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Case Report

Near-Complete Regression of Metastatic Typical Pulmonary Carcinoid After Five Month's Treatment with Somatostatin Analog in a 29-Years-Old Male

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1. Abstract

Typical pulmonary carcinoids are a rare form of neuroendocrine tumors (NETs) with a favorable outcome, whose treatment is still under evaluation for efficacy. We hereby report a case of a 29-years-old male who was diagnosed with typical pulmonary carcinoid after pulmonary lobectomy. In the follow-up PET-CT scan with ^{68}Ga-DOTATOC, there was evidence of multiple uptake areas, configuring a metastatic disease. He began therapy with octreotide LAR (30 mg every 28 days). After five months, a new PET-CT scan with ^{68}Ga-DOTANOC showed absence of significant uptake in the previous sites, configuring a near-complete regression of the metastatic lesions.

2. Introduction

Pulmonary neuroendocrine tumors (NET) are primary lesions showing different biological behavior according to their histotype. Typical and atypical carcinoids are the slowest replicating neoplasms and associated with a favorable outcome, whereas large cells and small cells pulmonary carcinomas often show a higher number of mitosis and a poorer outcome. The first step in treating most forms of gastroenteropancreatic (GEP) NETs is by using somatostatin analogs (SSA), whose adequacy was provided by two important studies, PROMID [1] and CLARINET [2]. However, as of now, in pulmonary NETs use of SSA is controversial because of lack of data.

3. Case Report

A 29 years old male was referred to our Endocrine Oncology Unit after lower right lobectomy for a pulmonary mass, which was diagnosed after a chest x-ray for persistent cough. Before lobectomy, he underwent bronchial biopsy, with a diagnosis of typical pulmonary carcinoid, which was confirmed on the final surgical specimen. The histopathology report on the surgical specimen indicated a typical pulmonary carcinoid infiltrating the bronchial wall, with areas of osseous metaplasia and a low mitotic count (1 per 10 HPF mitosis), along with a metastatic lymph node. According to UICC 2017 staging, the tumor was classified as a pT1b (PL0), pN2a1.

Blood tests and nuclear imaging were performed. Blood tests showed normal values of Chromogranin-A and Neuron-Specific Enolase (NSE). ^{68}Ga-DOTATOC PET-CT (Figure 1) showed an area of major uptake in the right pulmonary hilum and lesser uptake in the left pulmonary hilum, in the right anterior superior mediastinum, and in precarinal, subcarinal and lower right paratracheal lymph nodes. There also was an area of uptake in the right gluteus minimus. He didn't complain of any symptoms that could be related to carcinoid syndrome.

After collegial multidisciplinary evaluation (NETwork unit) we decided to begin therapy with a somatostatin analog, in the form of octreotide LAR 30mg every 28 days, along with deoxycholic acid 300mg q.d. and pancrelipase 5000 IU t.i.d. to avoid steatorrhea and biliary stones. The patient didn't complain of side effects, besides an initial increased bowel activity, which subsided at about six weeks into therapy.

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Five months after beginning SSA therapy, the patient underwent a follow-up ^{68}Ga-DOTANOC PET-CT (Figure 2) in a different Nuclear Medicine Unit, which was chosen for patient convenience. The imaging showed a complete absence of uptake in the known sites, with a faint uptake in the right pulmonary hilum; focal uptake was still detectable in the gluteus minimus. The patient underwent further testing with magnetic resonance imaging, which demonstrated the cystic and benign nature of the lesion. As of now, the patient is continuing therapy without complaints and further evaluations will be carried out in the following months to ensure continuation of clinical response.

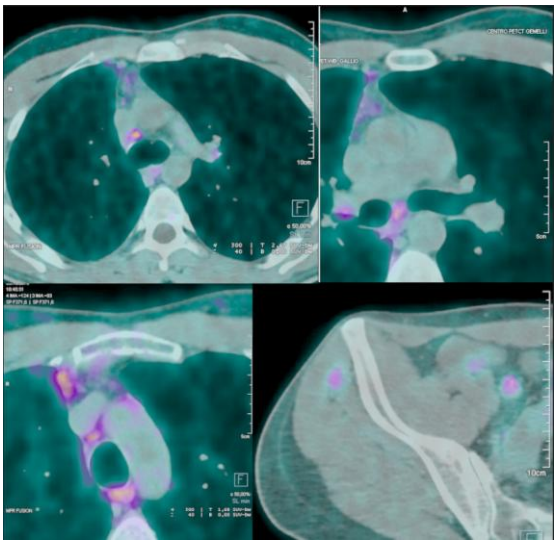

Figure 1: ^{68}Ga-DOTATOC PET-CT fusion images.

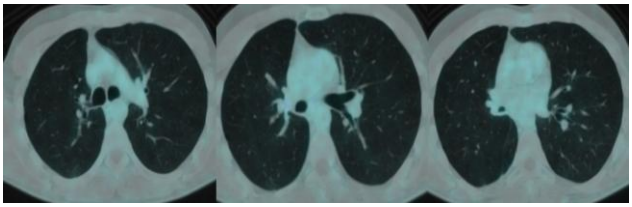

Figure 2: ^{68}Ga-DOTANOC PET-CT fusion images.

4. Methods

The first PET-CT was performed using 170 MBq of ^{68}Ga-DOTATOC; PET images were obtained sixty minutes after administration of the tracer. CT images were obtained by low-dose multilayer spiral technique with photon attenuation correction.

The second PET-CT was performed using 157.25 MBq of ^{68}Ga-DOTANOC; PET images were obtained sixty minutes after administration of the tracer. CT images were obtained by low-dose multilayer spiral technique with photon attenuation correction.

The Bronchial biopsy and the surgical specimen were reviewed by an experienced pathologist.

Magnetic Resonance Imaging was performed with Spin Echo and Short Tau Inversion Recovery with image acquisition on axial and coronal planes.

5. Discussion

Treatment with SSA in well differentiated pulmonary NETs is based mostly on midgut and foregut studies 1·2, and as of now, data is lacking. A subgroup analysis of the RADIANT-2 [3] study showed that a combination of octreotide LAR and everolimus resulted in an increase in progression free survival. The LUNA study [4] was a phase II randomized trial in which patients with advanced pulmonary NETs were treated with pasireotide, everolimus or both, with an initial evidence of efficacy and acceptable side-effects. However, data remain scarce and therefore the use of somatostatin analogs in pulmonary NETs remains controversial.

Our patient was a young man whose disease had a low profile of malignancy and a favorable outcome; however, we had evidence of a metastatic disease already from the first evaluation, prompting for a choice of adjuvant treatment. We had to choose between using SSA or everolimus, but most consensus [5,6] advice stepping up to everolimus after failure of other therapies; moreover, everolimus has a slightly worse side effect profile.

In conclusion, in our patient, treatment with octreotide LAR 30 mg every 28 days produced a near-complete regression of metastatic lesions from a typical pulmonary carcinoid.

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