

Case Report

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Takotsubo can also occur during minimally invasive treatment of paraganglioma liver metastasis!

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Abstract

Pheochromocytomas and paragangliomas are catecholamines' producing tumors with metastatic potential. In expert centers, new techniques have been developed, especially for the management of metastases in interventional radiology. However, during any therapeutic procedure, there is a risk of release of catecholamines. Intraoperative complications are poorly documented during interventional radiology. This case reports a cardiac decompensation during the management of liver metastasis of a paraganglioma by chemoembolization and cryotherapy. The main message of this case study is that cardiovascular events can occur also during interventional radiology management. These procedures require education and close collaboration between the radiologist and the anaesthetologist.

Keywords: Anesthesia; Paraganglioma; Metastasis; Interventional imaging; Ablation therapy; Hemodynamic instability; Complications; Catecholaminergic stress.

Abbreviations: CT scan: Computed Tomography Scanner; EKG: Electrocardiogram; MRI: Magnetic Resonance Imaging.

Introduction

Pheochromocytoma and paraganglioma are rare endocrine tumors located on adrenal medulla (pheochromocytoma) or extra adrenal lymph nodes (paraganglioma) and producing a variable but sometimes major amount of excess catecholamines [1]. Both can be malignant (i.e. inducing distant metastasis in lungs, liver or bones) in 10% to 15% of cases [2].

Whereas the main treatment of the primary tumor is surgery, often performed after sympathetic blockade preparation to minimize intraoperative catecholamine release, metastasis can be destroyed, or at least temporarily controlled by minimally invasive procedures such as percutaneous thermal destruction technique, chemoembolization or bone cementoplasty [3-5] and pharmacological preparation is inconsistent.

Although these metastasis are supposed, rightly or wrongly, to be less secreting than the primary tumor and interventional imaging is less invasive and stimulating than surgery, the risk of releasing catecholamines with acute cardiovascular consequences during local treatment of metastasis remains present. Very few publications have described this rare complication of a rare tumor.

The purpose of this case report is to describe an acute cardiomyopathy episode observed during mini-invasive treatment of a paraganglioma's unique liver metastasis, to remind to the readers it can also occur in this context, and how it may be treated.

Written consent has been obtained from the patient for the publication of this Case Report.

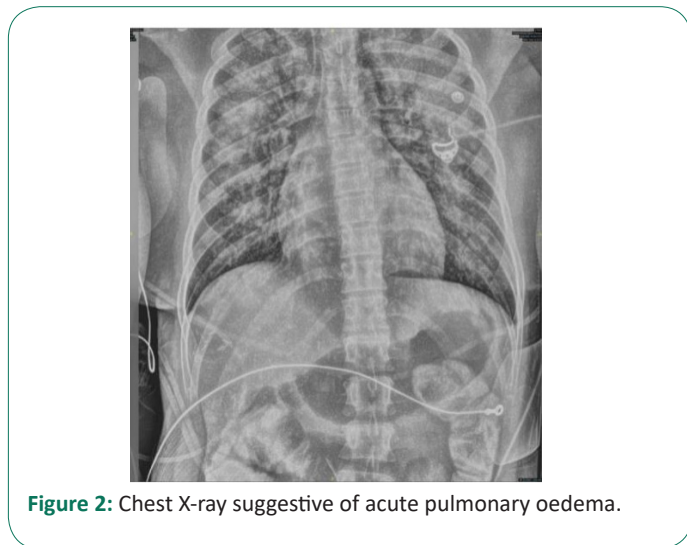
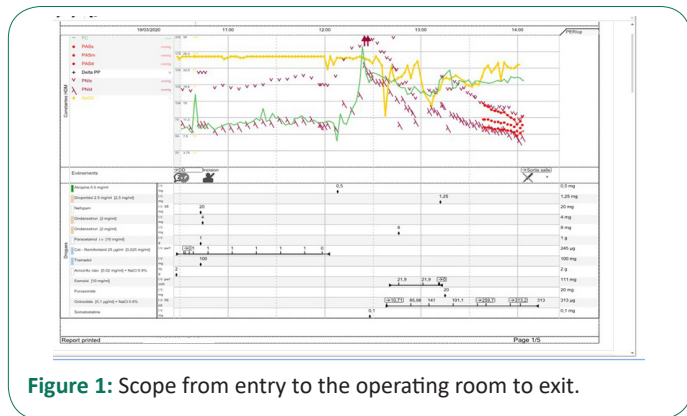
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Case report

A 31-years-old female patient was followed for a paraganglioma of the mesentery, diagnosed in 2005 on symptoms such as tachycardia, hypertension, and headache. The tumor was surgically removed then treated by somatulin. Two intrahepatic recurrences were treated by surgery in 2013 and 2016 without major cardiovascular event. In 2019, the follow-up Computed Tomography Scanner (CT scan) shows a 16 mm segment VI lesion, which was proposed for chemoembolization and cryotherapy.

