Annals of Medicine and Surgery 60 (2020) 566–570

Contents lists available at ScienceDirect

Annals of Medicine and Surgery

journal homepage: www.elsevier.com/locate/amsu

A case of multiple serous cystadenoma of pancreas

Naoko Sekiguchi, Shinsuke Nakashima, Masahiro Koh, Masami Ueda, Yujiro Tsuda, Tsukasa Tanida, Jin Matsuyama, Masakazu Ikenaga, Terumasa Yamada *

Department of Surgery, Higashiosaka City Medical Center, Nishiiwata 3-4-5, Higashiosaka, Osaka, 567-8588, Japan

ARTICLE INFO

Keywords:
Serous cystadenoma of pancreas
Multiple
Case report

ABSTRACT

Introduction: Typically, SCN is single and doesn't invade around tissue. In our case, tumors were multiple and had gradually grown and caused vein stenosis. This is extremely rare and unique resected multiple SCN case. In addition, I report that it was thought to be educational that even benign tumors could cause such changes.

Presentation of case: A 60-year-old female was diagnosed with 3 multilocular cystic tumors in distal pancreas by contrast enhanced computed tomography (CT) at the preoperative staging for rectal neoplasm. The diameters of cystic tumors were 22/23/29 mm. The CT showed that the tumors had multiple internal septa enhanced in the arterial phase and the second tumor contained internal calcifications located centrally. The main pancreatic duct was not dilated. Although SCN often occurred single and multiple SCN was very rare, we diagnosed that the tumors were suspected microcystic type SCN because they had typical image findings. So, we planned to follow up every six months after resection for rectal neoplasm. 2 years and half later, they had gradually grown, and splenic vein stenosis appeared. The pancreatic parenchyma atrophy and dilatation of the main pancreatic duct had been gradually progressing. We performed distal pancreatectomy because of possibility of malignancy. The histopathological findings showed that 2 cystic tumors the side of pancreatic head had a connection and had typical findings of SCA of pancreas. The other tumor was independent from two tumors. They had no malignant findings.

Discussion: At first, we expected tumor invasion had caused the changes. But tumors had no malignant findings, so we considered that compression from the tumor had caused stenosis, and obstructive pancreatitis had induced the pancreatic parenchyma atrophy.

Conclusion: We learned from this case that not only invasion but also compression caused vein stenosis and pancreatic duct dilation.

1. Introduction

Serous cystic neoplasm (SCN) was proposed in 1978 by Compagno and Hodgkinson [1,2]. SCNs comprise 1–2% of pancreatic neoplasms and 10–29% of pancreatic cystic neoplasms [3–5].They usually occur single, but occasionally multiple. The rate of multiple SCN like this case is very rare. The lesions are usually benign and asymptomatic, and present a benign clinical course in most cases. Surgical resection is recommended only when tumor is large and causing significant symptoms, or when preoperative diagnosis is uncertain in spite of extensive workup. It is also required in the exceptional cases where concern of malignancy exists [3,6].

We experienced a resected case of multiple SCNs, which we couldn't differentiate from local infiltration preoperatively because they were getting large and caused splenic vein stenosis and main pancreatic duct (MPD) dilation. We learned even benign tumors could cause such changes, so we reported our case along with the relevant literature.

This report has been reported in line with the SCARE 2018 criteria [7].

2. Presentation of case

A 60-year-old female was referred to our hospital for detail examination of rectal neoplasm detected by fiber scope. She didn't have any family, drug and medical histories and allergies. On physical examination, there was no remarkable findings. Hyperlipidemia showed only at the first visit, and tumor markers, CEA and CA 19-9, were not elevated.

The contrast-enhanced computed tomography (CT) happened to

* Corresponding author.
E-mail address: nao.rocky0226@gmail.com (T. Yamada).

