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Case Report

Undifferentiated carcinoma of the pancreas with osteoclast like giant cells: Literature review with CT/MR imaging findings in 3 cases ☆,☆☆

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ABSTRACT

Undifferentiated carcinoma with osteoclast-like giant cells of pancreas (UCOGCP) is a relatively rare tumor worldwide. Its accurate preoperative diagnosis is extremely difficult. Because the mass is usually large and closely related to neighboring structures, it is difficult to locate the tumor and it is often misdiagnosed as pancreatic cancer, neuroendocrine tumor or gastrointestinal stromal tumor. Combining literature to analyze UCOGCP clinical features (including age of onset, prevalent location) and imaging features (including lesion size, mass nature), to explore the value of preoperative CT and MRI in the diagnosis and differential diagnosis of UCOGCP and hope to help clinical diagnosis and treatment.

Introduction

Undifferentiated carcinoma with osteoclast-like giant cells of pancreas (UCOGCP) is a relatively rare tumor worldwide [1,2]. If we can rely on imaging diagnostics to make an accurate diagnosis of UCOGCP before surgery, including the extent of the disease, whether there is invasion of adjacent structures, whether there is lymph node metastasis and distant metastasis, etc., it can help clinicians to formulate the corresponding surgical plan. Therefore, preoperatively accurate imaging di-

Abbreviations: CT, computed tomography; MRI, magnetic resonance imaging; UCOGCP, Undifferentiated carcinoma with osteoclast-like giant cell of pancreas; T1WI, T1-weighted image; T2WI, T2-weighted image; DWI, diffusion-weighted imaging.

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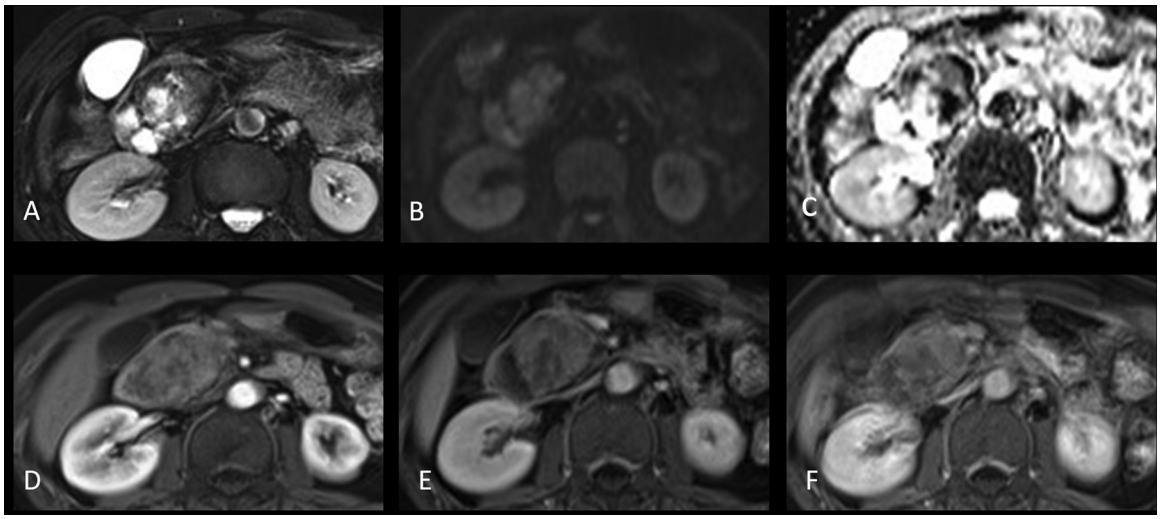

Fig. 1 – The abdominal magnetic resonance imaging (MRI) shows a 10 × 5 cm heterogeneous mass arising from the head of the pancreas. The T2WI image shows heterogenous high-signal intensity with multifocal cystic lesions (A). Diffusion-weighted imaging shows a mass with high signal intensity (B). The ADC value corresponding to the high signal part in the diffusion-weighted image of the lesion is significantly reduced (C). The contrast-enhanced MRI shows a solid portion slightly enhanced in arterial phase (D) and continuously enhanced in portal venous and delayed phases (E, F).

agnosis is extremely important. We report 3 cases of patients with UCOGCP confirmed by surgery and pathology with complete clinical, pathological, and imaging data collected in our hospital.

Clinical presentation

Three cases of UCOGCP patients confirmed by surgery and pathology in our hospital were reported.

