



Case report

A case report of pancreatic mucoepidermoid carcinoma responded to gemcitabine and paclitaxel

Yuan Chen^a, Changren Zhu^b, Peng Xu^{c,**}, Jie Yao^{c,*}^a Yangzhou University Medical College, Yangzhou, Jiangsu Province, China^b Pathology Department, Clinical Medical College, Yangzhou University, Yangzhou, Jiangsu Province, China^c Department of Hepatobiliary and Pancreatic Surgery, Clinical Medical College, Yangzhou University, Yangzhou, Jiangsu Province, China

ARTICLE INFO

Keywords:

Mucoepidermoid carcinoma
Pancreas
Gemcitabine
Paclitaxel
Case report

ABSTRACT

Mucoepidermoid carcinoma (MEC) typically manifests in the salivary glands, but occurrences in the pancreatic gland are exceedingly rare. Surgical resection proves effective; however, pancreatic MEC is prone to metastasis, and lacking a standardized postoperative treatment. We discussed the experience of a 51-year-old female patient with pancreatic MEC who received paclitaxel and gemcitabine as postoperative care. Within a predetermined amount of time, this regimen successfully stopped the spread of metastatic tumors and returned tumor markers to normal. A Stable Disease status was achieved within 6 months after chemotherapy. In summary, gemcitabine and paclitaxel display efficacy in treating pancreatic MEC.

1. Introduction

Mucoepidermoid Carcinoma (MEC) is a common primary malignant tumor of the salivary glands [1], first described by Stewart et al. in 1945 [2]. Pancreatic mucinous epidermoid carcinoma is even rarer, to our knowledge, with only 13 cases reported in English literature [3]. Diagnosis of MEC still relies on pathological examination due to the absence of typical imaging characteristics and tumor markers. The prognosis of pancreatic MEC is extremely poor, with patients often struggling to survive beyond one year even after radical surgery [4]. Besides surgical resection, there is no standardized treatment strategy for pancreatic MEC. Here, we present a case of pancreatic MEC and discuss its treatment process.

2. Case presentation

A 51-year-old female with a two-month history of upper abdominal pain was admitted to the Subei People's Hospital of Jiangsu province (China), in November 2022. She denied any history of gastrointestinal illness or immunological disease previously. Physical examination showed mild tenderness in the upper abdomen. The computed tomography (CT) reveals a 28mm × 19mm mass-like low-density lesion in the head of the pancreas with indistinct borders and irregular enhancement (Fig. 1A and B). The tumor marker examination showed a carbohydrate antigen (CA) 19-9 level of 51.30 U/mL (normal: <30 U/mL) and a CA 50 level of 59.67 U/mL (normal: <30 U/mL), while the levels of alpha-fetoprotein (AFP) and carcinoembryonic antigen (CEA) were within the normal range.

* Corresponding author.

** Corresponding author.

E-mail addresses: pengxu1986@126.com (P. Xu), dryaojie@yzu.edu.cn (J. Yao).

The patient did not receive any treatment before admission. As a result, suspicion of a pancreatic carcinoma, a curative pancreaticoduodenectomy was performed on December 5, 2022, after a multidisciplinary team discussion.

The postoperative pathology revealed tumor tissue with a gray-yellow appearance, moderate texture, and no lymph nodes observed around the pancreas. Microscopically, the tumor tissue exhibited a nested distribution consisting of epidermoid cells, intermediate cells, and mucous cells (Fig. 2A). Immunohistochemical staining of the tumor showed positive expression for CKpan, CK7, P63, and P40, and negative staining for Hepa, AFP, and CK20 (Fig. 2B–H). In addition, the proliferation index by Ki-67 staining was above 30 % (Fig. 2I). These pathological results were consistent with the diagnosis of pancreatic MEC.