https://doi.org/10.1016/j.amsu.2020.11.052
Received 23 October 2020; Received in revised form 16 November 2020; Accepted 17 November 2020
Available online 22 November 2020

reveal 3 cystic lesions in pancreatic body and tail, the sizes were 22/23/29mm respectively. The CT showed that the tumors had innumerable internal septa enhanced in the arterial phase (Fig. 1A), and the second tumor contained internal calcifications located centrally (Fig. 1B). Main pancreatic duct (MPD) was not dilated. We assessed the tumors of the pancreas had a low potential for malignancy. By contrast, the rectal neoplasm had been diagnosed rectal advanced cancer (Stage IIA), so she was performed laparoscopic sigmoidectomy before detail examination for pancreatic tumors. The course after the operation was uneventful. One month after the operation, she was performed magnetic resonance imaging (MRI), endoscopic retrograde cholangiopancreatography (ERCP) with scraping cytology for pancreatic neoplasms, and fluorodeoxyglucose-positron emission tomography (FDG-PET). In MRI, the tumors were low intensity in T1 weighted image (T1WI) and high intensity in T2WI. MRI demonstrated septa and gathering microcysts inside the tumors, so called honeycomb appearance (Fig. 1C/D). ERCP showed the tumors didn't communicate MPD. There were no abnormal uptakes on FDG-PET. The result of the cytology was no malignancy. Because the tumor consisted of the glycogen rich cuboidal cells, we diagnosed the tumors were multiple serous cystadenomas and considered lymphangioma and IPMN as differential diagnosis. Then, we selected not surgical procedure but follow up every six months by image findings.

After 2 and a half years, the tumor sizes increased gradually (31/33/33mm), and splenic vein stenosis appeared (Fig. 2A). The pancreatic parenchyma atrophy and dilatation of the MPD had been gradually progressing (Fig. 2B/C/D). We could not deny malignant transformation and infiltration, so we decided to perform distal pancreatectomy. The operation was performed by specialists of pancreatic resection who had been in practice for over 10 years. In operation findings, there were 3 cystic lesions, which sizes were 35/35/50mm respectively. The tumor surfaces were smooth, and cysts contained serous fluid. The pathological findings revealed that they consisted of multiple spongy microcysts, and the cells which had small nucleus and clear cytoplasm (Fig. 3). The three cysts had similar pathological findings and 2 lesions from the pancreatic head had the connection with each other. The stenosis of the splenic vein was caused by not invasion of malignancy, but compression of cystic lesions. We finally diagnosed them multiple serous cystadenomas. The

