

Pulmonary inflammatory myofibroblastic tumor in a breast cancer patient, a rare presentation

Julia Pastorello 1, 2, Emanuela Lando 2, 3, *, Marina Ractz Bueno 1, 2, Cristiane Pagnussat Cechetti 1, 2, Camila dos Santos do Amaral 1, 2, Giovana Zerwes Vacaro 1, 2, Nicoli Taiana Henn 1, 2, Douglas Carvalho Cogo 1, 2

¹ Department of Clinical Oncology, Hospital de Clínicas de Passo Fundo - HCPF, Passo Fundo, Rio Grande do Sul/RS, Brazil.

² Academic League of Oncology and Hematology -LAONC-HCPF, Hospital de Clínicas de Passo Fundo -HCPF, Passo Fundo, Rio Grande do Sul/RS, Brazil.

³ Medical Student, Faculdade Meridional -IMED, Passo Fundo, Rio Grande do Sul/RS, Brazil.

*Corresponding author: Emanuela Lando. Rua Paisandú, 358. Cep: 99010100 –Passo Fundo, Rio Grande do Sul/RS. Brasil. Fone: +55 (54) 9 91861467. E-mail: manu.lando@hotmail.com.

Research Ethics Committee Approval (if necessary): We declare that the patient approved the study by signing an informed consent form and the study followed the ethical guidelines established by the Declaration of Helsinki.

Received on: Oct 31, 2021. Accepted on: Nov 11, 2021. Available online: Dec 13, 2021.

Abstract

Inflammatory myofibroblastic tumor (IMT), a benign and rare neoplasm, of mesenchymal origin, pseudoarcomatous, is responsible for about 0.7% of all neoplastic presentations in the pulmonary region, being the most frequent cause of pulmonary nodules in pediatric patients, approximately 50% of the presentations. Thus, the present study aims to report a rare case of IMT in the lung site in a 31-year-old patient undergoing treatment for breast cancer, according to a literary review, relating the data according to the clinical presentation. Due to scarce documentation in the medical literature, no cases were found similar to the one described in relation to the reported oncological primary condition of the patient, making the present case a clinical challenge in modern medical practice.

Keywords: Breast Cancer; Oncology; Carcinogenesis.

Introduction

The inflammatory myofibroblastic tumor (IMT), a benign

tumor characterized by diagnostic rarity, responsible for about 0.7% of all neoplastic presentations in the pulmonary region, being the most

frequent cause of pulmonary masses in pediatric patients, about 50% of the presentations [1-3].

The diagnosis is confirmed by histological and immunohistochemical findings, since the clinical presentations in most cases are nonspecific, surgical treatment is considered the main method of choice and the chemotherapy approach is restricted to extremely reserved cases [1-3].

Thus, the present study aims to report a rare case of IMT in the lung site of a patient with breast cancer, according to a literary review, relating the data according to clinical presentation. Making the reported case extremely relevant since no case was found similar to that presented in current medical literature.

Case report

Female patient, 31 years old, diagnosed with left breast cancer cT2N3M0, hormone receptors and HER 2 negative, Ki-67: 60%, instituted neoadjuvant chemotherapy treatment based on AC-TC (carboplatin, docetaxel, doxorubicin and cyclophosphamide), performed 8 cycles.

However, patient after termination of neoadjuvant comes to the hospital in an emergency due to tremors, associated with hyperthermia (39°C) and severe myalgia in lower limbs, a hypothesis related to COVID-19 was discarded, echocardiogram showed mass vegetating in a right ventricle.

Computed tomography scans described thin scattered opacities in both lungs, nonspecific, suggesting an inflammatory process, investigating as possible a picture of associated sepsis, blood culture of a central venous catheter confirmed the presence of *Burkholderia cepacia*, and antibiotic therapy-based on cefepime and metronidazole was instituted, with Port-A-Cath catheter withdrawal, presenting frame resolvability.

However, in restaging tests, PET CT showed a hypermetabolic nodular lesion in the union of lateral quadrants of the left breast, associated with hypermetabolism in small bilateral pulmonary nodules and in secondary hilar lymph nodes (Figure 1), mediastinoscopy was contraindicated due to non-approach in hilar lymph nodes, thus video-assisted thoracoscopic segmentectomy followed by biopsy was the method of choice, the anatomopathological study confirmed the rare benign diagnosis of an inflammatory myofibroblastic tumor in the left lung.

Therefore, the patient continued with the surgical treatment related to the oncological condition, performed mastectomy, currently due to toxicity associated with adjuvant chemotherapy treatment based on capecitabine was suspended, so the patient was referred for radiotherapy treatment due to local control, if there is a plan to reintroduce capecitabine which patient is under treatment, after radiotherapy treatment,

15 fractions of 45 Gy, due to an evident extension of associated breast and lymph node disease.

The patient reported in this case has been followed since her diagnosis of

breast cancer until the moment when she is undergoing chemotherapy treatment after having surgical and radiotherapy treatment.