The final pathology diagnosis was mucoepidermoid carcinoma. The postoperative course was uneventful, and the patient was discharged two weeks after surgery. From two months post-surgery, the patient received paclitaxel (albumin-bound) (1st day: 140 mg/m², 8th day: 70 mg/m²) and 1000 mg/m² of gemcitabine (on the 2nd and 9th day), repeated every 21 days.

Two months post-surgery, the patient underwent a follow-up abdominal CT, which revealed liver metastasis of the tumor (Fig. 1C). After undergoing five rounds of chemotherapy, the patient's tumor marker levels gradually decreased to within the normal range (Fig. 3), and there was some reduction in the size of the metastatic tumor foci (Fig. 1D–F). Within 6 months after chemotherapy, the patient's outcome was evaluated as Stable Disease (SD) according to the RECIST criteria (version 1.1). The patient is still undergoing regular follow-up examinations at our hospital and has survived for 12 months post-surgery.

3. Discussion

MEC is a common malignant tumor in the salivary glands, composed of epidermoid cells, intermediate undifferentiated cells, and poorly differentiated adenocarcinoma cells [5]. However, it is exceedingly rare in the pancreas. Since Franz first reported pancreatic MEC in 1959, only 13 cases have been reported in English literature [3] (Table 1). Pancreatic MEC is more commonly found in the body and tail of the pancreas, and the liver and lymph nodes are the sites where metastasis is more likely to occur [3].

At present, there is no standardized treatment regimen for this disease, and curative surgery remains an effective approach for extending survival. Additionally, postoperative multimodal therapy shows promise in enhancing the patient's prognosis [6]. However, there is currently no evidence-based recommendation for the choice of chemotherapy regimen. Hepatic artery infusion of oxaliplatin in combination with intravenous infusion of bevacizumab, 5-fluorouracil, and cetuximab has shown a favorable treatment response [6]. Adjuvant therapy with gemcitabine and cisplatin has also shown promise in improving the patient's prognosis [7]. Supplemental chemotherapy has been shown to potentially prolong the patient's survival [8]. However, a more detailed analysis of the effectiveness of this regimen in tumor suppression cannot be determined due to the lack of disclosed follow-up data from the patient.

In this case, the patient developed liver metastases just three months after surgery. After undergoing treatment with gemcitabine and paclitaxel, the patient's CA 19-9 and CA 50 expression levels gradually returned to normal. Additionally, CT results indicated that the size of the metastatic lesions was suppressed. These results show that the chemotherapy regimen can effectively inhibit the progression of metastatic tumors. However, in the later stages of chemotherapy, the patient's tumor marker levels gradually increased again, indicating disease progression. This could be associated with drug resistance in the tumor. Despite the potential decrease in effectiveness during the later stages, chemotherapy still plays a role in controlling the tumor progression to some extent.

This patient underwent the first CT scan two months after the surgery, revealing tumor metastasis. Conducting an earlier

