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## <sup>68</sup>Ga–Prostate-Specific Membrane Antigen-11–Avid Cardiac Metastases in a Patient With Adenoid Cystic Carcinoma, A Rare Presentation of a Rare Cancer

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**Abstract:** Adenoid cystic carcinoma is a rare cancer that arises from secretory glands. Metastases are mainly seen in the lungs and bones. Cardiac metastases are extremely rare; only 1 case has previously been described. In this patient, cardiac metastases were suspected based on CT imaging. Tumor imaging by <sup>68</sup>Ga–prostate-specific membrane antigen (PSMA)-11 PET is mainly used in prostate cancer patients but is also of interest in adenoid cystic carcinoma patients because of high PSMA-ligand uptake in salivary glands. <sup>68</sup>Ga-PSMA-11 PET imaging supported the suspicion of cardiac metastases, because the cardiac lesions showed similar tracer uptake compared with other metastases.

**Key Words:** <sup>68</sup>Ga-PSMA-11 PET, adenoid cystic carcinoma, cardiac metastases, prostate specific membrane antigen

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## **REFERENCES**

- Spiro RH. Distant metastasis in adenoid cystic carcinoma of salivary origin. Am J Surg. 1997;174:495–498.
- Sung MW, Kim KH, Kim JW, et al. Clinicopathologic predictors and impact
  of distant metastasis from adenoid cystic carcinoma of the head and neck.
   *Arch Otolaryngol Head Neck Surg*. 2003;129:1193–1197.
- Laurie SA, Ho AL, Fury MG, et al. Systemic therapy in the management of metastatic or locally recurrent adenoid cystic carcinoma of the salivary glands: a systematic review. *Lancet Oncol*. 2011;12:815–824.
- Goyal J, Sidana A, O'Malley M, et al. Large renal metastasis from rare pulmonary neoplasm. *Urology*. 2011;77:1097–1098.
- Perera M, Papa N, Roberts M, et al. Gallium-68 prostate-specific membrane antigen positron emission tomography in advanced prostate cancer updated diagnostic utility, sensitivity, specificity, and distribution of prostate-specific membrane antigen-avid lesions: a systematic review and meta-analysis. Eur Urol. 2019.
- Hofman MS, Violet J, Hicks RJ, et al. [(177)Lu]-PSMA-617 radionuclide treatment in patients with metastatic castration-resistant prostate cancer (LuPSMA trial): a single-centre, single-arm, phase 2 study. *Lancet Oncol*. 2018:19:825–833.
- van Boxtel W, Lütje S, van Engen-van Grunsven ICH, et al. <sup>68</sup>Ga-PSMA-HBED-CC PET/CT imaging for adenoid cystic carcinoma and salivary duct carcinoma: a phase 2 imaging study. *Theranostics*. 2020;10:2273–2283.
- Klein Nulent TJW, van Es RJJ, Krijger GC, et al. Prostate-specific membrane antigen PET imaging and immunohistochemistry in adenoid cystic carcinoma—a preliminary analysis. Eur J Nucl Med Mol Imaging. 2017;44: 1614–1621.
- Lutje S, Sauerwein W, Lauenstein T, et al. In vivo visualization of prostate-specific membrane antigen in adenoid cystic carcinoma of the salivary gland. Clin Nucl Med. 2016;41:476–477.

