe (SUV_{max} 15.1) with mild to moderate uptake in left lower paratracheal lymph node (SUV_{max} 3.6), and in aortopulmonary window lymph nodes.

Case presentation

A 68-year-old gentleman with a background history of prostate cancer on surveillance presented with several months of persistent cough associated with fatigue, night sweats and weight loss of about 10 kg over 6 months. He denied any significant smoking history or occupational exposure to carcinogens including asbestos or other inhaled carcinogens. Physical examination of the chest was unremarkable. A CT scan of the chest showed airspace opacities with some mass-like lesions in the left upper lobe associated with left hilar lymphanopathy. An ¹⁸F-FDG PET was arranged for further evaluation.

PET/CT demonstrated intensely FDG avid confluent pulmonary consolidation in the left upper lobe (SUV_{max} 15.1) with mild to moderate uptake in left lower paratracheal lymph node (SUV_{max} 3.6), and in aortopulmonary window lymph nodes (SUV_{max} 3.8) (Fig. 1). No FDG avid disease was identified elsewhere. Histopathological analysis of the lesion showed broad bands of eosinophilic collagen traversed by small blood vessels containing predominantly lymphoplasmacytic inflammatory infiltrates with scattered polymorphs. The features were consistent with a fibro-inflammatory process with moderate background inflammatory changes and in keeping with an IMT. Immunohistochemical analysis was negative for anaplastic lymphoma kinase (ALK). Following discussion at multidisciplinary meeting, he was started on a course of oral steroid treatment with antibiotics (Trimethoprim and/or Sulfamethoxazole).

His symptoms including cough resolved whilst on steroid therapy. A repeat PET/CT and diagnostic CT 3 months following initial presentation and 3 weeks after initiation of steroid therapy demonstrated significant metabolic response (SUV_{max} up to 5.4) and near complete resolution of consolidative changes with residual cystic changes and bronchiectasis in the left upper lobe (Fig. 2). Previously FDG-avid lymph nodes were no longer evident. No new sites of FDG avid disease was demonstrated.

CT chest 4 months following initial presentation and 7 weeks after initiation of steroid therapy later shows complete resolution of consolidation and further improvement in the irregular opacities in the left upper lobe (Fig. 3). There is however, residual left upper lobe fibrosis, bronchiectasis and cystic changes. The patient underwent left upper lobe lobectomy due to evidence of persistent cystic disease and malignant potential of IMT. Final histopathology was consistent with IMT with no evidence of malignancy.



Fig. 2 – (A) MIP, (B) axial ¹⁸F-FDG PET, and (C) axial CT 3 weeks after initiation of steroid and antibiotics demonstrates significant metabolic response (SUV_{max} 5.1) and near complete resolution of consolidative changes with residual cystic changes and bronchiectasis in the left upper lobe. Previously FDG-avid lymph nodes are no longer evident. No new sites of FDG avid disease was demonstrated.



Fig. 3 – Axial CT chest 4 weeks later shows complete resolution of consolidation and further improvement in the irregular opacities in the left upper lobe. There is however, residual left upper lobe fibrosis, bronchiectasis and cystic changes.

Discussion

IMT is a subgroup of a broad category of "inflammatory pseudotumors" and has emerged as a distinct entity with characteristic clinical and pathological features and is composed of myofibroblastic proliferation accompanied by inflammatory infiltrate of plasma cells, lymphocytes and eosinophils [1]. IMT is most frequently in the lung, but have reported to occur in virtually all major organs [3].

IMT have been reported to show FDG avidity on ¹⁸F-FDG PET/CT [5-7]. Dong and colleagues reported that FDG uptake in IMTs correlate with cellularity, proliferative index and degree of inflammation, which tends to be elevated in malignancy, inflammation as well as infection, making separation of these entities difficult [5]. Hence histological diagnosis is the gold standard for diagnosis. Due to the malignant potential of IMTs with reported recurrent and metastatic spread, complete surgical resection have been reported to be the treatment of choice for localized pulmonary IMTs and has shown excellent prognosis [8]. In a case series of 25 patients whom received complete resection by Fabre and colleagues, the 10 year survival was 89% with no adjuvant therapy [9]. The response of IMTs to steroid therapy along with antibiotics has been welldocumented, however worsening of IMTs with steroids as well as reoccurrences following steroid treatment have been reported [10–12]. Hence, careful monitoring of patients with IMTs is crucial and ¹⁸F-FDG PET/CT has been reported to be a useful modality in the monitoring of local recurrence and distant metastases [5].