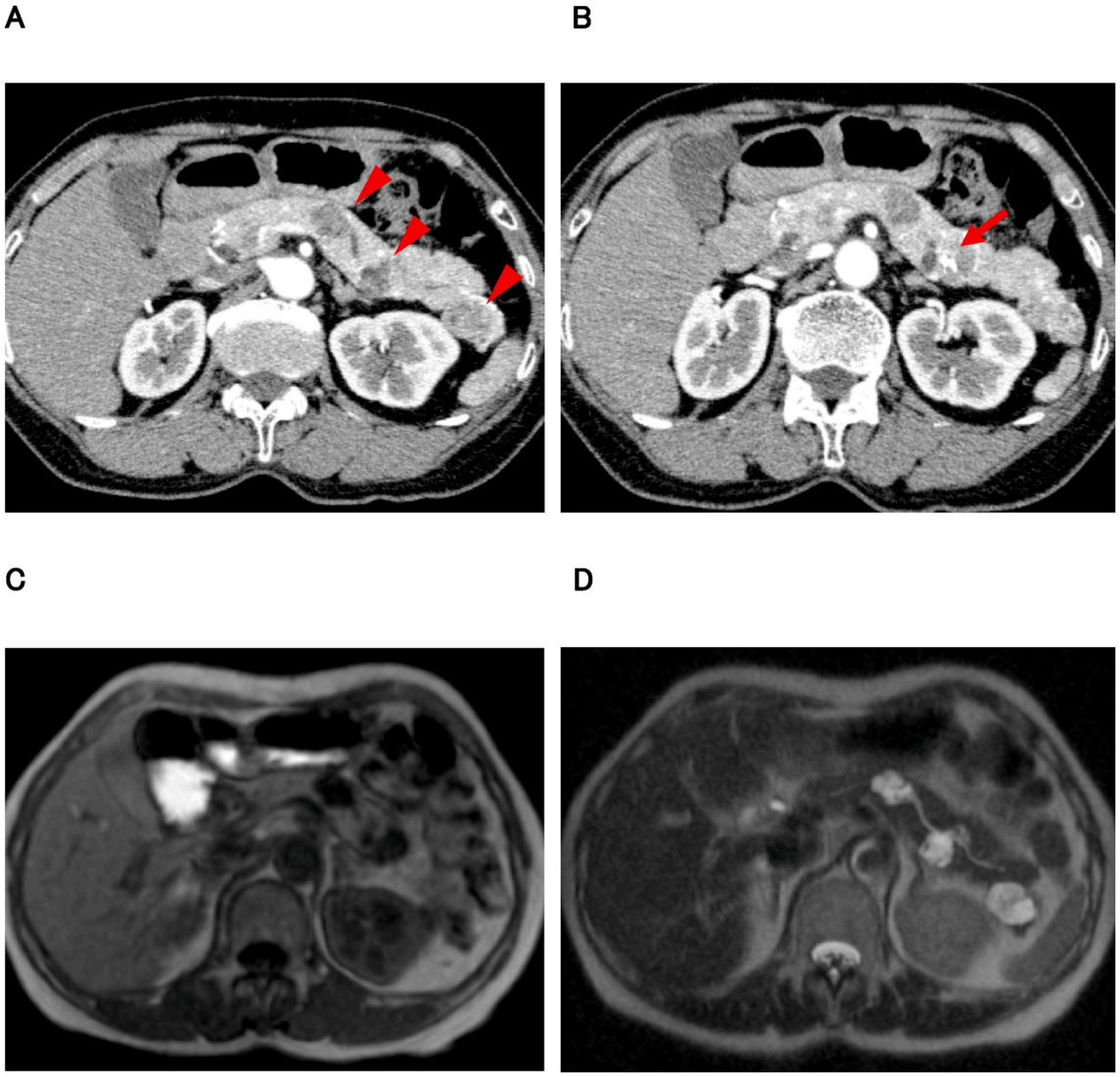

Fig. 1. A/B; CT
The contrast-enhanced CT revealed 3 cystic lesions in pancreatic body and tail, the sizes were 22/23/29mm respectively (red arrow heads). The second tumor contained internal calcifications located centrally(red arrows).
C; MRI T1WI/D; MRI T2WI
The tumors were low intensity in T1WI and high intensity in T2WI. MRI demonstrated septa and gathering microcysts inside the tumors. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

