Activation of cardiac sarcoidosis associated with development of gastric cancer: a case report

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Background
The high 18F-fluorodeoxyglucose (FDG) uptake in sarcoidosis lesions reflects infiltration of inflammatory cells such as macrophages. An increased incidence of cancer in patients with sarcoidosis has been suggested, and some combination of the following mechanisms has been proposed: chronic inflammation, immune dysfunction, shared aetiological agents, and genetic susceptibility to both cancer and autoimmune diseases.

Case summary
A 73-year-old man was admitted to our hospital due to effort dyspnoea. Initial investigations showed complete atrioventricular block on electrocardiography, basal thinning of the interventricular septum, and preserved left ventricular (LV) systolic function on echocardiography, and late gadolinium enhancement (LGE) in all layers of the basal interventricular septum on cardiac magnetic resonance imaging. FDG positron emission tomography/computed tomography (FDG-PET/CT) showed no abnormal uptake in the whole-body including myocardium. After discussion, corticosteroid was not initiated then. One year later, he developed stomach adenocarcinoma. Repeated investigations demonstrated enlargement of the LV (LV diastolic diameter 63 mm) and diffuse systolic impairment of LV function (LV ejection fraction 31%) on echocardiography, and abnormal focal uptake at the lateral walls of LV and hilar lymph nodes on FDG-PET/CT imaging. One more year after the surgery for gastric cancer and corticosteroid initiation, echocardiography showed recovery of systolic function and FDG-PET/CT showed no uptake in either the myocardium or hilar lymph nodes.

Discussion
In the present case, it is speculated that the first inflammation which left scarred areas showing LGE was already completed before the first FDG-PET/CT. The development of gastric cancer may be associated with the reactivation of cardiac sarcoidosis.

Keywords
Case report • Cardiac sarcoidosis • PET-CT • 18F-FDG • Malignant tumour

Learning points
• Multimodality cardiac imaging is essential for identification of patients with the early stage of cardiac sarcoidosis (CS).
• In the present case, the development of gastric cancer may be associated with the reactivation of CS.
• Cardiac re-evaluation in patients with CS may be needed before cancer surgery.
Introduction

Sarcoidosis, characterized by non-caseating granulomas, is a multisystem chronic condition of unknown aetiology, but in which some combination of infectious, genetic, and environmental factors is suspected. It has been estimated that two of three patients with sarcoidosis die of cardiac involvement. Despite the need for early diagnosis and therapeutic interventions, diagnostic confirmation of cardiac sarcoidosis (CS) is difficult because endomyocardial biopsy has low sensitivity (less than 20%) due to the focal nature of the disease. That is the reason why multimodality cardiac imaging is essential for identification of patients with the early stage of CS.

18F-fluorodeoxyglucose (18F-FDG) is a glucose analogue that is taken up by cells that have increased glucose use such as brain cells, myocytes, cancer cells, and inflammatory cells, and positron emission tomography (PET) delineates areas of high 18F-FDG uptake. The high 18F-FDG uptake in sarcoidosis lesions reflects infiltration of inflammatory cells such as macrophages. Meanwhile, an increased incidence of cancer in patients with sarcoidosis compared with the general population has been suggested. We report a case with worsening left ventricular (LV) contraction on echocardiography and newly developed myocardial focal uptake on repeated 18F-FDG PET/computerized tomography (FDG-PET/CT) images before surgery for gastric cancer.

