Breast Sparganosis Incidentally Detected by FDG PET/CT

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Abstract Sparganosis is a rare, parasitic infection that is caused by the plerocercoid tapeworm larvae of the genus *Spirometra*. Sparganosis is transmitted by ingestion of larvae-containing water or intermediate hosts, such as raw amphibians, and usually presented with a subcutaneous nodule in the abdominal wall, extremities, and genital organs. Among the various involved organs, the breast is a seldom encountered site for sparganosis. However, breast sparganosis has clinical importance, since it is generally presented with an indolent palpable mass that mimics malignancy, even without evidence of inflammation. Herein, we report a case of breast sparganosis that was detected incidentally by FDG PET/CT during staging work ups in a patient with gall bladder cancer.

Keywords Human sparganosis · ^{18}F -FDG · PET/CT · Breast

Introduction

Sparganosis is a rare parasitic infection that is caused by *Spargana*, the plerocercoid tapeworm larvae of the genus

Spirometra, and usually found in East Asia (including Korea) [1]. Though it is rarer than in East Asia, sparganosis has been reported in Western countries, as well [2, 3]. Sparganosis is transmitted by ingestion of larvae-containing water or by ingestion of intermediate hosts, such as frogs or snakes [4, 5]. In humans, sparganosis frequently involves the abdominal wall, the urogenital organs, the extremities, and the central nervous system, in descending order, and presents as a subcutaneous nodule, often accompanied by vague migrating pain [6, 7]. Among the various organs involved, the breast is a seldom encountered site for sparganosis, and only few cases have been reported [8, 9].

Since it is often presented as a painless breast lump, sparganosis of the breast has clinical significance as it needs to be differentiated from breast malignancy. Breast sparganosis can mimic breast neoplasms on conventional radiological imaging of the breast, such as mammography or ultrasonography [10, 11]. However, in most cases, breast sparganosis has unique radiological features on mammography or breast ultrasonography, because infected larvae form tubular structures in the subcutaneous fat layer of the breast [12, 13]. Here, we report a case of incidentally detected breast sparganosis by ^{18}F -fluorodeoxyglucose (FDG) positron emission tomography/computed tomography (PET/CT), which the patient was undergoing for staging work ups of gall bladder (GB) cancer.

Case

An 81-year-old woman presented for an FDG PET/CT for the staging work ups of GB cancer. Complaining of discomfort in the epigastrium, she visited the emergency room at our institute and had laboratory tests and a biliary-pancreas protocol CT. The patient's CT images revealed multiple GB stones, diffuse GB wall thickening (as determined by contrast enhancement), and multiple enlarged lymph nodes in

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the portocaval region, and in the region of the hepatoduodenal ligament. According to past medical history review, the patient had recurrent attacks of epigastric pain of the same nature and had been kept under observation without further medication or intervention during the previous decade. Hence, the patient underwent a whole body FDG PET/CT under the impression that GB cancer and chronic cholecystitis are combined with cholelithiasis.

The whole body FDG PET/CT images were acquired using a Philips Gemini TF PET/CT scanner (Philips Healthcare, OH, USA), after intravenous injection of 325 MBq (8.8 mCi) FDG. FDG PET/CT revealed a nodular lesion with FDG uptake that had a maximal standardized uptake value (SUV_{max}) of 3.7 within the GB neck area, and multiple enlarged lymph nodes with FDG uptake up to SUV_{max} of 2.5 in the portocaval and hepatoduodenal ligament areas. In addition, a mass like lesion with mild FDG uptake with SUV_{max} of 1.2 was found in the right breast (Fig. 1). No other suspicious FDG uptake was presented in scanned images.

Further evaluation of the right breast mass lesion included radiological imaging modalities for breast, such as mammography, ultrasonography, and magnetic resonance (MR) imaging. The well-defined tubular structures were demonstrated by mammography, as well as by breast sonography (Fig. 2). Breast MR with gadolinium enhancement revealed tubular structures in the right breast mass (Fig. 2). Based on the imaging findings, breast sparganosis was suspected and final diagnosis was made upon the excisional biopsy; a cord-like tapeworm was extracted. At the same time, the patient underwent laparoscopic cholecystectomy for GB cancer.

