

## CASE REPORT

---

# Combined Serous Cystadenoma and Pancreatic Endocrine Neoplasm. A Case Report with a Brief Review of the Literature

Harsh Mohan<sup>1</sup>, Sukant Garg<sup>1</sup>, Raj Pal Singh Punia<sup>1</sup>, Ashwini Dalal<sup>2</sup>

Departments of <sup>1</sup>Pathology and <sup>2</sup>Surgery, Government Medical College and Hospital,  
Chandigarh, India

### ABSTRACT

**Context** The presence of a combined serous cystadenoma and pancreatic endocrine neoplasm is a distinct clinicopathological entity rather than the incidental concurrence of two separate entities.

**Case report** We report the case of a 52-year-old woman admitted to our hospital who had suffered from epigastric pain, nausea and vomiting for 4 months. Imaging techniques showed an irregular mass having a mixed solid and cystic consistency, arising from the body of the pancreas and involving the lesser sac. This mass went beyond the stomach and above the lesser curvature of the stomach. The diagnosis of combined microcystic adenoma and pancreatic endocrine neoplasm was made. The patient had an uneventful postoperative course and is well two months after surgery.

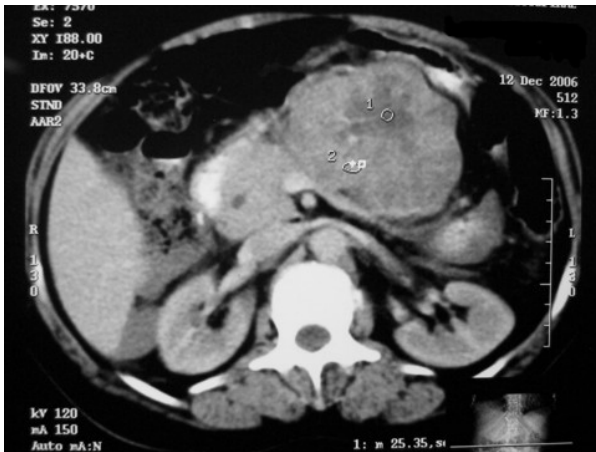
**Conclusion** This case emphasizes the importance of careful gross examination, sampling and reporting of pancreatic tumors. The coexistence of pancreatic endocrine neoplasms with potential malignant behavior may be overshadowed by obvious benign tumors such as a microcystic serous cystadenoma. The malignant potential and prognostic features of this neoplasm require long-term follow-up and additional data from subsequent reports of such cases.

### INTRODUCTION

Microcystic serous cystadenoma also known as glycogen-rich adenoma, is a rare, usually benign cystic pancreatic tumor which most often occurs in older men from 60-80 years of age [1, 2]. These tumors are often solitary, arising in an otherwise normal pancreas. Less frequently, these may be associated with underlying pancreatic disorders, such as chronic pancreatitis, possibly the result of duct obstruction secondary to the compressive effects of an expanding serous cystadenoma [3]. These may occasionally occur in association with other pancreatic neoplasms such as pancreatic ductal carcinoma or pancreatic endocrine tumors [3, 4, 5]. We describe a mixed neoplasm, composed of both a serous cystadenoma and a pancreatic endocrine neoplasm.

### CASE REPORT

A 52-year-old woman who had suffered from epigastric pain, nausea and vomiting for four months was admitted to our hospital. She had an eight year history of diabetes mellitus. Physical examination revealed a palpable abdominal mass in the epigastrium which moved with respiration. Ultrasonography showed an irregular mass having a mixed solid and cystic consistency, arising from the body of the pancreas and involving the lesser sac. The mass went beyond the stomach and above the lesser curvature of the stomach. A