Case report

Case 1

A 66-year-old female patient with a pancreatic mass found on physical examination for 1 week. On physical examination, there was a mild tenderness in the upper abdomen, with no other discomfort. Abdominal magnetic resonance imaging (MRI) (Fig. 1) showed a large (approximately 10 × 5 cm) mass arising from the head of the pancreas that invaded the adjacent descending duodenum. Contrast-enhanced MRI showed a mixed cystic and solid mass that was slightly enhanced in the arterial phase and continuously enhanced in the portal venous phase. No regional lymphadenopathy, ascites, or metastasis was observed on MRI. The patient underwent pancreaticoduodenectomy; she recovered and was discharged 8 days postoperatively.

Case 2

This was a 68-year-old female patient who presented with repeated dull pain in her left upper abdomen for 3 months. A gastroscope from a local hospital revealed “gastric ulcer,” but she was admitted to our hospital after conservative treatment with drugs. She had a 10-year history of hypertension and diabetes and a surgical history of appendectomy. Abdominal computed tomography (CT) scan (Fig. 2) showed the presence of a round-shaped mass (5 × 6 cm in diameter) arising from the tail of the pancreas, with a clear boundary and uneven high density on plain scan. Contrast-enhanced CT scan showed a mixed cystic and solid portion that was slightly enhanced in the arterial phase and continuously enhanced in the portal venous and delayed phases; it appeared to have an uneven density. The abdominal magnetic resonance (MR) T2-weighted image showed a mass with heterogenous high-signal intensity and multifocal cystic and hemorrhagic lesions. The T1-weighted image showed a mass with low-signal intensity and multifocal hemorrhagic lesions. In an enhanced scan, most of the lesions were not obviously enhanced; only the edge of the mass was slightly enhanced. The T1-weighted image also showed a small amount of exudate adjacent to the splenic hilum, uninvaded splenic arteries and veins, and an unenlarged pancreatic duct. Thus, the diagnosis of pancreatic tail cystadenoma with hemorrhage was made based on the preoperative CT scan and MRI.

Case 3

This was a 44-year-old male with repeated upper abdominal pain and discomfort for 2 months, which was intolerable and worsened at night. A weight loss of 3 kg was noted in over 2 months. His medical history revealed hypertension. Abdominal CT scan (Fig. 3) showed a large (approximately 17 × 11 cm), dumbbell-shaped mixed cystic and solid mass in the left upper abdomen. The main body of the mass was uneven and had low density. The small, patchy, high-density shadow in the mass may indicate bleeding. The mass was closely adherent to the adjacent stomach, duodenum, jejunum, and colon.