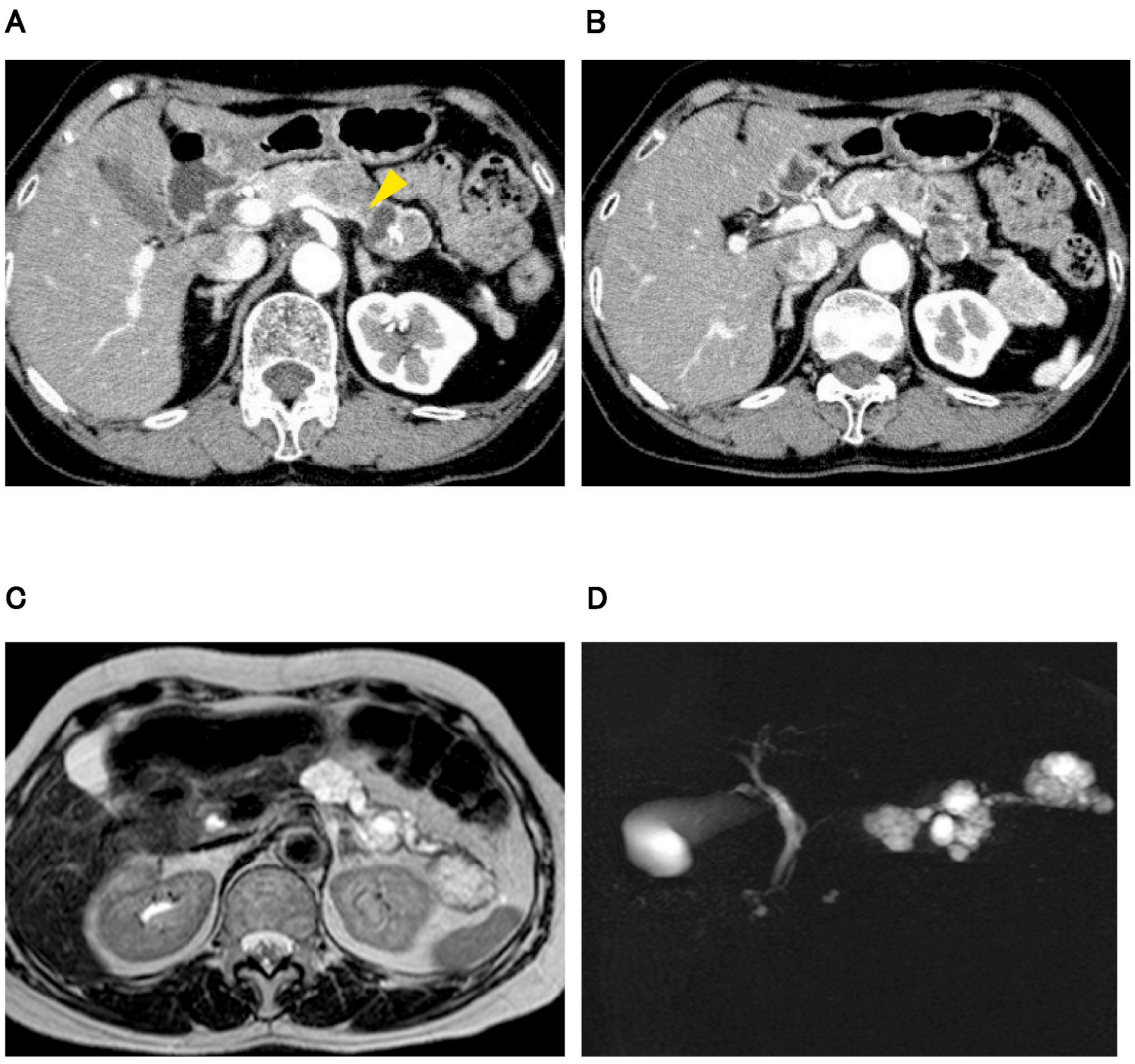

Fig. 2. A/B; CT after 2 and a half year
The tumor sizes increased gradually (31/33/33mm), and splenic vein stenosis appeared (yellow arrow head). The pancreatic parenchyma atrophy had been gradually progressing
C/D; MRI after 2 and a half year. The dilatation of the MPD had been progressing. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

course after the operation was uneventful. She discharged to her home on the 13 day after operation, and was followed up every 6 month by image findings, CT and MRI. She had lived for 10 years after surgery without any evidence of metastasis and recurrence.

3. Discussion

Pancreatic cystic lesions have several species. Ones are benign cysts without any malignant potential such as pancreatic pseudocysts and serous cystic neoplasms (SCNs). Others are premalignant cysts such as mucinous cystic neoplasms (MCNs) and intraductal papillary mucinous neoplasms (IPMNs). Among them, SCNs comprise 1–2% of pancreatic neoplasms and 10–29% of pancreatic cystic neoplasms [3–5]. They are more frequently detected in women, between the sixth and seventh decade [8].They usually occur single, but occasionally multiple. The rate of multiple SCNs like this case is 2.3% [9].

SCNs have 4 types, microcystic type, macrocystic type, mixed type and solid type [9,10]. Most SCNs are exclusively microcystic type that has become easier to diagnose as microcystic type SCN with advances in imaging. Microcystic type SCN has typical image findings so called honeycomb appearance. The diagnostic accuracy of honeycomb appearance is 100% in EUS, 85% in CT, 89% in abdominal ultrasound sonography, and 86% in MRI [8]. In this case, we were able to diagnose as SCN preoperatively because MRI showed thin septa and gathering microcysts inside the tumors.

The surgical resection for SCNs is recommended only when the lesions are symptomatic, difficult to definitively differentiate from other surgical lesions, or large in size [3,6]. In this case, firstly, we chose non-surgical procedure because the tumors didn't indicate the criteria for surgery. We have decided to follow images up every six months. About 2 and a half years later, the tumor sizes increased gradually. SCN shows little increase in tumor size, or only a slight increase, if any. El-Hayek KM et al. estimated an overall mean growth rate of SCN was 2.8 mm per year [11].Jais B et al. analyzed the growth rate was different from the original tumor size [10].This case showed a slow growth reflecting the nature of SCN but the dilatation of the MPD and stenosis of splenic vein had been progressing. By the reason of findings, the tumors were suspected to have invaded the surrounding tissue. Therefore, we performed surgery. The pathological findings revealed that the dilatation of the MPD was caused by the compression of them and they were diagnosed serous cystadenomas. We considered that obstructive pancreatitis induce the pancreatic parenchyma atrophy. This is unique

