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Calcific pancreatitis uncovering a multiple endocrine neoplasia type 1: A case report in compliance with the scare guidelines

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ABSTRACT

INTRODUCTION: Chronic pancreatitis are mostly linked to alcoholic consumption or biliary lithiasis; and Primary hyperthyroidism (PHPT) is still a very rare association and the exact physiopathology is yet to be fully unveiled to the human knowledge. We present the first case report of a calcific pancreatitis associated with not only PHPT but a multiple endocrine neoplasia (MEN) type 1.

CASE PRESENTATION: We report the case of a 52 years old woman suffering from mellitus diabetes consulting the emergency rooms for acute pancreatitis with hyperlipasemia and hypercalcemia whom final imaging discovered a pituitary gland adenoma, a left surrenal adenoma, and a parathyroid adenoma, and for the pancreas it revealed an acute mild pancreatitis with a background of calcifications, no gallstones, no bone or renal abnormalities; and the parathyroidectomy was performed following the minimally invasive selective technique.

DISCUSSION: Calcemia levels testing routinely performed help discover hyperparathyroidism. The associations of chronic pancreatic inflammation to hyperparathyroidism needs to be studied, even if hypercalcemia is proven to be a risk factor of pancreatitis; the mechanism behind this association is briefly described. parathyroidectomy is the definitive cure for hyperparathyroidism, the technique advances has shown effective localization of the responsible adenoma and the intraoperative testing of parathormon levels after resection decreasing is a very reliable extemporaneous sign for the success of the procedure.

CONCLUSION: The endocrine system is synchronized; meaning the injury of one gland should start the search for others. In our case, the first main lead should not have been the acute pancreatitis but her diabetes. In the future we suggest that diabetes primary explorations may need a pancreatic imaging and endocrine explorations even though it could get pricier for the healthcare system, but giving the complications that we could prevent; it is to be considered.

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1. Introduction

Repeated inflammatory episodes of the pancreatic gland conduct its replacement with a fibrotic tissue leading to an endocrine and exocrine deficiency generating a chronic pancreatitis (CP) [1]. on the other hand the multiple endocrine neoplasia (MEN) type 1 is diagnosed by the occurrence of at least 2 of the 3 tumors of the endocrine system (parathyroid, pituitary, adrenal gland and endocrine pancreas) and it is an autosomal dominant disease related to menin gene mutation [2]. The associations of calcific pancreatitis and primary hyperparathyroidism (PHPT) have been

outlined as a rare association and the study of the pancreatic disease caused by PHPT is not fully elucidated [3].

In compliance with the scare guidelines [10] We present a case of an acute pancreatitis on a background of chronic calcific pancreatitis bringing out into open a multiple endocrine neoplasia type 1 with a parathyroid adenoma, pituitary adenoma and surrenal gland adenoma, giving birth to an analytical and interesting case of endocrine diseases, we believe that we have the first ever case aligning such associations (Figs. 1–4).

2. Case presentation

We present the case of a 52 years old lady with uncontrolled diabetes type II under oral anti-diabetics, also suffering from asthma under fluticasone, with no relevant drug or family history, consulting at the emergency room for an acute abdominal pain with vomiting for about 10 days beforehand, the clinical exam found a hemodynamically and respiratory stable patient with

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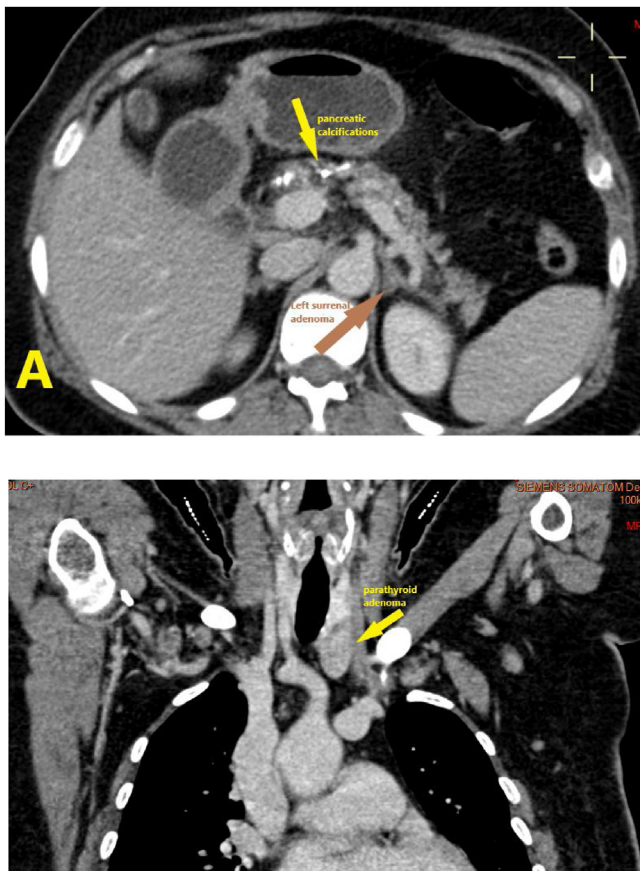