Discussion

Incidental breast lesions are occasionally detected by FDG PET/CT [9, 14, 15]. It has been reported that the incidence of incidental FDG uptake in the breast was 0.82 %, and more than half of these findings (57 %) were malignant lesions [14]. Kang et al. analyzed incidental FDG uptake in the breast after dichotomizing the patient group with a

Fig. 1 Breast mass lesion detected by FDG PET/CT. **a** The axial fused PET/CT image showed a focal FDG uptake in the GB neck with a SUV_{max} of 3.7 (black arrow). **b** The whole body PET/CT detected an incidental mass-like lesion with mild hypermetabolism in the right breast (white arrows). The maximum SUV of the lesion is 1.2

Fig. 2 Representative images of radiological imaging modalities for breast. **a** & **b** Well-demarcated, radiopaque tubular structures are observed in both the cranio-caudal view (**a**) and the medio-lateral view (**b**) of mammography. **c** Well-defined, tubular structures are also demonstrated by breast ultrasonography. **d** Breast MR with gadolinium enhancement reveals multiple round and tubular-shape lesions with enhancement

cut-off value of SUV_{max} 2.0, which revealed that malignancy was confirmed in 66.7 % of the patients with high focal FDG uptake ($SUV_{max} > 2.0$) [9]. In addition, the researchers recommended further investigations, such as breast ultrasonography to assist differential diagnosis of the incidental breast lesion.

Breast sparganosis is a rare parasitic infection transmitted by ingestion of impure water that contains larvae or by eating raw amphibians. Clinical symptoms include migrating masses, pain or itching sensations accompanied with the mass or a painful migrating mass, and no sign specific to infection, such as warm sensations, edema, or tenderness was revealed upon physical examination by clinicians [16]. In this regard, thorough investigations are needed to differentiate breast sparganosis from malignancy even though it is a rare clinical condition. The final diagnosis of breast sparganosis should be confirmed by extracting the worm or examining surgical pathology specimens [6, 17].

Radiological exams, including mammography, ultrasonography, and breast MR, have revealed tubular structures caused by sparganosis of the breast [16]. In the present case, typical radiopaque tubular structures with multiple curves are clearly demonstrated in the upper outer quadrant of the right breast. Sometimes, it would be presented as a radiopaque lobular, solid mass without microcalcification in the breast, which is similar to that of breast cancer or benign tumors, such as fibroadenomas [12, 13, 16]. Also, the tubular structures with hypoechoic shadow are demonstrated by breast ultrasonography in the present case. These tubular structures are developed as the worm migrates in subcutaneous tissue or fat of the breast, since the larva has a preference for subcutaneous location and migration [8, 18]. This migrating tendency of the larva also induces the inflammation of the surrounding tissues to varied extent. Hence, breast sparganosis generates various enhancing patterns, from delayed to progressive enhancements, on breast MR according to the perilesional edema and the surrounding inflammatory reaction [17, 19, 20].

Inflammatory processes as well as malignancy can cause increased FDG uptake, since it merely reflects glucose metabolism of cells. Although not many studies on FDG uptake of parasitic infection have been performed, the degree of FDG uptake in parasitic infection seems to have a broad spectrum, from mild uptake like in the present case to high uptake causing false positive results. Several cases have been reported to have high FDG uptake (SUV_{max} 3.9–4.0) in pulmonary paragonimiasis [21]. Active inflammatory processes, including eosinophil infiltration in response to viable worms, can cause high FDG uptake in the infected sites [20]. It is known to elicit various degrees of immune reactions according to parasite types, infection site, host immune status, etc., which could be attributed to the broad spectrum of FDG uptake in parasitic infection.

Consequently, in cases of having intense FDG uptake, parasitic infection could be often misdiagnosed with cancer on FDG PET/CT.

To the best of our knowledge, this is the first case of breast sparganosis, which was incidentally found by FDG PET/CT. Initially, the incidental breast mass lesion was considered as a benign breast tumor or FDG non-avid cancer, such as lobular carcinoma, due to low FDG uptake (SUV_{max} 1.2). The patient had no specific symptoms and signs, with the exception of the breast lump. With the assistance of radiological imaging modalities of the breast including mammography, we could suspect breast sparganosis. In addition, it was revealed that the patient had a history of eating frog meat. Owing to the unique features on radiological imaging, it would not be difficult to diagnose breast sparganosis, if radiologists have enough experience and knowledge of all the possible differential diagnoses, including parasitic infection of the breast. In conclusion, breast sparganosis should be included in the differential diagnoses for incidental breast lesions, though it is a rare clinical occurrence.

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