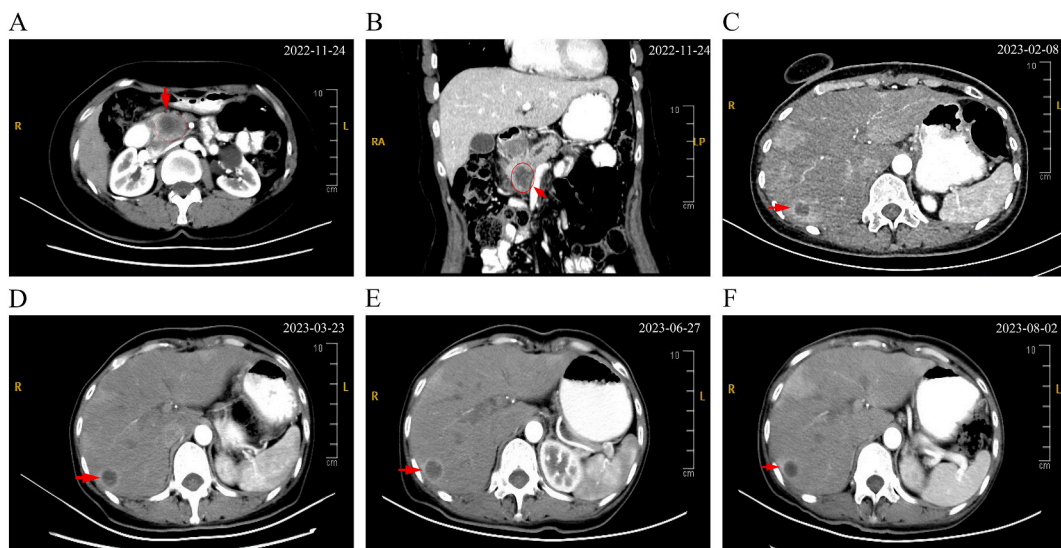


Fig. 1. Preoperative and postoperative enhanced computed tomography. (A–B) There is a mass-like relatively low-density lesion visible in the pancreatic head region, with an indistinct border, measuring approximately 28mm × 19mm. (C–F) The patient developed tumor metastasis to the liver post-surgery, and the tumor size was inhibited after regular chemotherapy.

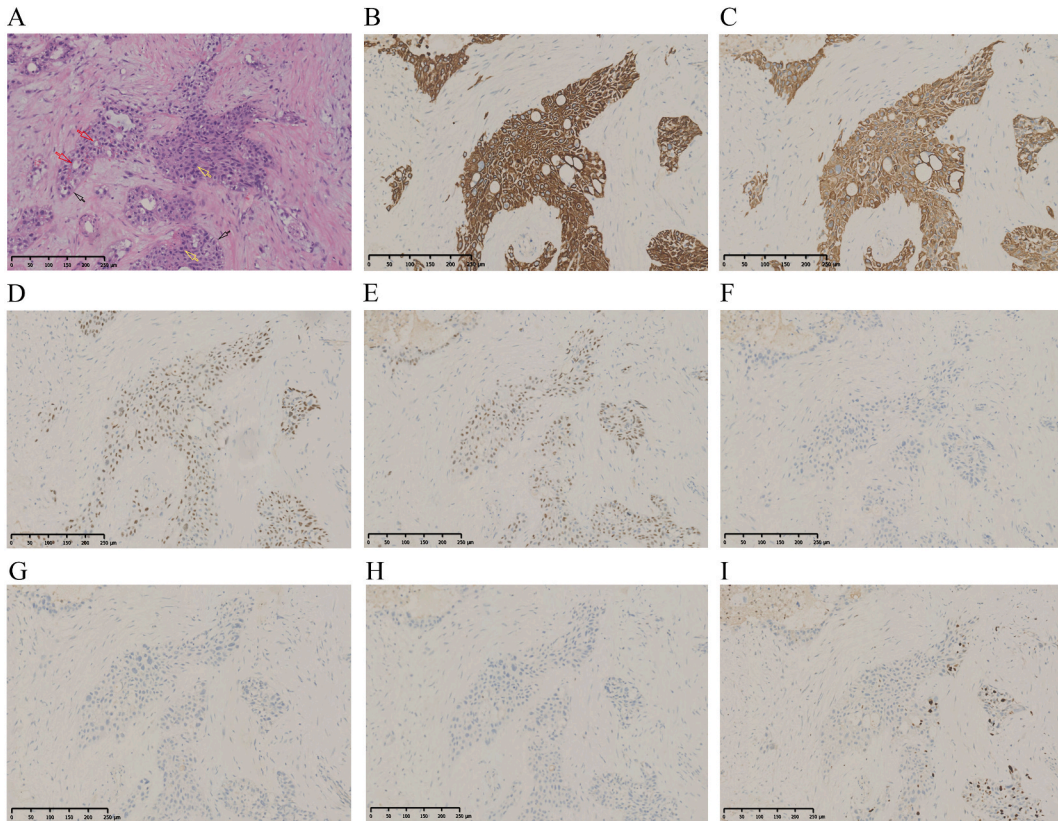


Fig. 2. Pathological examination of the tumor. (A) Hematoxylin–eosin staining revealed the tumors consisted of 3 cell types: mucous cells (red arrow), epidermoid cells (black arrow), and intermediate cells (yellow arrow). (B)The tumor was positive for CKpan. (C) The tumor was positive for CK7. (D) The tumor was positive for P63. (E) The tumor was positive for P40. (F) The tumor was negative for Hepa. (G) The tumor was negative for AFP. (H) The tumor was negative for CK20. (I) The tumor Ki-67 staining was above 30 %.