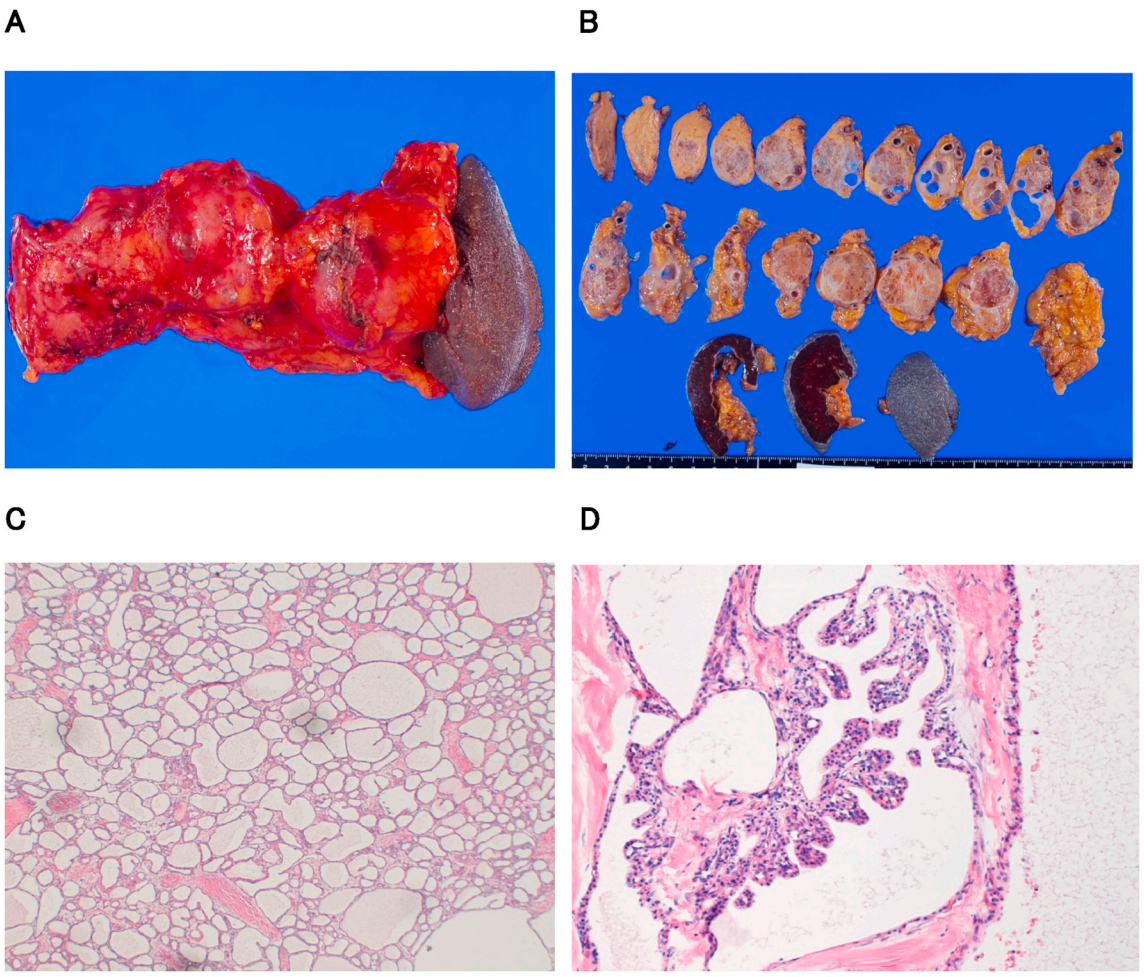

Fig. 3. A/B; Resected specimen
The tumor surfaces were smooth, and the tumor consisted of multiple spongy microcysts.
C/D; HE stain
The tumor had multiple microcysts. Each tumor had similar pathological finding. They were consisted the cells which had small nucleus and clear cytoplasm. A part of microcysts had papillary projection.

and educational that even benign tumors could cause such changes.

Almost SCNs are considered benign tumors without malignant potential. The possibility of malignant transformation in SCNs is 0–2.2% [9,12,13].Since this risk is unremarkable, surgical resection is advised only in symptomatic patients [3]. In the Japanese classification of pancreatic carcinoma 7th edition, only cases with metastases such as liver metastases are defined as serous cystadenocarcinoma [14]. However, Galanis C et al. [13]reported three of 158 patients were noted in final pathology demonstrated locally aggressive disease on histologic examination. By definition, these cases were diagnosed as SCA. But one recurred 13 years later, with disease in the liver and retroperitoneal tissue and was therefore diagnosed with serous cystadenocarcinoma. The preoperative differentiation between serous cystadenoma and serous cystadenocarcinoma remains difficult. Most of the cases followed a benign course, but some cases died of the primary disease. In addition, Nodell CG et al. reported one case of SCA in which the dilation and stenosis of MPD were caused due to coexistence of pancreatic cancer [15]. Therefore, surgery is considered if the MPD changes appear or we can't deny the existence of malignancy.