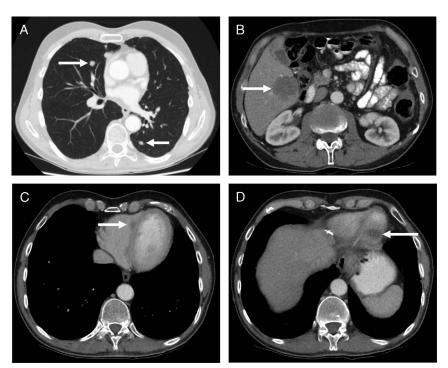


FIGURE 1. Adenoid cystic carcinoma (ACC) is a rare cancer that occurs most often in the salivary glands, but can also arise from other secretory glands, for example, in the bronchus. Adenoid cystic carcinoma is known for its often indolent growth, and patients can remain asymptomatic for years, even with distant metastases, which mainly occur in the lungs and bones. 1,2 A 55-year-old man with metastatic ACC arising from the left bronchus previously received tumor resections of the primary tumor in 2011 and of the liver (in 2013) and subcutaneous metastases (in 2014). From 2015, very slow-growing metastases arose in the liver, spleen, lungs, and bones. Because of relatively stable disease and no disease complaints, the patient did not receive systemic therapy, but was under active surveillance with contrast-enhanced CT of the chest and abdomen every 3 months. During one of these scans in 2017, cardiac lesions were observed and suspected to be metastases. Cardiac metastases in ACC patients are extremely rare; only one other case is described in literature. A shows lung metastases, B shows a liver metastasis. C and D show lesions in the cardiac septum and left ventricle wall. In retrospect, these lesions were also present on scans from 2014 and increased in size over time.

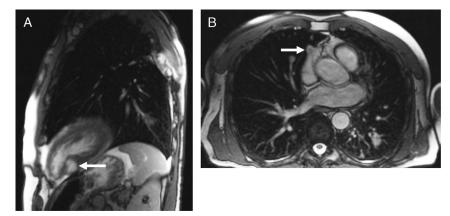
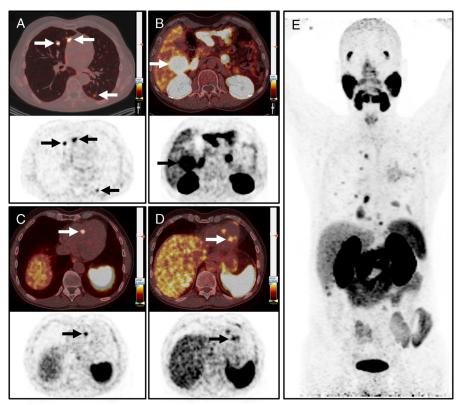


FIGURE 2. In accordance with CT, multiple intracardiac lesions were observed on cardiac MRI. The largest lesion was seen in the left ventricle wall near the apex (A, sagittal True FISP). Additionally, a pericardial lesion was seen near the right atrium (B, axial True FISP). A slightly reduced left ventricular function was observed with cardiac sonography (ejection fraction, 56%) with a normal right ventricular function. The patient experienced some heart palpitations, mostly related to exercise. However, he was generally able to cycle 140 km, as a hobby, without complaints.



**FIGURE 3.** <sup>68</sup>Ga–prostate-specific membrane antigen (PSMA)-11 PET is a relatively new imaging technique, which enables visualization of PSMA-expressing tumors. It is mostly performed in prostate cancer patients. <sup>5</sup> Patients with high PSMA expression might benefit from PSMA-targeted therapy, such as <sup>177</sup>Lu-PSMA-617. <sup>6</sup> Interestingly, healthy salivary glands also show high uptake of PSMA ligands. Therefore, salivary gland tumors have been investigated for PSMA-ligand uptake. <sup>7–9</sup> The patient presented above participated in a study (NCT03319641), <sup>7</sup> shortly after the suspicion of cardiac metastases on CT. **A** (axial fusion and PET images) visualizes tracer uptake of the lung metastases corresponding to Figure 1A and the pericardial lesion corresponding to Figure 2B. **B** shows tracer uptake of the liver metastasis (different scaling). Because the cardiac lesions (**C**, **D**) showed similar ligand uptake compared with other metastases, the PET scan supported the suspicion of cardiac metastases. **E** shows the maximum intensity projection image. Because of disease progression and cardiac metastases, the patient received 6 cycles of chemotherapy (cisplatin, doxorubicin, and cyclophosphamide), resulting in stable disease. Disease progression occurred after 1 year for which the patient is actively monitored. Currently, the patient is doing well.