Fig. 1. A- abdominal CT showing the pancreatic calcifications and the left adrenal gland adenoma B-neck CT showing the parathyroid adenoma.

epigastric sensibility, the laboratory exams showed an elevated calcium rate and a normal rate of amylasemia and lipasemia, the computed tomography (CT) aligned with a magnetic resonance cholangiopancreatography(MRCP) found a mild acute pancreatitis and a background of chronicity with calcifications of the pancreas without gallstones, a left surrenal gland adenoma and a parathyroid adenoma (P3). the patient at this stage has been hospitalized at the surgery department for treatment of her acute pancreatitis and explorations for her hypercalcemia and multiple endocrine neoplasia.

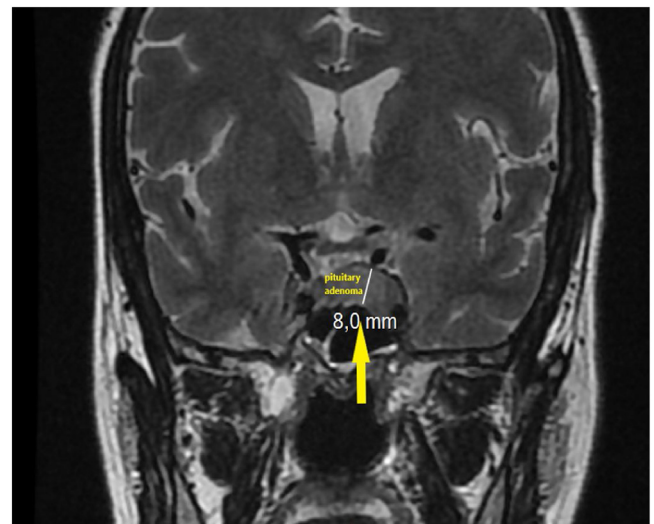


Fig. 2. Cerebral pituitary MRI showing the pituitary gland adenoma.

Further evaluation included an ultrasound of the neck viewing the parathyroid nodule along with a unilateral thyroid node and the intact parathormone (iPTH) assay. A raised iPTH along with a raised serum calcium and low phosphate clinched the diagnosis. Normal levels of urinary catecholamins were confirmed. The cerebral magnetic resonance imaging (MRI) found a pituitary adenoma completing the multiple endocrine neoplasia type1 syndrome. At this stage, a pet-scan was indicated but due to the covid19 crisis and the non-availability of this imaging technique, the decision of undergoing surgery was made.

The procedure was performed by our Head surgeon and the patient underwent a minimal invasive approach with a selective unilateral parathyroidectomy with measurement of parathyroid hormone. Total time of surgery was 20 min. post operative biological evaluation showed a normalized calcemia, no supplements were needed.

There were no postoperative complications. Mean duration of hospital stay was two days.

The patient undergoes a follow up with our endocrinology unit to control the diabetes along with the surveillance of the multiple endocrine neoplasia

The enlarged gland's histopathology showed an adenoma. Adequate intraoperative parathyroid hormone decrease was observed.



Fig. 3. Operative view after the resection of the parathyroid adenoma – specimen of parathyroidectomy.

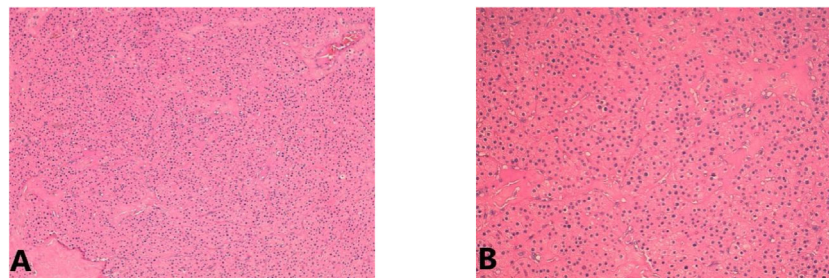


Fig. 4. Histological image of the parathyroid adenoma.

A- Microphotography showing a cellular, homogenous lesions, made of oxyphil cells in delicate capillary network. (HE, 100X).

B- Microphotography showing bland appearing cells. There were no capsular invasion, no vascular invasion, or invasion of adjacent tissue (HE, 400X).