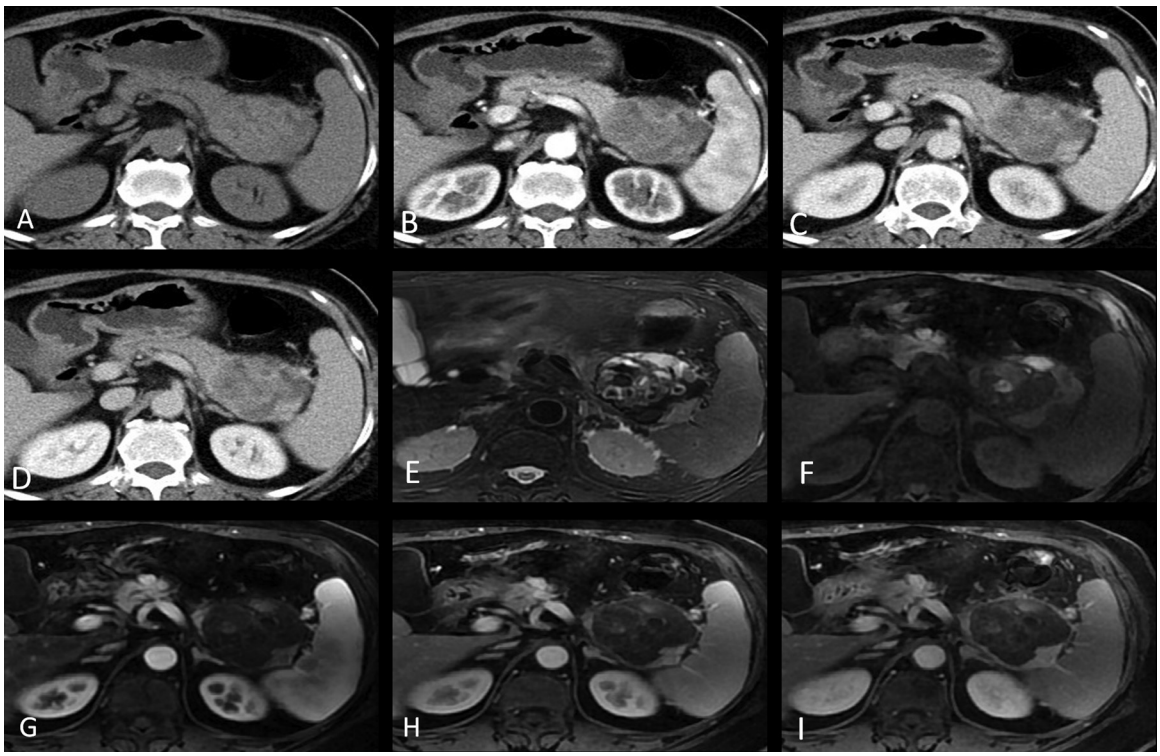

Fig. 2 – Abdominal computed tomography (CT) shows the presence of a round-shaped mass (5 × 6 cm in diameter) arising from the tail of the pancreas, with clear boundary and uneven high density (A). The contrast-enhanced CT scan shows a solid portion slightly enhanced in arterial phase (B) and continuously enhanced in portal venous and delayed phases (C, D). Abdominal MR T2-weighted image shows multifocal cystic and hemorrhage lesions with heterogenous high-signal intensity (E). The T1-weighted image shows a mass with multifocal hemorrhagic lesions with low-signal intensity (F). In an enhanced scan, most of the lesions were not obviously enhanced, and only the edge of the mass is slightly enhanced (G, H, I).

In an enhanced scan, the solid component and partition of the mass had delayed enhancement, whereas the cystic component was not significantly enhanced. The patient underwent surgical treatment. The postoperative pathologic examination showed UCOGCP (Fig. 3F). The patient refused any adjuvant chemotherapy. He died due to tumor recurrence and liver metastasis 5 months postoperatively.

Laboratory inspection indicators

All 3 of our patients had relevant laboratory tests (including CA125, CA19-9, APF and ferritin) before surgery. Among them, 2 cases of ferritin were slightly increased, and 1 case was accompanied by a slight increase of CA125, and the other indexes were all within the normal range.

Discussion

UCOGCP is extremely rare, accounting for <1% of pancreatic malignancies [1–4]. The true histogenesis of UCOGCP is still unknown. Researchers have concluded the possibility of mesenchymal or epithelial origin based on immunohistochemistry and molecular biology findings [5].

UCOGCP is also called osteoclast-like giant cells pleomorphic carcinoma/sarcomatoid carcinoma of the pancreas, as first reported by Rosai in 1968 [6]. It is composed of undifferentiated epithelial and/or mesenchymal cells admixed with nonneoplastic osteoclast-like giant cells [7]. UCOGCP is classified by the World Health Organization as a rare variant of pancreatic ductal adenocarcinoma, with 2 distinct cell lines, including a mononuclear cell line population and osteoclastic tumor cells [8].

UCOGCP is more common in middle-aged and elderly patients (mostly 60-70 years old), but its incidence in male and female is controversial [8–9]. The tumor is usually found in a large volume, with an average diameter of 8 cm, which might be related to rapid tumor growth, with relatively low malignancy and late clinical symptoms. In our study, the tumors were usually large in size; the shortest diameter of the tumor was ≥5 cm in 3 cases, of which 1 case had shortest diameter of the tumor >8 cm. The neoplasms are mostly located in the body and tail of the pancreas [3,10–11]. In this paper, 3 cases of tumor located in the head, body and tail of the pancreas accounted for 1 case each. We speculate that it may be due to fewer cases.

Despite its large volume, tissue infiltration and lymph node metastases are not common. In some studies, the 10-year survival rate of patients with UCOGCP has been reported, which