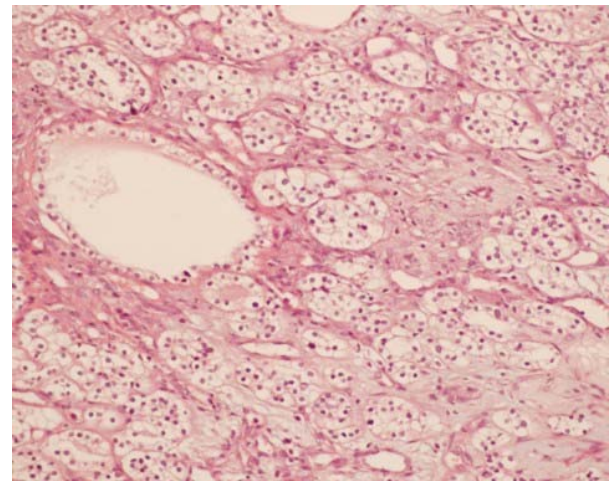


**Figure 1.** A contrast-enhanced CT scan of the pancreas showing a mass in the region of pancreas with hypodense areas.

contrast enhanced computed tomography (CECT) scan of the patient showed a growth measuring 10x8x6.9 cm arising from the body of the pancreas (Figure 1). This was compressing the stomach. Biochemical analyses, including serum amylase and bilirubin, were within normal limits. A complete hemogram showed no abnormality. A clinical diagnosis of cystic pancreatic neoplasm was made. Exploratory laparotomy with complete excision of the pancreatic tumor was carried out and sent for histopathological examination. The surgical specimen consisted of a tannish pink, multilocular soft tissue mass measuring 10x8.8x7.2 cm. It went from well-circumscribed to encapsulated. A cut section showed cystic areas ranging from 0.2 cm to 2.5 cm in diameter alternating with grayish white solid areas (Figure 2). The cystic spaces



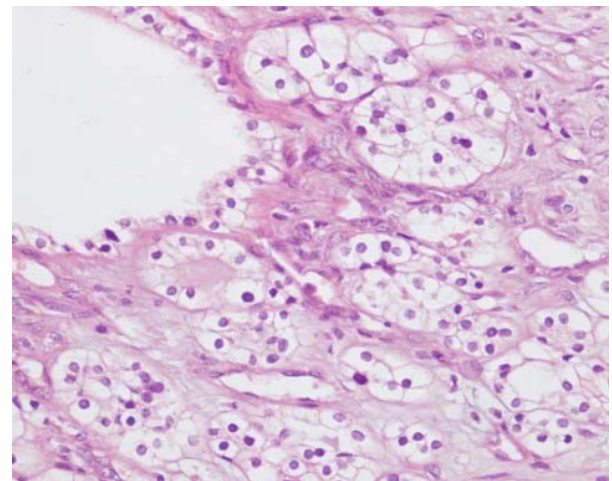
**Figure 2.** Gross photograph of a cut section of the pancreas showing cystic areas alternating with gray white solid and myxoid areas.



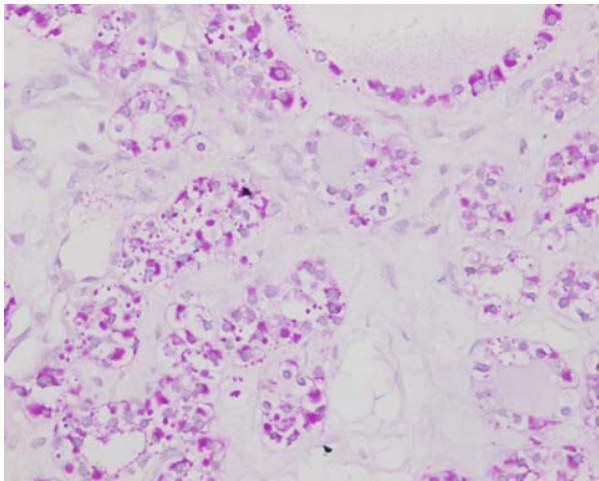
**Figure 3.** Photomicrograph showing solid areas composed of nests of cells arranged in an organoid, trabecular and acinar pattern (H&E, x200).

were filled with hemorrhagic serous fluid. The sections were stained with hematoxylin and eosin (H&E), periodic acid Schiff (PAS) with and without diastase and mucicarmine stain. Immunohistochemistry was performed with chromogranin immunostain.

The sections examined showed a tumor with alternating cystic spaces and solid areas. The cystic spaces were variable and lined with cuboidal to flattened epithelium containing clear to eosinophilic cytoplasm and uniform round nuclei and inconspicuous nucleoli. The solid areas were composed of nests of cells arranged in an organoid, trabecular and acinar pattern (Figure 3). These cells showed the presence of clear to eosinophilic cytoplasm and uniform round nuclei (Figure 4). Numerous congested capillary-sized blood



**Figure 4.** Photomicrograph showing cells with the presence of clear to eosinophilic cytoplasm and uniform round nuclei. (H&E, x400).