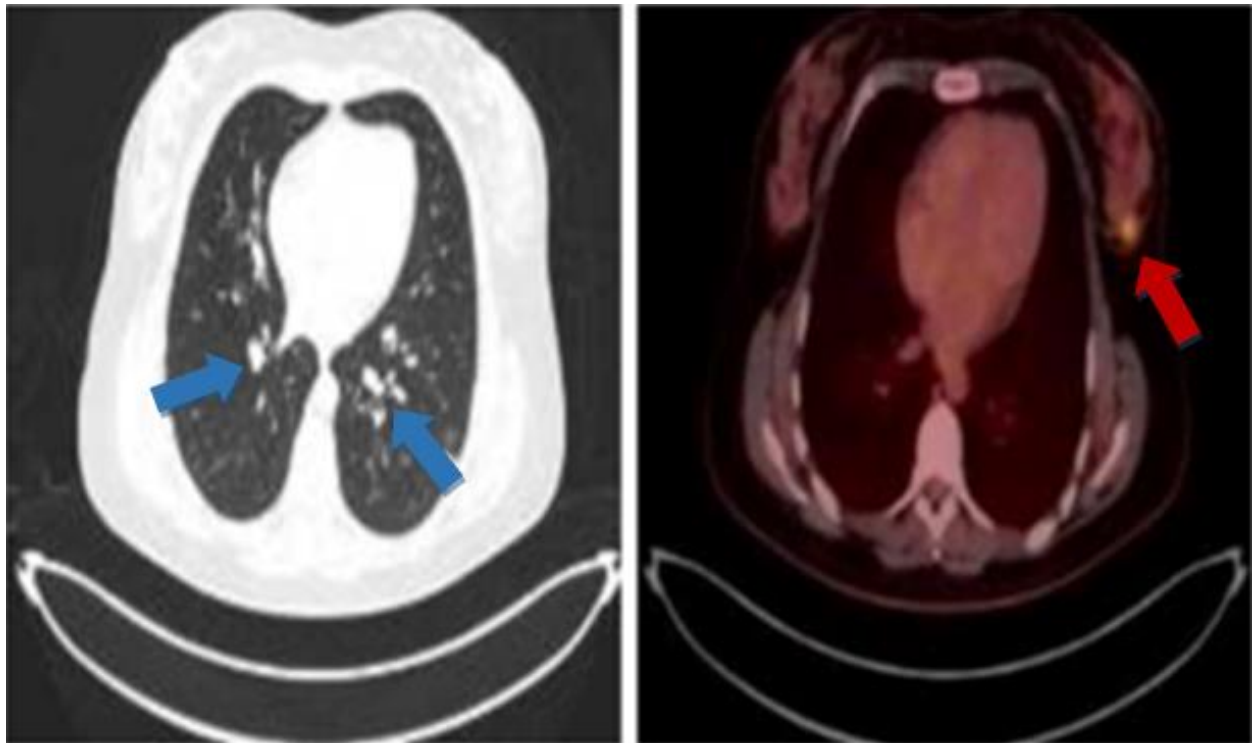


Figure 1. PET CT showed, in red arrow, a hypermetabolic nodular lesion in the union of lateral quadrants of the left breast, associated with hypermetabolism in small bilateral pulmonary nodules and in secondary hilar lymph nodes in blue arrow.

Discussion and Conclusion

Include a very brief review of similar published cases the inflammatory myofibroblastic tumor (IMT) is characterized by being a benign and rare neoplastic entity, of mesenchymal origin, pseudosarcomatous, associated with a slow growth with a mortality rate related to the affected tissue extension [1-4].

Although the location in the pulmonary site is the most prevalent, such tumor can affect any organ, being responsible for 0.7% of all lung tumors [5-6]. However, cases of IMT in patients with cancer, especially breast cancer, are extremely rare in diagnosis [5-6].

Keeping in mind literature review, which did not show previously reported cases similar to the present one. The first description of such diagnosis was made by Roth in 1980 [1-

2]. Tumor composed of a still unknown etiopathology, associated with a secondary inflammatory process, it was believed that there was a greater diagnostic predilection for the juvenile age group, especially for the female sex, however recent studies have not confirmed a direct relationship between sex and age and the clinical and radiological findings associated with it. most cases are nonspecific [5-7].

Regarding the diagnosis of IMT, this is confirmed through histological analysis associated with numerous atypical, fusiform cells, and the immunohistochemistry exam does not demonstrate great diagnostic relevance since it presents phenotypic characteristics such as staining and reactivity similar to the cases of leiomyosarcoma, rhabdomyosarcoma and sarcomatoid transitional cell carcinoma, which should be discarded [8-9].

Recent studies indicate that the presence of some antibody markers, such as the translocation of the Anaplastic Lymphoma Kinase (ALK) gene, which results in the ALK-1 variant, presents positivity between 30% and 75% of the cases of myofibroblastic tumor, another associated marker would be 1 A4, these complementary data to be interpreted in the diagnostic investigation [8-9].

