& Diagnosis

Appendiceal Adenoma on F-18 FDG PET-CT in a Patient with Hodgkin Lymphoma: A Case Report and Literature Review

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Introduction

Appendiceal tumors including adenocarcinoma and carcinoid are rare, accounting for less than 1 percent of appendectomy pathology specimens [1-3]. Adenomas of the appendix are even more rare, with an incidence of less than 0.2 percent [3]. By definition, appendiceal adenomas (cystadenoma, villous adenoma, etc.) are benign tumors confined to the muscularis mucosa layer without invasion. Although the gastrointestinal tract is the most common site for extranodal non-Hodgkin lymphoma, lymphomatous infiltration of the appendix is uncommon with a frequency of 1%-3% [4,5]. Furthermore, Hodgkin lymphoma, rarely involves extra-nodal sites [6]. Knowledge of the different patterns of extranodal lymphoma manifestations is important when analyzing foci of metabolic uptake on F-18 fluorodeoxyglucose (FDG) positron emission tomography-computed tomography (PET-CT).

Case Report

A 72-year-old woman with stage 2B Hodgkin lymphoma presented as an outpatient for restaging PET-CT. The patient originally presented with weight loss, night sweats and left lower extremity swelling. Physical exam was significant for palpable firm and matted lymph nodes in the left groin. White blood cell count, hemoglobin and platelets were within normal limits, however laboratory results were notable for a mildly elevated lactate of 277 U/L. CT of the abdomen and pelvis confirmed the presence of extensive left external iliac and pelvic side wall lymphadenopathy. Percutaneous biopsy demonstrated atypical large lymphoid cells described as large mononuclear cells with large nuclei and cherry-red nucleoli, morphologically consistent with Reed-Sternberg cells. Immunohistochemical staining was CD15 and CD30 positive.

Staging PET-CT demonstrated hypermetabolic inguinal and pelvic sidewall lymphadenopathy, as well as focal intense hyper metabolism (maximum standard uptake value [SUVmax] 9.3) at the base of the appendix (Figures 1A and 2C). No corresponding discrete lesion or inflammatory changes were identified on CT. The patient endorsed depressed mood and difficulty sleeping, but denied any gastrointestinal

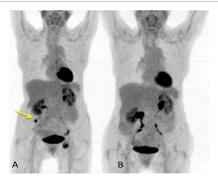


Figure 1: Staging PET-CT demonstrated hypermetabolic inguinal and pelvic sidewall lymphadenopathy, as well as focal intense hypermetabolism (maximum standard uptake value [SUVmax] 9.3).

complaints, including abdominal pain, melena or hematochezia. No right lower quadrant abdominal tenderness or palpable abnormality was identified on physical exam. Review of systems was negative for fever or chills.

Despite resolution of pelvic and inguinal adenopathy following completion of chemotherapy, metabolic activity at the base of the appendix persisted on two follow-up PET-CTs at 3 and 5 months (not shown), raising suspicion for lymphomatous involvement of the appendix. The patient continued to deny right lower quadrant abdominal pain and was clinically asymptomatic with improved lower extremity edema.

The patient subsequently underwent laparoscopic appendectomy. Gross pathology revealed a pedunculated tumor measuring at least 0.7 cm in greatest dimension. Histopathology showed increased number of glands with no significant mucin production consistent with tubular adenoma of the appendix (Figure 3). Subsequent PET-CT obtained 10 months after the original PET-CT, confirmed resolution of the hyper metabolism in the right lower abdominal quadrant (Figures 1B and 2D).

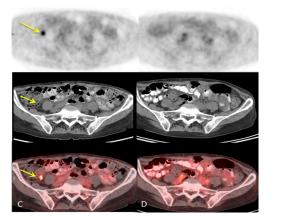


Figure 2: Histopathology showed increased number of glands with no significant mucin production consistent with tubular adenoma.

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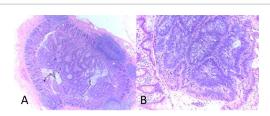


Figure 3: PET-CT obtained 10 months after the original PET-CT, confirmed resolution of the hypermetabolism in the right lower abdominal quadrant.

Discussion

A common pitfall of PET-CT in demonstrating focal metabolic activity in non-malignant lesions is well documented. To the best of our knowledge, there has been only one reported case of noninvasive appendiceal tubular adenoma, demonstrating increased metabolic uptake, in a woman with a hepatic tumor [7]. In two studies by van Hoeij et al. and Oh et al. 37-41 percent of incidental foci of increased FDG uptake on PET-CT within the colon were biopsy-proven adenomas [8,9]. Of note, these studies did not specify the presence of focal mucosal abnormality fusing to the corresponding CT images, such as a polypoid lesion or wall thickening. With almost half of incidental focal metabolic activity in the colon resulting from benign etiologies, the question arises in how to distinguish benign from malignant lesions without having to proceed with more invasive measures, such as colonoscopy, biopsy or surgical resection.

In the study of 242 patients by van Hoeij et al., malignant lesions in the colon had a statistically significantly higher SUV max than nonmalignant lesions, with median SUVmax 16.6 (interquartile range (IQR) 12.0-20.8) in adenocarcinomas compared to median SUVmax 8.3 (IQR 6.1-10.5) in non-advanced adenomas [8,10]. In our patient, the SUV max on the original PET-CT was 9.3, fitting within the reported values for a non-malignant lesion in that study. However, appendiceal tumors do not always follow this rule, as evidenced by the first described case report of appendiceal tubular adenoma in the literature, which cited a SUVmax of 15.0 [7]. Similarly, in a case report by Souza and Israel, a patient with large B-cell non-Hodgkin lymphoma and lymphomatous infiltration of the vermiform appendix had a SUVmax of only 8.0 on PET-CT [11]. This study also demonstrated a thickened and enlarged appendix on CT, while our patient showed no appendiceal mucosal abnormality on CT (Figure 2C). Other studies describing appendiceal involvement in lymphoma also reported enlargement of the appendix at pathologic examination, with an average diameter of approximately 3 cm [12]. While the presence of mucosal abnormality may be useful in predicting disease, it is not pathognomonic. In a study by Kei et al. only 12 out of 21 patients (57%) with true positive lesions (focal hyper metabolism and biopsy-proven malignancy) demonstrated soft-tissue abnormality on CT. Given the conflicting data regarding the expected SUV range for benign and malignant lesions, as well as their variable appearance on CT, many current recommendations advise that all incidentally detected colonic foci be evaluated by colonoscopy, even in the absence of any corresponding focal abnormality on CT [13, 14]. Our patient demonstrated persistent uptake in the region of the appendix and decision was made to proceed with appendectomy.

Conclusion

In conclusion, we present a rare case of an appendiceal tubular adenoma in a patient with Hodgkin lymphoma. The adenoma demonstrated focal intense metabolic uptake, raising suspicion for

lymphomatous involvement. This case serves as a reminder of the common disease presentations of non-Hodgkin versus Hodgkin lymphoma, as well as important diagnostic considerations for incidental foci of colonic FDG uptake on PET-CT.

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