Postoperative calcemia was normal. With no suspicion of persistent hyperparathyroidism.

3. Discussion

1 alpha-hydroxylase production is increased by high levels of parathyroid hormone (PTH) therefore increasing renal and intestinal absorption of the calcium leading to hypercalcemia [3]; and the most common cause of hyperparathyroidism is the parathyroid adenoma; mostly sporadic but also may be familiar (MEN 1–MEN 2 and MEN 4) in about 10 % of the cases, this familiar form has changed overtime its main clinical form once outlining bone disease have switched to be asymptomatic (80–90 %) or originating very atypic general symptoms including gastrointestinal manifestations caused by hypercalcemia that increases gastrinemia and reduces neuromuscular excitability therefore being a risk factor for gastric ulcers and pancreatitis [4] although numerous studies have established the association between pancreatitis and PHPT; in 2006 Jacob et al. have found that 13 % of patients with PHPT had pancreatitis all of them presenting high levels of blood calcium, suggesting a causal effect between the pancreatic disease and PHPT through hypercalcemia [5] and following the same course SK Bhadada et al. in 2007 compared PHPT pancreatitis with alcoholic and idiopathic pancreatitis concluded that the chronic pancreatitis associated with PHPT have characterized biochemical and clinical manifestations that are extremely relieved after parathyroidectomy [6] and also in 2018 Thareja et al. colliged 70 patients with acute and chronic pancreatitis from whom 9.4 % had PHPT concluding to the necessity of dosing the calcium levels for every case of pancreatitis and hypercalcemia should draw attention for endocrine or malignant diseases association thus the physiopathology of the pancreatic disease is yet to be fully uncovered leaving space for skepticism [3]. If histopathological study can prove the formal features of chronic pancreatitis (CP) it definitely cannot distinguish it's etiology according to the international consensus guidelines for CP and the gold standard for the diagnosis is not the histopathology but it will differentiate between CP from neoplasia or autoimmune disease [7].

Imaging is key to identify the localization of the adenomas and the complications of the chronic pancreatitis, the adenoma is localised by 99-Technetium scanning and high-resolution ultrasonography. Computed tomography (CT) and magnetic resonance imaging (MRI) are useful to identify ectopic glands and other endocrine tumors [9].

If the association of these 2 diseases is established but yet still controversy, the treatment consists of pain treatment according to its assessment in term of intensity and pattern following the pain relief ladder; surgical or endoscopic treatment are only discussed for complicated or symptomatic CP, and medical treatment for exocrine pancreatic insufficiency by oral pancreatic enzymes is only allowed after a nutritive assessment and malabsorption identification; however the diabetes mellitus secondary to CP is

to be treated with insulin therapy and oral metformin only with a low risk for hypoglycemia according to the United European Gastroenterology evidence-based guidelines [8].

On the other hand PHPT treatment will provide an etiologic treatment of the CP, and the Parathyroidectomy is the only definitive treatment for symptomatic PHPT and also indicated for MEN associated PHPT, under general or local anesthesia and with a full explorative or selective surgery with intraoperative study of iPTH levels that should drop by 50 % 10 min after extraction of the gland, if the iPTH levels doesn't decrease by half or stay within a normal range, an ectopic secreting gland should be sought [9].

4. Conclusion

There is no doubt that PHPT engenders either acute or chronic pancreatitis, and it is very unconventional how this association is often brought to light, moreover laying hands on one endocrine tumor must initiate the search for all of them endocrine tumors; that is why a hypercalcemia found on a routine blood work should aggregate the systematic endocrine explorations ; in our case we believe it to be one of a kind associating not only chronic calcifying pancreatitis to hyperparathyroidism but to a whole MEN type 1 breaking the ice around an unknown endocrine world of associations.

Declaration of Competing Interest

The authors report no declarations of interest.

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Ethical approval

It's a one case report needing no ethnical approval.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

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Have written the article, Have consulted the patient, prescribed all of the tests and prepared the patient for surgery and participated in the surgery.

A. Miry: Achraf Miry: achrafmiry@outlook.com (anatomopathology resident): have helped writing the article, confirm the histological diagnosis.

H. Mirali: Houda Mirali: mirali.houda@gmail.com (radiology resident) : have helped with the imaging analysis.

A. Bennani: Amalbennani: (anatomopathology professor): confirm the histological diagnosis.

M. Bouziane: Mohammed bouziane: bouzianemohammed@hotmail.com (oncology surgery professor): have supervised the writing of the paper, and has been the leader surgeon of the case).

Guarantor

M. Bouziane: Mohammed bouziane: bouzianemohammed@hotmail.com (oncology surgery professor).

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