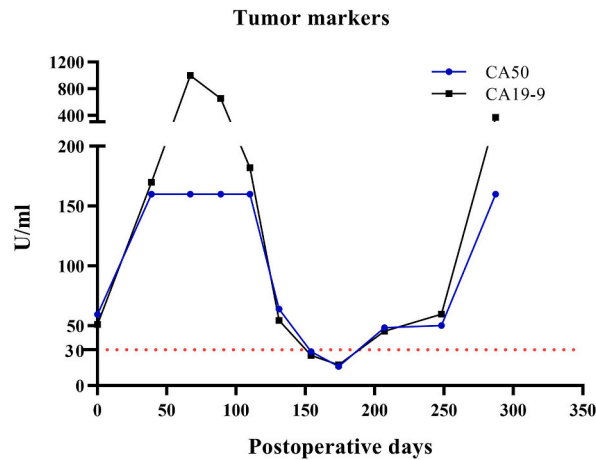


Fig. 3. Postoperative levels of tumor markers in the patient. After 5 cycles of postoperative chemotherapy, the patient’s tumor marker levels returned to normal. The normal value for CA 19-9 is < 30 U/ml, and the detection limit is 1000 U/ml. The normal value for CA 50 is < 30 U/ml, and the detection limit is 120 U/ml.

postoperative CT scan can facilitate the detection of small metastatic lesions more easily. It is crucial to regularly monitor tumor markers and conduct imaging examinations to achieve better chemotherapy outcomes. Making timely adjustments to the chemotherapy regimen may enhance the survival time of patients.

In summary, we presented a rare case of pancreatic MEC and shared the postoperative chemotherapy regimen along with the

Table 1
Reported cases of pancreatic mucoepidermoid carcinoma in the English literature.

Case	Year	Sex/Age (year)	Diameter (cm)	Location	Treatment	Disease Progression	Survival (Month)
1	1987	M/58	10	pancreatic tail	None	Multiple organ metastases (Lung, gallbladder, spleen, adrenals) and lymph node metastases	2
2	1987	F/69	T2	pancreatic head	PD	Vessel invasion (mesocolon and superior mesenteric vein)	3
3	1991	F/48	3.5	pancreatic body	TP	Single liver metastasis	11
4	1992	M/58	3.5	pancreatic head	PD	Lymph node metastases	6
5	1993	M/57	8	pancreatic body	Intraoperative radiation	Multiple organ metastases (Lung, liver, colon, stomach)	2
6	1995	M/64	8	pancreatic tail	DP	Multiple organ metastases (spleen, kidney, colon, adrenal, liver) and peritoneal dissemination	11
7	2002	M/65	10	pancreatic body	UN	None	UN
8	2012	F/63	4.5	pancreatic body and tail	DP	None	12
9	2016	M/50	17	pancreatic head	TPD	Liver, lymph node metastases	45
10	2018	M/48	4	pancreatic body and tail	DP	Lymph node metastases	23
11	2018	F/67	2	pancreatic body	UN	Multiple liver metastases	UN
12	2020	M/56	4.5	pancreatic tail	DP	Multiple liver metastases, multiple organ metastases	3
13	2022 (current case)	M/51	2.8	pancreatic head	PD	Multiple liver metastases, Lymph node metastases	12 (live)

Abbreviations: F, female; M, male; PD, pancreaticoduodenectomy; DP, distal pancreatectomy; TPD, total pancreaticoduodenectomy; UN, unknown.

patient's disease progression. The chemotherapy regimen of gemcitabine and paclitaxel resulted in a measurable but time limited beneficial effect with metastasis tumor volume reduction and reduction of CA 19-9 and CA 50 expression levels. These findings demonstrate tumor response to the gemcitabine and paclitaxel regimen. Further studies are needed to better define the role of gemcitabine and paclitaxel for the treatment of MEC. This contributes to the exploration of the optimal treatment strategies for this disease.