**Figure 5.** Photomicrograph showing PAS positive, magenta colored granules in solid areas. (PAS, x200).

vessels were present. Large myxoid areas were also seen. Periodic acid Schiff (PAS) stains with and without diastase digestion revealed the lining of the cells which contained PAS positive, diastase digestible material consistent with glycogen. No mucin-containing cells were identified histochemically. In addition, the tumor showed PAS positive (Figure 5) and chromogranin positive granules (Figure 6) in the solid areas.

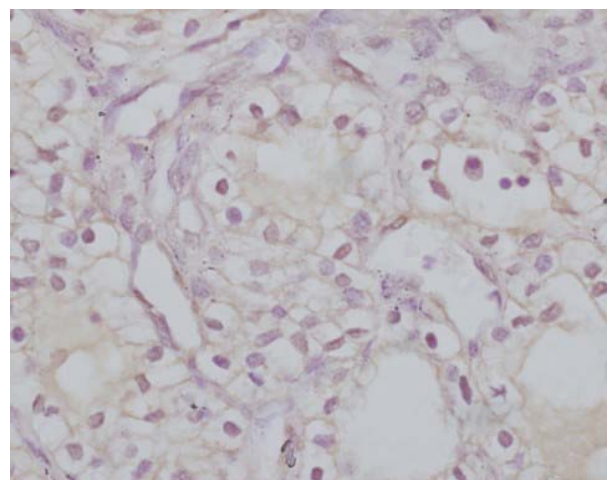
Based on the organoid (endocrine) pattern of the tumor cells in the solid areas on H&E staining and the lining of the cells of the cystic spaces containing glycogen along with chromogranin-positive neurosecretory granules in the solid areas, a final diagnosis of combined microcystic adenoma with pancreatic endocrine neoplasm was made. The patient had an uneventful postoperative course and is well two months after surgery.

## DISCUSSION

In this case report, we have described the rare occurrence of a combined microcystic adenoma and pancreatic endocrine neoplasm in the same patient. There are only five case reports in the English literature describing the coexistence of pancreatic endocrine neoplasms with serous cystadenomas [5].

With regard to the histogenesis of combined microcystic serous cystadenoma and pancreatic endocrine neoplasms, two possibilities exist. Both components of the

neoplasm arose closely but independently and are simply admixed (collision tumor). Alternatively, the tumor was derived from a neoplastic clone capable of exocrine and endocrine differentiation. From the embryological point of view, the biphasic differentiation potential of pancreatic ductal stem cells along either the glandular epithelial or the neuroendocrine pathways would not be unexpected. The presence of endocrine cells within an exocrine carcinoma has also previously been reported in the literature [6]. Although we believe that there can be coincidental occurrence of both tumors (i.e., a collision tumor); in this event, separate tumor epicenters would be expected. Indeed, a few such cases of combined microcystic serous cystadenoma, cystadenocarcinoma and pancreatic endocrine neoplasm have been reported in the literature [4]. In contrast, the endocrine component in a combined neoplasm is embedded within and intermixed with a microcystic serous cystadenoma. Therefore, for the combined neoplasm with the features summarized in this report, we favor the concurrent development and differentiation of distinct epithelial and endocrine cells within the same tumor. In support of this mechanism, biphasic differentiation has been observed in the pancreas in patients with conditions such as nesidioblastosis [7, 8], pancreatoblastoma [9], amphicrine neoplasms [10] and mixed acinar-



**Figure 6.** Photomicrograph showing cytoplasmic chromogranin positive granules in solid areas (chromogranin immunostain, x400).

endocrine neoplasms [11]. Moreover, we observed PAS positive endocrine granules within the epithelial lining of the cystic structures in the vicinity of the pancreatic endocrine neoplasm in the present case. This further supports the development of a combined neoplasm from the same precursor clone.

In addition, this finding indicates that, as in the case of nesidioblastosis, the endocrine components of the neoplasm can be formed by the neogenesis of endocrine cells from transformed duct cells. Since a combined microcystic serous cystadenoma and pancreatic endocrine neoplasm has distinct general and pathologic features, albeit in a small number of cases, we believe that this tumor may represent a separate clinicopathological entity.

A combined microcystic serous cystadenoma and pancreatic endocrine neoplasm may have a higher malignant potential when compared to a microcystic serous cystadenoma. This appears to be related to the pancreatic endocrine neoplasm component, since features suggestive of the malignant potential of the pancreatic endocrine neoplasm component such as perineural and lymphatic invasion or duodenal invasion and lymphatic permeation have been reported in two cases of combined neoplasm; however, no metastatic tumor was seen in either case [12, 13].