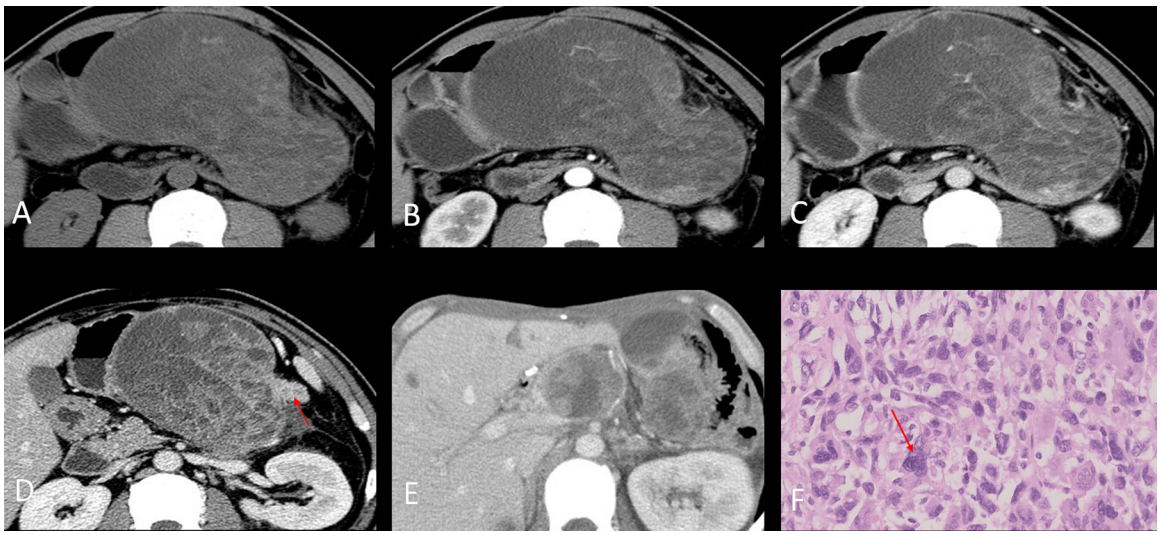

Fig. 3 – CT scan shows a large dumbbell-shaped cystic solid mass in the body of the pancreas. The main body of the mass is uneven and of low-density, with small, patchy, high-density shadows(A). The contrast-enhanced CT scan shows the solid component and partition of the mass with gradual enhancement, while the cystic component did not significantly enhance (B, C). A thin-layer image shows the mild pancreatic duct dilation in the tail of the pancreas (short arrows, D). The reexamination at 5 mo after the operation shows tumor recurrence and liver metastasis (E). The postoperative pathology shows that the tumor tissues mainly consist of monocyte-like tumor cells and osteoclast-like giant cells (short arrows, F). The nuclei of the tumor cells were round or spindle-shaped, with different shapes and rich eosinophilic cytoplasm. The cytoplasm of osteoclast-like giant cells contains several oval nuclei. (HE staining, 200x magnification).

shows that this tumor may have a better prognosis than undifferentiated carcinoma of the pancreas without osteoclast-like giant cells [3,4,12–15].

The main signs and symptoms of UCOGCP include abdominal pain, palpable mass, fatigue, and weight loss [16]. The related tumor markers, such as CA-125, CA 19-9, AFP, CEA and ferritin, are usually normal or slightly elevated [17]. In our 3 patients, we found that ferritin was slightly elevated in 2 cases, 1 of which was accompanied by a slight increase in CA-125, while the other indexes had normal indicators, which is consistent with previous studies.

Based on relevant literature reports, the CT scan features of UCOGCP are mainly mixed cystic and solid masses with clear boundaries [18]. The masses are generally large and uneven in density, with cystic necrosis and bleeding and small separation shadows in it [19–20]. The tumors in the head and neck of the pancreas tend to cause pancreatic duct dilatation [8]. The CT scan findings of all cases in our study were similar to those mentioned in the literature. Slight peripheral enhancement, internal solid parts in the arterial phase, and continuous enhancement in the portal venous and delayed phases were observed. The MRI features of UCOGCP may be related to the complex pathological changes inside the tumor. The literature mentions that the MRI features of UCOGCP include low-signal intensity in T1WI and high-intensity central cystic part in T2WI, with low-intensity interval and solid surrounding tissue [21–22]. MRI often exhibits characteristic changes, such as high T1W1 patchy signal and low T1W1 and T2W1 signals caused by hemosiderin deposition [23]. In an enhanced MRI scan, the mass is unevenly enhanced and the enhancement is more obvious in the venous and delayed phases than the arterial phase. CT scan and MRI can show tumor progression and whether the tumor is hemorrhagic and necrotic to some extent [23], but UCOGCP cannot be confirmed.

CT or EUS-guided fine-needle aspiration (FNA) is an effective method for tumor cytology diagnosis. However, most literatures have reported that preoperative FNA can increase the incidence of postoperative complications, and the long-term survival rate is lower than that of patients without FNA [20]. None of the 3 cases we provided did not do FNA before surgery.

Because most of UCOGCP are cystic and solid masses, they should be differentiated from absorption phase of pancreatitis. The disease has a history of pancreatitis, CT/MR shows peripancreatic inflammatory exudation, localized as a wrapped mass, but the density/signal of it is relatively uniform, usually called pseudocyst.

Conclusions

UCOGCP is more common in elderly patients and its imaging finding is a large mixed cystic and solid mass in the pancreas. It can be pathologically diagnosed postoperatively; its accurate preoperative diagnosis is extremely difficult. The final diagnosis needs to be determined based on histopathology and immunohistochemistry findings [8].

Author contributions

Subjective image analysis: Kun Zhan, Shizheng Zhang. Conceptualization, Data curation, Methodology, Resources, Writing - original draft, Writing - review & editing: Kun Zhan. Resources, Writing - review & editing: Zhongfeng Niu.

Patient consent

This retrospective study was approved by the institution's Committee on Human Research, and the patient's written informed consent has been obtained.

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