The preoperative evaluation did not reveal any signs of catecholamine excess secretion and the urinary catecholamine dosage was negative. The anesthetic protocol was a sedation by remifentanyl Target Control Infusion targeting the effect-site, (Minto pharmacokinetic model, base Primea, Fresenius-Kabi Brezins France) with non-invasive intraoperative monitoring including humeral cuff blood pressure, continuous 3-lead Electrocardiogram (EKG) and digital oximetry. The patient had an initial blood pressure at operation room entry of 116/75 mmHg and a heart rate of 60 beats per minute.

Ninety minutes later, while the procedure was almost completed, hemodynamic instability appeared (Figure 1), starting with bradycardia, then hypertensive peaks (systolic blood pressure >200 mmHg), tachycardia (heart rate at 180 beats per minute) and rhythm disorders (ventricular extra systoles with bigeminy and bursts of unsustained ventricular tachycardia). They were treated by octreotide then esmolol continuous infusions.



After a few minutes, polypnea and hypoxemia then crackling sound on the lungs and red foamy sputum were observed. Hypoxemia improved with furosemide, dobutamine, norepinephrine and non-invasive ventilation and the patient was transferred to the intensive care unit.

In Intensive Care, transthoracic echography showed myocardial sideration (only apex contracting), Left Ventricular Ejection Fraction at 30%, and chest X-ray showed a typical pulmonary edema (Figure 2). Dobutamine could be stopped immediately, norepinephrine few hours later and non invasive ventilation the next day.

Echographic left ventricula kinetics and chest X-rays normalized by day 3. Troponin level was initially at 8700 pg/ml then decreased gradually.

The diagnosis of cardiogenic shock due to inverse TakoTsubo on probable catecholaminergic release was retained. The patient was discharged from the intensive care unit at Day 4 and from hospital at Day 7. Control cardiac Magnetic Resonance Imaging (MRI) at 4 months was normal.

Discussion

This case shows that pheochromocytoma hypertensive crisis may occur during the less invasive procedure when performed on an endocrine tumor.

Induced by stress, drugs, or food, it can occur in the perioperative period and reveal the diagnosis [6,7]. Most of time but not often reversible [8] with an appropriate symptomatic treatment, it should be cured before surgery to optimize the outcome.

It can be targeted by the stress of induction, intubation or tumor manipulation but can also occur during another type of surgery far from the endocrine tumor location as for example during a knee arthroscopy [9].

Described during the main tumor removal, hypertensive crisis of cardiac failure was rarely observed during secondary locations, especially if treated by mini-invasive technique. Only Deljou published a small serie of percutaneous ablation of extra-adrenal metastasis with 2 patients / 21 showing cardiac events but none with acute cardiac dysfunction [10]. Takotsubo syndrome or stress cardiomyopathy has been quite recently associated with pheochromocytoma [11].

Our case shows that such cardiac event can occur in this context. To detect and treat it without delay, physicians should all be aware of the risk and patients should be assessed before the procedure by a multidisciplinary team including the anesthesiologist, surgeon, radiologist and endocrinologist to identify the number and locations of metastasis, assess the level of secretion (more secreting is the tumor higher is the risk of cardiovascular event [12]), chose the right procedure and discuss the rational for a pharmacological preparation, even if this preparation is still debated [13].

Per operative monitoring including EKG, repeated Non Invasive Blood Pressure measures, oxygen saturation, and cardiovascular drugs and ventilation uses should be as narrow as for surgery, even for procedures performed with minimal or with-

out anesthesia.

In our case, the patient had a favorable outcome despite severe hypoxemia because of symptomatic treatment by Non Invasive Ventilation, antiarrhythmics and diuretics immediately available.

Conclusion

In summary, severe acute cardiovascular events may occur during removal surgery or mini-invasive treatment of metastasis for malignant paraganglioma or pheochromocytoma. Favorable outcome is supported by the education of all the interventional team, closed cardiovascular monitoring, rapid symptomatic treatment with resuscitation equipment and professionals available.

Declarations

Conflicts of interest: None of the co-authors has conflict of interest related to the publication.

Author's contributions: SANCELME Elise: This author wrote the manuscript. DEMIRI Migena: This author conceived of the presented idea and contributed to the writing of the manuscript. RIVES Jean-Phillipe: This author conceived of the presented idea and contributed to the writing of the manuscript. TSELIKAS Lambros: This author contributed to the final version of the manuscript. BILLARD Valerie: This author contributed to the writing of the manuscript, provided critical feedback, and helped supervise the project.

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