In summary, our case emphasizes the importance of the careful gross examination, sampling and reporting of pancreatic tumors. The Coexistence of pancreatic endocrine neoplasms with potential malignant behavior may be overshadowed by obvious benign tumors such as microcystic serous cystadenoma. The malignant potential and prognostic features of this neoplasm require long-term follow-up and additional reports of such cases.

---

Received April 9<sup>th</sup>, 2007 - Accepted May 15<sup>th</sup>, 2007

**Keywords** Carcinoma, Pancreatic Ductal; Cystadenoma, Serous; Diabetes Mellitus;

Duodenum; Endocrine Gland Neoplasms; Nesidioblastosis; Stomach

**Conflict of interest** The authors have no potential conflicts of interest

### Correspondence

Sukant Garg

Department of Pathology

Govt. Medical College, Sector-32A,

Chandigarh 160047

India

Phone: +91-172.266.5375

Fax: +91-172.266.5375

E-mail: sukantgarg@yahoo.com

Document URL: <http://www.joplink.net/prev/200707/14.html>

---

### References

1. Alpert LC, Truong LD, Bossart MI, Spjut HJ. Microcystic adenoma (serous cystadenoma) of the pancreas. A study of 14 cases with immunohistochemical and electron microscopic correlation. *Am J Surg Pathol* 1988; 12:251-63. [PMID 3354751]
2. Compagno J, Oertel JE. Microcystic adenomas of the pancreas (glycogen-rich cystadenomas): a clinicopathologic study of 34 cases. *Am J Clin Pathol* 1978; 69:289-98. [PMID 637043]
3. Keel SB, Zukerberg L, Graeme-Cook F, Compton CC. A pancreatic endocrine tumor arising within a serous cystadenoma. *Am J Surg Pathol* 1996; 20:471-5. [PMID 8604814]
4. Heresbach D, Robert I, Le Berre N, Raoul JL, Siproudhis L, Bretagne JF, et al. Cystic tumors and endocrine tumor of the pancreas. An unusual association. *Gastroenterol Clin Biol* 1993; 17:968-71. [PMID 8125232]
5. Ustün MO, Tuğyan N, Tunakan M. Coexistence of an endocrine tumour in a serous cystadenoma (microcystic adenoma ) of the pancreas, an unusual associaton. *J Clin Pathol* 2000; 53:800-2. [PMID 11064680]
6. Pour P. Islet cells as a component of pancreatic ductal neoplasms. I. Experimental study: ductular cells, including islet cell precursors, as primary progenitor cells of tumors. *Am J Pathol* 1978; 90:295-318. [PMID 341720]
7. Zumkeller W. Nesidioblastosis. *Endocr Relat Cancer* 1999; 6:421-8. [PMID 10516855]
8. Goossens A, Gepts W, Saudubray JM, Bonnefont JP, Nihoul-Fekete , Heitz PU, Klöppel G. Diffuse and focal nesidioblastosis. A clinicopathological study of

24 patients with persistent neonatal hyperinsulinemic hypoglycemia. *Am J Surg Pathol* 1989; 13:766-75. [PMID 2669541]

9. Klimstra DS, Wenig BM, Adair CF, Heffess CS. Pancreatoblastoma. A clinicopathologic study and review of the literature. *Am J Surg Pathol* 1995; 19:1371-89. [PMID 7503360]

10. Klimstra DS, Rosai J, Heffess CS. Mixed acinar-endocrine carcinoma of pancreas. *Am J Surg Pathol* 1994; 18:765-78. [PMID 8037290]

11. Schron DS, Mendelsohn G. Pancreatic carcinoma with duct, endocrine, and acinar differentiation. A histologic, immunocytochemical, and ultrastructural study. *Cancer* 1984; 54:1766-70. [PMID 6089999]

12. Kim YW, Park YK, Lee S, Park JH, Lee SM, Hong SW, et al. Pancreatic endocrine tumor admixed with a diffuse microcystic adenoma. A case report. *J Korean Med Sci* 1997;12:469-72. [PMID 9364309]

13. Jung HK, Son HY, Lee HC, Yi SY. Microcystic adenoma coexistent with low grade malignant islet tumor of the pancreas. *J Clin Gastroenterol* 2001; 11:441-3. [PMID 11319320]