4. Conclusion

We experienced a rare case of multiple SCNs. We should keep in mind that even small SCNs can cause the stenosis of MPD and atrophy of pancreatic parenchyma by the compression.

Funding

No sources of funding.

Author contribution

Clinical treatment: Naoko Sekiguchi, Shinsuke Nakashima, Masahiro Koh, Masami Ueda, Yujiro Tsuda, Tsukasa Tanida, Jin Matsuyama, Masakazu Ikenaga and Terumasa Yamada.

Collected data: Naoko Sekiguchi, Shinsuke Nakashima.

Assessment and discussion: Naoko Sekiguchi, Shinsuke Nakashima and Terumasa Yamada.

Wrote the paper: Naoko Sekiguchi, Shinsuke Nakashima and Terumasa Yamada.

Registration of research studies

We confirm that the work is not necessary to registry. The report of one case is not necessary to ethnical approval in our institution.

Guarantor

The guarantor of this study is Terumasa Yamada, corresponding author.

Consent

Informed consent has been obtained from the patient and all identifying details have been omitted. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

This study of case report is exempt from ethnical approval in ethics committee of our institution.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Declaration of competing interest

No conflicts of interest.

Appendix A. Supplementary data

Supplementary data related to this article can be found at https://doi.org/10.1016/j.amsu.2020.11.052.

Abbreviations

CT	Computed tomography
IPMN	Intraductal papillary mucinous neoplasm
MCN	Mucinous cystic neoplasm
MRI	Magnetic resonance imaging
T1WI	T1 weighted image
T2WI	T2 weighted image

References

[1] J. Compagno, J.E. Oertel, Microcystic adenomas of pancreas (glycogen-rich cystadenomas): a clinicopathologic study of 34 cases, Am. J. Clin. Pathol. 69 (1978) 289–298.
[2] D.J. Hodgkinson, W.H. ReMine, L.H. Weiland, Pancreatic cystadenoma. A clinicopathologic study of 45 cases, Arch. Surg. 113 (1978) 512–519.
[3] Terris B, Fukushima N, Serous Neoplasms of the Pancreas.
[4] Hruban R, Pitmen M. Serous Cystic Neoplasms.
[5] Colonna J, Plaza JA. Serous cystadenoma of the pancreas clinical and pathological feature in 33 patients.
[6] J.F. Tseng, A.L. Warshaw, Serous cystadenoma of the pancreas tumor growth rates and recommendations for treatment, Ann. Surg. 242 (2005) 413–421.
[7] R.A. Agha, M.R. Borrelli, R. Farwana, et al., The SCARE 2018 statement: updating consensus surgical CAse REport (SCARE) guidelines, Int. J. Surg. 60 (2018) 132–136.
[8] O. Strobel, Z'graggem. Risk of malignancy in serous cystic neoplasms of the pancreas, Digenst 68 (2003) 24–33.
[9] W. Kimura, T. Moriya, Multicenter study of serous cystic neoplasms of the Japan pancreas society, Pancreas 41 (2012) 380–387.
[10] B. Jais, V. Rebours, G. Malleo, et al., Serous cystic neoplasms of the pancreas: a multinational study of the International Association of pancreatology and European pancreatic club, Gut 65 (2016) 305–312.
[11] K.M. El-Hayek, N. Brown, C. O'Rourke, et al., Rate of growth of pancreatic serous cystadenoma as an indication for resection, Surgery 154 (2013) 794–802.
[12] J.C. King, T.T. Ng, S.C. White, et al., Pancreatic serous cystadenocarcinoma: a case report and review of literature, J. Gastrointest. Surg. 13 (2009) 1864–1868.
[13] C. Galanis, A. Zamani, J.L. Cameron, et al., Resected serous cystic neoplasms of the pancreas: a review of 158 patients with recommendations for treatment, J. Gastrointest. Surg. 11 (2007) 820–826.
[14]) General Rules for the Study of Pancreatic Cancer, seventh ed., Japan pancreas society, 2016.
[15] C.G. Nodell, P.C. Freeny, D.H. Dale, J.A. Ryan, Serous cystadenoma of the pancreas with a metachronous adenocarcinoma, AJR Am. J. Roentgenol. 162 (1994) 1352–1354.