Timeline

| December 2016 | Effort dyspnoea  
Electrocardiogram: complete atrioventricular block  
Transthoracic echocardiogram (TTE): basal thinning of interventricular septum, 59% of left ventricular ejection fraction (LVEF), and 50 mm of left ventricular diastolic diameter (LVDd)  
Cardiac magnetic resonance imaging: late gadolinium enhancement in all layers of basal interventricular septum  
Permanent pacemaker implantation  
Fluorodeoxyglucose-postion emission tomography/computerized tomography (FDG-PET/CT): no uptake in the whole-body including myocardium  
Endomyocardial biopsy: non-caseating granuloma |
| March 2018 | Epigastric discomfort  
Diagnosed as gastric cancer  
TTE: 31% of LVEF and 63 mm of LVDd  
FDG-PET/CT: abnormal focal uptake at the lateral walls of left ventricle and hilar lymph nodes |
| May 2018 | Gastric surgery |
| June 2018 | Oral corticosteroid initiation  
TTE: 52% of LVEF and 49 mm of LVDd  
FDG-PET/CT: no uptake in either myocardium or hilar nodes |
| July 2019 | |
Discussion

We report a case with CS showing deteriorating cardiac function and newly developed focal uptake in myocardium and hilar lymph nodes on FDG-PET/CT before surgery for gastric cancer. After the gastric surgery and initiation of corticosteroid, LV contraction improved and the uptake in both myocardium and hilar lymph nodes disappeared. To our knowledge, this is the first report about a patient who developed abnormal myocardial uptake in FDG-PET/CT after the development of cancer, and was diagnosed to have CS.

Myocardial uptake in 18F-FDG PET imaging reflects active inflammation in sarcoidosis. In current clinical practice, 18F-FDG PET scanning has a pivotal role in the diagnosis, therapeutic evaluation, and determination of prognosis for patients with CS. Although several reports have documented the utility of serial 18F-FDG PET scanning for evaluating the therapeutic response and effect,7–9 the usefulness of serial 18F-FDG for making the diagnosis of CS remains unknown.10

An increased incidence of cancer in patients with sarcoidosis compared with the general population has been suggested by several case reports and series, and some combination of the following mechanisms has been proposed: chronic inflammation, immune dysfunction, shared aetiologic agents, and genetic susceptibility to both cancer and autoimmune diseases. A meta-analysis study

Figure 1 Imaging findings at the first admission. (A) Complete atrioventricular block on electrocardiography. (B) Basal thinning of interventricular septum on echocardiography. (C and D) Late gadolinium enhancement of basal interventricular septum and lateral wall of the left ventricle on cardiac magnetic resonance imaging.
noted the relative risk of developing a malignant tumour in the upper digestive tract in patients with sarcoidosis to be 1.73 (95% confidence interval 1.07–2.79). In the present case, it is speculated that the first inflammation which left scarred areas showing LGE was already completed before the first FDG-PET/CT. Although the reason for the change in myocardial uptake from negative to positive in FDG-PET/CT is unclear, the development of gastric cancer may be associated with the reactivation of CS. Cardiac re-evaluation in patients with CS may be needed before cancer surgery.

**Figure 2** Serial change of \(^{18}\text{F}-\text{fluorodeoxyglucose}\) positron emission tomography findings. Baseline images showed no \(^{18}\text{F}-\text{fluorodeoxyglucose}\) uptake including in the heart (A–C). However, abnormal focal uptake was noted at the lateral walls of left ventricle (red and white arrow) and hilar lymph nodes (blue arrow) on repeated fluorodeoxyglucose-positron emission tomography/computerized tomography before surgery for gastric cancer (D–F). One year after corticosteroid initiation, fluorodeoxyglucose-positron emission tomography/computerized tomography showed no uptake in either myocardium or hilar lymph nodes (G–I).
Conclusion

We report a case with CS showing deteriorating cardiac function and new uptake in myocardium and hilar lymph nodes on 18F-FDG PET before surgery for gastric cancer.

Lead author biography

Hideki Kawai graduated from Nagoya University in 2002. His research interest is mainly focused on multimodal cardiac imaging, in particular ischaemic heart disease and cardiomyopathy.

Supplementary material

Supplementary material is available at European Heart Journal - Case Reports online.

Slide sets: A fully edited slide set detailing this case and suitable for local presentation is available online as Supplementary data.

Consent: The authors confirm that written consent for submission and publication of this case report including images and associated text has been obtained from the patient in line with COPE guidance.

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