



## Pancreatic IPMN Cancer: A Rare Case of Bone Metastases

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### Abstract

We report a case of a 70-year-old male with 1-month history of 9 kg weight loss and abdominal pain. Physical examination was normal and hepatic tests revealed a cholestasis. CT scan showed a pancreatic head mass associated with biliary ducts dilatation and an echo-endoscopic-guided fine needle aspiration of this lesion suggested an intraductal papillary mucinous neoplasm (IPMN) with mild dysplasia.

A duodenopancreatectomy of the head of the pancreas was performed and histological images have shown a mixed adenocarcinoma with endocrine differentiation developed over an IPMN with one ganglion metastasis and negative resection margins. Subsequently, the patient received a treatment with 6 cycles of gemcitabine.

A pulmonary metastasis appeared after one year of follow up and it was resected with success.

Three years after the 4th lumbar vertebrae presented a lytic pattern and was treated with cementoplasty, but lumbar pain got worse. Lumbar vertebrae magnetic resonance imaging (MRI) showed a multi-lobulated lesion with peripheral enhancement in this vertebrae and a pseudo-collection along the subcutaneous and bone path of the cementoplasty's needle. A percutaneous biopsy was performed and revealed a metastatic disease. The patient presented later multiple bone cystic lesions in the pelvis and spine.

This case illustrates a rare feature of IPMN bone metastases, rarely described on literature. This diagnosis should be considered when a history of cystic pancreatic tumour is known and an invasive procedure like biopsy is required to confirm the diagnosis.

**Keywords:** Pancreatic cancer-IPMN-cystic; Bone metastases

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### Introduction

IPMN of the pancreas are a pancreatic neoplasm originating from the mucinous epithelium of the pancreatic duct. There are three classifications of IPMN disease among cystic pancreatic tumours, according to their localization and extension within duct system as main duct (MD), branch duct (BD) or mixed.

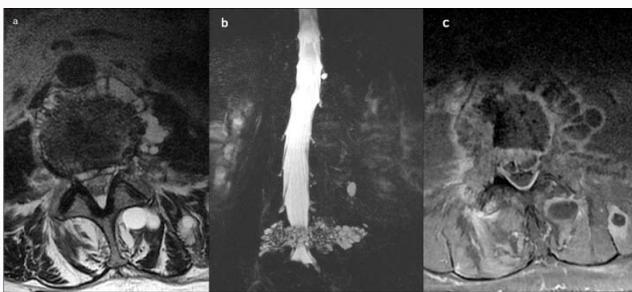
IPMN are classified as benign or malignant (parenchymal invasive adenocarcinoma) according to their degree of dysplasia. Some prognostic factors for malignancy include their localization, size, histology subtype, positive margins, nodal metastases, vascular and perineural invasion [1,2]. Main duct involvement as intestinal or biliopancreatic pathological phenotype means a higher risk of malignancy[1].

The outcome of a cystic pancreatic carcinoma, as well as pancreatic adenocarcinoma, once invasive is poor [3]. IPMN malignancy can occur in 30 to 88% of patients, as *in situ* or invasive carcinomas. *In situ* carcinoma is considered when confined to the ductal structure, while invasive carcinoma is suggested when it infiltrates the pancreatic parenchyma [1]. IPMN metastases are very rare and bone metastases have been rarely described.

The pattern of metastases may be unusual mimicking abscess, which makes the diagnosis challenging in imaging methods.

### Case Presentation

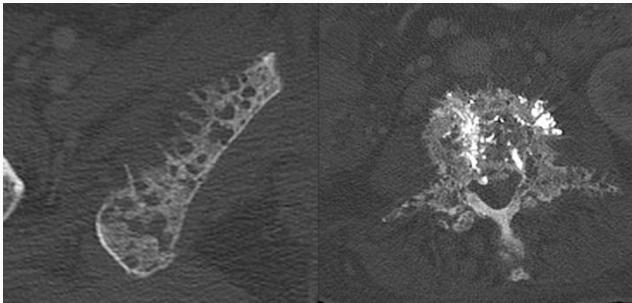
A 70-year-old male diagnosed with IPMN with mild dysplasia underwent a duodenopancreatectomy of the pancreas' head. Histological images showed a mixed adenocarcinoma with endocrine differentiation developed over a mixed localisation, main (MD) and branch ducts (BD). The margins were negative but there was one nodal metastasis. The pathological phenotype



**Figure 1:** MRI examination, axial T2 (a) myelo-MR on coronal view (b) and axial T1 fat saturation after contrast injection (c) cystic pattern of vertebral metastases with paravertebral and posterior multiple cysts and peripheral enhancement mimicking abscess after cementoplasty.



**Figure 3:** 3D reconstruction shows bone lacuna in both ischium and 4<sup>th</sup> vertebral body.



**Figure 2:** CT examination on bone kernel showing ischium and vertebral involvement by metastases with multiple "holes", a specific feature due to the initial nature of pancreatic tumour.

was intestinal and biliopancreatic. Subsequently, the patient received a treatment with 6 cycles of gemcitabine.

During follow up, one year after the end of the chemotherapy, a hypermetabolic pulmonary lesion was diagnosed and treated with inferi or right lobe resection.

Two years after, the patient presented with backache in lumbar region and a lumbar CT scan revealed one single lytic lesion in the fourth lumbar vertebrae treated with cementoplasty, without release of the pain. An MRI was performed and showed a multi-lobulated lesion with peripheral enhancement in the vertebra and a pseudo-collection along the subcutaneous and bone path of the needle of cementoplasty (Figure 1). There was no modification of the disc sign or endplate of the adjacent vertebrae. A percutaneous biopsy was performed and showed a metastatic spread of the disease with cystic pattern, similar to the pancreatic tumour. The patient presented later multiple bone cystic lesions in the pelvis and the spine (Figures 2 and 3).

## Discussion

Cystic lesions of the pancreas are increasingly recognized. While some lesions show benign behaviour (serous cystic neoplasm), others have an unequivocal malignant potential as mucinous cystic neoplasm, IPMN involving the pancreatic duct and solid pseudopapillary neoplasm.

Partial resection with intra-operative frozen section examination of the transaction margin is recommended for treatment [3]. Eight per cent of the patients have recurrence even with negative margins and a yearly follow-up with preferably non-radiating imaging (e.g.

MRI or EUS) is suggested. The liver, the chest and the peritoneum are the most frequent sites of metastases in the majority of patients who recurred with invasive IPMN [1]. This case is unusual because the bone metastases of IPMN are rarely described.

Concerning to the differential diagnosis of bone cystic lesions, abscess and primitive lesions as unicameral bone cysts (UBC) and aneurysmal bone cysts (ABC) may be considered in case of unique localisation.

Unicameral bone cysts are common benign non-neoplastic bony lesions, seen mainly in childhood, typically in the metaphysis of long bones as humerus and femur. UBC's can be rarely seen in adults, and when present is in unusual locations such as in the talus, calcaneus or iliac wing. MR signal characteristics are T1 low signal and T2 high signal, with no fluid-fluid levels unless there has been a complication with haemorrhage.

ABC are also benign lesions and are mostly diagnosed in children and adolescents, typically located in long bones of lower limb, spine and sacrum. MRI demonstrates the characteristic fluid-fluid levels with a solid component.

In conclusion, bone metastases in case of IPMN are really rare, and in our knowledge this is the first case illustrating the multi-cystic pattern of IPMN metastases. That entity should be considered when a patient as a history of IPMN and a cystic pattern is seen in the bone.

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