Conclusion

An IMT is a rare lesion of the lung and diagnosis of IMT has proven to be difficult and often only possible after resection of the tumor and immunohistochemical analysis. Incomplete resection can result in malignant recurrence and worse clinical outcome, highlighting its potential malignant behavior. We presented a case of an intensely FDG avid pulmonary IMT which showed remarkable anatomical and metabolic response to oral steroid and antibiotic therapy.

Patient consent

Patient consent for publication was obtained.

REFERENCES

- [1] Coffin CM, Hornick JL, Fletcher CD. Inflammatory myofibroblastic tumor: comparison of clinicopathologic, histologic, and immunohistochemical features including ALK expression in atypical and aggressive cases. Am J Surg Pathol 2007;31(4):509–20.
- [2] Marino-Enriquez A, Wang WL, Roy A, Lopez-Terrada D, Lazar A, Fletcher C, et al. Epithelioid inflammatory myofibroblastic sarcoma: an aggressive intra-abdominal variant of inflammatory myofibroblastic tumor with nuclear membrane or perinuclear ALK. Am J Surg Pathol 2011;35:135Y144.

- [3] Palaskar S, Koshti S, Maralingannavar M, Bartake A. Inflammatory myofibroblastic tumor. Contemp Clin Dent. 2011;2(4):274–7.
- [4] Sonomura T, Hasegawa S, Takeuchi H, Ishii S, Sato Morio. Inflammatory myofibroblastic tumor of the lung indistinguishable from adenocarcinoma on imaging studies. Clin Nucl Med 2014;39(8):740–1.
- [5] Dong A, Wang Y, Dong H, Cheng C, Zuo C, Lu J. Inflammatory myofibroblastic tumor: FDG PET/CT findings with pathologic correlation. Clin Nucl Med 2014;39(2):113–21.
- [6] van den Heuvel DA, Keijsers RG, van Es HW, Bootsma GP, de Bruin PC, Schramel FM, et al. Invasive inflammatory myofibroblastic tumor of the lung. J Thorac Oncol 2009;4(7):923–6.
- [7] Dua SG, Purandare N, Pramesh CS, Karimundackal G, Menon S, Shah S, et al. Fluoro-deoxy glucose-avid endobronchial inflammatory myofibroblastic tumor mimicking bronchial malignancy: report of a case. J Cancer Res Ther 2011;7(3):340–3.
- [8] Kim TS, Han J, Kim GY, Lee KS, Kim H, Kim J. Pulmonary inflammatory pseudotumor (inflammatory myofibroblastic tumor): CT features with pathologic correlation. J Comput Assist Tomogr 2005;29(5):633–9.
- [9] Fabre D, Fadel E, Singhal S, Montepreville VD, Mussot S, Mercier O, et al. Complete resection of pulmonary inflammatory pseudotumors has excellent long-term prognosis. J Thorac Cardiovasc Surg 2009;137(2):435–40.
- [10] Khatri A, Agrawal A, Sikachi RR, Mehta D, Sahni S, Meena N. Inflammatory myofibroblastic tumor of the lung. Adv Respir Med 2018;86(1):27–35.
- [11] Schaeffer CJ, Minai OA, Sharma N, Kanne JP, Mohammed TH. Inflammatory myofibroblastic tumor of the lung: recurrence after steroid treatment. J Thorac Imaging 2008;23(3):191–3.
- [12] Panigada S, Sacco O, Girosi D, Magnano GM, Tuo P, Tarantin V, et al. Corticosteroids may favour proliferation of thoracic inflammatory myofibroblastic tumors. Pediatr Pulmonol 2014;49(3):E109–11.