4. Patient perspective

When the patient was diagnosed with malignant tumor, they exhibited extreme anxiety. They once hesitated about whether to undergo postoperative chemotherapy. However, upon learning that their chemotherapy was effective, they felt relieved about their past decision.

Ethics statement

All participants/patients (or their proxies/legal guardians) provided informed consent for the publication of their anonymised case details and images.

Funding

This work was supported by The National Natural Science Foundation of China under Grant number 81772516 and Provincial-level discipline leader of the NJPH under Grant number DTRB202213.

Data availability statement

Data included in article/supp. Material/referenced in article.

CRedit authorship contribution statement

Yuan Chen: Writing – review & editing, Writing – original draft, Resources, Formal analysis, Data curation. **Changren Zhu:** Writing – review & editing, Resources, Formal analysis. **Peng Xu:** Writing – review & editing, Resources, Conceptualization. **Jie Yao:** Writing – review & editing, Funding acquisition, Conceptualization.

Declaration of Competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to

influence the work reported in this paper.

References

- [1] C.H. McHugh, D.B. Roberts, A.K. El-Naggar, et al., Prognostic factors in mucoepidermoid carcinoma of the salivary glands, *Cancer* 118 (16) (2012) 3928–3936, <https://doi.org/10.1002/cncr.26697>.
- [2] F.W. Stewart, F.W. Foote, W.F. Becker, Muco-epidermoid tumors of salivary glands, *Ann. Surg.* 122 (5) (1945) 820–844, <https://doi.org/10.1097/00000658-194511000-00005>.
- [3] Z. Chen, L. Zhang, J. Huang, et al., Targeted-gene sequencing and bioinformatics analysis of patients with pancreatic mucoepidermoid carcinoma: a case report and literature review, *OncoTargets Ther.* 14 (2021) 3567–3581, <https://doi.org/10.2147/OTT.S305248>.
- [4] R. Ma, Y.Q. Yu, J.T. Li, et al., Mucoepidermoid carcinoma of the pancreas: a case report and a review of literature, *J. Res. Med. Sci.* 17 (9) (2012) 886–889.
- [5] J. McKenzie, J. Lockyer, T. Singh, et al., Salivary gland tumours: an epidemiological review of non-neoplastic and neoplastic pathology, *Br. J. Oral Maxillofac. Surg.* 61 (1) (2023) 12–18, <https://doi.org/10.1016/j.bjoms.2022.11.281>.
- [6] P. Pandey, R.N. Al-Rohil, J.B. Goldstein, et al., Cutaneous metastasis of a mucoepidermoid carcinoma of the pancreas: first reported case, *Am. J. Dermatopathol.* 38 (11) (2016) 852–856, <https://doi.org/10.1097/DAD.0000000000000604>.
- [7] H.J. Hu, R.X. Zhou, F. Liu, et al., You cannot miss it: pancreatic mucoepidermoid carcinoma: a case report and literature review, *Medicine (Baltim.)* 97 (11) (2018) e9990, <https://doi.org/10.1097/MD.00000000000009990>.
- [8] F.F. Zhou, L.M. Lu, Clinicopathologic analysis of mucoepidermoid carcinoma in the pancreas with literature review, *J of Wannan Medical College* 32 (5) (2013) 371–373. 周芳芳, 卢林明胰腺黏液表皮样癌临床病理分析并文献复习 [J]. 皖南医学院学报, 2013, 32(05): 371-373.