Regarding the treatment of IMT, the associated evolution must be taken into account, since there may be spontaneous resolution, stability, cases

of local invasion and rarely, sarcomatous transformation [7,10].

However, the treatment of choice should be complete tumor surgical resection, and treatment associated with corticosteroids shows significant improvement in pediatric patients [11-12].

Chemotherapy treatment becomes an option in extremely reserved cases, and cases with the presence of a positive *ALK* gene, unresectable tumor or metastatic activity, anti-tyrosinakinase agents, especially crizotinib, according to the Phase II EORTIC 90101 study, become a new therapeutic possibility [13].

Thus, a clinical case becomes relevant due to the diagnostic rarity associated with the manifestation of an inflammatory myofibroblastic tumor (IMT) in the lung site in patients with breast cancer, considering the little documented medical literature, about 0.7% of all cases of lung tumors [1-3].

Since there were no cases similar to the one described above related to the primary oncological condition of the reported patient, the present case becomes a clinical challenge in modern medical practice.

References

- [1] Gentile JKA, Ruiz DF, Moricz A, et al. Tumor miofibroblástico inflamatório da vesícula biliar: relato de caso. *GED gastroenterol. Endosc Dig.* 2014;33(3):112-4.

- [2] Oliveira RF, Couto RWG, Dias AFM, et al. Tumormiofibroblástico. *Urologia*. 2017;1(4):37-40.
- [3] Pinilla I, Herrero Y, Torres MI, et al. Tumor inflamatório miofibroblástico pulmonar. *Radiología*, 2007;49(1):53- 5. doi:10.1016/s0033-8338(07)73718-9.
- [4] Ereño C, López JL, Grande J, et al. Inflammatory myofibroblastic tumor of the larynx. *J Laryngol Otol*. 2001;115(10):856-8.
- [5] Arber DA, Weiss LM, Chang KL. Detection of Epstein-Barr virus in inflammatory pseudotumor. *Semin Diagn Pathol*. 1998;15(2):155-60. PMID:9606806.
- [6] Fernández Villar A, Mosteiro Añón M, Corbacho Abelaira D, et al. Pulmonary inflammatory pseudotumor: report of 2 cases and review of the literature [Article in Spanish]. *An Med Interna*. 1997;14(9):469-72.
- [7] Donner LR, Tompler RA, White, RR. Progression of inflammatory myofibroblastic tumor (inflammatory pseudotumor) on soft tissue into sarcoma after several recurrences. *Hum Pathol*. 1996;27(10):1095-8. doi:10.1016/S0046-8177(96)90291-9.
- [8] Powell CL, Holzbeierlein JM, Wetzel L, et al. Inflammatorymyofibroblastic tumor: a case study. *Urol Case Reports*. 2014;2(5):173-175. doi:10.1016/j.eucr.2014.05.010.
- [9] Tsuzuki T, Magi-Galluzzi C, Epstein JI. ALK-1 expression in inflammatory myofibroblastic tumor of the urinary bladder. *Am J Surg Pathol*. 2004;28(12):1609-14.
- [10] Berardi RS, Lee SS, Chen HP. Inflammatory pseudotumors of the lung. *Surg Gynecol Obstet*. 1983;156(1):89-96.
- [11] Hedlung GL, Navoy JF, Galliani CA, et al. Aggressive manifestations of inflammatory pulmonary pseudotumor in children. *Pediatr Radiol*. 1999;29:112-6. doi:10.1007/s002470050553.
- [12] Verbeke JIM, Verberne AAP, Hollander JD, et al. Inflammatory myofibroblastic tumour of the lung manifesting as progressive atelectasis. *Pediatr Radiol*. 1999;29:816-9. doi:10.1007/s002470050703.
- [13] Schoffski P, Suflarsky J, Gelderblom H, et al. Prospective precision medicine trial of crizotinib (C) in patients (pts) with advanced, inoperable inflammatory myofibroblastic tumor (IMFT) with and without ALK alterations: EORTC phase II study 90101 "CREATE". In: Proc Annu Meet Am Assoc Cancer Res; 2018 Apr 14-18; Chicago, IL. Philadelphia (PA): AACR; Cancer Res 2018;78 (13 Suppl):Abstract nr CT045.
- Conflict of interest:** The author declares no conflicts of interest.
- Acknowledgements:** We thank very much the whole team for the excellent

multidisciplinary management of the case, and the patient for having trusted in our work and accepting the publication of this case report.

Funding: The author declares no funding to develop this article.

How to cite this article: Pastorello J, Lando E, Bueno MR, Cechetti CP, Amaral CS, Vacaro GZ, Henn NT, Cogo DC. Pulmonary inflammatory myofibroblastic tumor in a breast cancer patient, a rare presentation. *Brazilian Journal of Case Reports*. 2022 Jan-Mar;02(1):4-9.