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## Wirsungocele: evaluation by MRCP and clinical significance

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### Abstract

**Purpose**—Wirsungocele is a rare cystic dilatation of the main pancreatic duct seen at the terminal portion of the duct of Wirsung. The purpose of our study is to evaluate the diagnostic value of MRCP in detection of Wirsungocele and the association between the MRCP-determined size of Wirsungocele and the MRCP-clinical findings of pancreatitis.

**Methods**—Thirty-four patients with reported ‘Wirsungocele’ were analyzed in the study. Two radiologists reviewed MRCP/S-MRCP images for the presence and diameter of Wirsungocele (WD), main pancreatic duct dilatation (MPDD), side branch ectasia (SBE), acinarization, and duodenal filling grade. Electronic medical record review included symptoms (abdominal pain), signs (recurrent acute/chronic pancreatitis), and select laboratory testing (serum amylase and lipase). Inter-reader agreement values were calculated by ICC. Pearson correlation analysis was performed to evaluate the association of WD with radiological and clinical findings. The comparison of WD on MRCP versus S-MRCP was calculated by Wilcoxon test. Mann–Whitney *U* test was used for two independent variable comparisons.

**Results**—The sensitivity of MRCP for the detection of Wirsungocele calculated using the S-MRCP and ERCP as the reference method was 76.9% and 100%, respectively. There was a significant difference in the diameter of Wirsungocele measured by MRCP vs S-MRCP ( $p < 0.001$ ). There was good inter-reader agreement for the detection of Wirsungocele on MRCP and measurement of WD on MRCP and S-MRCP (ICC: 0.79, 0.89, and 0.80, respectively,  $p < 0.001$ ). There was a significant difference in WD between the patients with and without MPDD ( $p < 0.05$ ). There was a significant positive correlation between WD and MPDD ( $r = 0.66$ ,  $p < 0.05$ ). WD was significantly associated with recurrent acute pancreatitis ( $p < 0.05$ ).

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**Ethical approval** Institutional Review Board approval was obtained.

**Informed consent** Written informed consent was waived by the Institutional Review Board.

**Conclusion**—MRCP is a highly sensitive and non-invasive imaging tool for detection of Wirsungocele. Greater Wirsungocele diameter is associated with MPDD and recurrent acute pancreatitis.

### Keywords

Magnetic resonance imaging; Pancreatic duct; Pancreatitis; Wirsungocele

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## Introduction

Wirsungocele is an uncommon cystic dilatation of the terminal portion of the ventral pancreatic duct just upstream to the major papilla [1]. Santorinicele is more common and generally accompanied by complete or partial pancreas divisum [2, 3]. Santoriniceles may result from increased intraductal pressure, impediment of pancreatic flow, and/or congestion of pancreatic juice [1, 2, 4]. Another proposed mechanism is decreased autonomic innervation of the sphincter of Oddi, causing functional obstruction of the papilla [5]. There are only a few case reports of Wirsungocele in the literature associated with acute pancreatitis, chronic abdominal pain, and chronic asymptomatic pancreatic hyperenzymemia. However, the mechanism of these clinical findings has not been confirmed, and it is unclear whether Wirsungocele is among the causes of recurrent acute pancreatitis [6–10].

Evaluation of pancreatic ductal anatomy is required in patients with recurrent acute pancreatitis and chronic pancreatitis. MRCP, ERCP, and EUS are the imaging methods used for this purpose [11]. MRCP with use of IV synthetic secretin significantly improves visualization of the main pancreatic duct, abnormal side branches, and ductal anomalies such as pancreas divisum and Wirsungocele. Secretin is a hormone secreted by the duodenum in response to gastric acid which stimulates the secretion of water and bicarbonate by the exocrine pancreatic cells. This leads to increased fluid in the pancreatic duct if the pancreatic secretory capacity is adequate [12–15]. Secretin-enhanced MRCP (S-MRCP) enables evaluation of pancreatic parenchyma, ductal anatomy, and function in a single modality and used for evaluation of ductal diseases and exocrine reserve in patients with chronic pancreatitis [16, 17].

The purpose of our study is to evaluate the diagnostic value of MRCP in detection of Wirsungocele and the association between the size of Wirsungocele and imaging and clinical findings of pancreatitis. To our knowledge, this is the first study to investigate the diagnostic value of MRCP for this purpose.

## Methods

### Patients

This HIPAA-compliant retrospective study was approved by the Institutional Review Board (IRB) with a waiver of consent. The MRI database of 6892 patients who underwent MRCP or S-MRCP between 2008 and 2020 was reviewed for the word ‘Wirsungocele.’ Thirty-four MRCP/S-MRCP were detected and analyzed. Post-secretin MRCP images were obtained in

26 and ERCP was performed in 16 of these 34 patients. ERCP findings performed after MRCP within 3 months were collected. ERCP images obtained by the endoscopists were also reevaluated by the radiologist. The indications of ERCP and MRCP were abdominal pain, hyperenzymemia, pancreatitis, and the evaluation of pancreatic ductal anatomy. Parameters assessed via electronic medical record review included symptoms (abdominal pain), signs (recurrent acute/chronic pancreatitis), and select laboratory testing (serum amylase and lipase) at the time of MRCP. Hyperenzymemia was defined as the serum amylase and/or lipase levels above the normal upper reference limit [18]. Recurrent acute pancreatitis cases were defined as cases who had more than one episode of acute pancreatitis with relief of symptoms between acute episodes [19].

### Imaging technique

Patients fasted for at least 4 h prior to the MR examination. MRCP was performed on either a 1.5 Tesla or a 3.0 Tesla scanner (Magnetom Avanto Harmony, or Verio, Siemens Medical Solutions, Erlangen, Germany). S-MRCP images were obtained after intravenous injection of 0.2 mcg/kg secretin over 1 min. Coronal 2D single-shot turbo spin-echo sequence (HASTE, Siemens Medical Solutions, Malvern, PA) was repeated every 30 s for 10 min.

### Image analysis

Two abdominal radiologists with 4 and 5 years of experience, blinded to clinical information, evaluated the pre- and post-secretin MRCP images separately. Wirsungocele diameter (WD) was measured both on pre- and post-secretin images. Images were assessed for concomitant pancreas divisum and Santorinicele, main pancreatic duct dilatation (MPDD), side branch ectasia (SBE), acinarization, and duodenal filling grade. MPDD was defined as a duct caliber larger than 3.5 mm on MRCP [20]. Side branch ectasia (SBE) was scored in concordance with the Cambridge classification used in chronic pancreatitis as follows: 0: no side branch ectasia, 1: fewer than three, 2: three or more. Acinarization was determined by the progressive increase in signal intensity of the pancreatic parenchyma on either side of the pancreatic duct on S-MRCP. The exocrine secretory capacity was assessed by the grade of duodenal filling on S-MRCP images, where 0: no visible filling, 1: filling only in the duodenal bulb, 2: filling also in descending duodenum up to the genu inferius, 3: filling in the entire duodenum beyond the genu inferius [21, 22].

### Statistical analysis

Statistical analyses were performed using SPSS (IBM Inc., Chicago, IL, USA) with version 24.0. Descriptive statistics were presented as frequencies [percentage] for categorical variables, and as mean  $\pm$  SD for numerical variables. Sensitivity values with 95% confidence interval (CI) were calculated for the detection of Wirsungocele on MRCP. Continuous variables were analyzed for normality by the Kolmogorov–Smirnov and Shapiro–Wilk test. Since the distribution of the measurements of WD on MRCP was normal, Pearson correlation test was used for the correlation analysis. The comparison of WD on MRCP vs S-MRCP was calculated by Wilcoxon test. Mann–Whitney *U* test was used for two independent variable, whereas Kruskal–Wallis test was used for  $> 2$  independent variable comparisons, since  $n < 30$  in each group. Inter-reader agreement values for detection of Wirsungocele by MRCP and WD measurements were calculated by intraclass correlation

coefficient (ICC). A  $p$  value of less than 0.05 ( $p < 0.05$ ) was considered statistically significant by taking 5% for type-I error.

## Results

The mean age of the patients was 64 (23–80). The mean age of female patients ( $n = 25$ ) was 65 (31–80), and the mean age of male patients ( $n = 9$ ) was 61 (23–77). There was no significant difference in the mean age between genders ( $p = 0.56$ ).

The sensitivity (95% CI) of MRCP for the detection of Wirsungocele calculated using ERCP as the reference method was 100% (29.24–100%). The sensitivity (95% CI) of MRCP calculated using S-MRCP as the reference method was 76.9% (56.35–91.03%) (Table 1).

The mean diameter of Wirsungocele on MRCP and S-MRCP was  $4.12 \pm 1.90$  mm and  $5.40 \pm 1.95$  mm, respectively. There was a significant difference in the diameter of Wirsungocele before and after secretin enhancement ( $p < 0.001$ ) (Fig. 1). There was no significant difference in the mean diameter of Wirsungocele between genders (male: 5.19, female: 4.23,  $p = 0.19$ ). There was no significant correlation between the diameter of Wirsungocele and age ( $p = 0.07$ ). Inter-reader agreement was calculated for the detection of Wirsungocele on MRCP and measurement of WD on MRCP and S-MRCP. The ICC values were good, calculated as 0.79, 0.89, and 0.80, respectively ( $p < 0.001$ ) (Table 2). The mean WD measured on MRCP of patients with MPDD was  $3.98 \pm 1.46$  mm, and the mean WD of patients without MPDD was  $7.40 \pm 1.13$  mm. The WD measured on MRCP was significantly larger in the group with MPDD than the group without ( $p = 0.001$ ).

All of the S-MRCP images exhibited grade 3 duodenal filling. There was no significant impediment of duodenal filling in the Wirsungocele patients. Eight cases showed acinarization. Fifteen patients had none, 10 had fewer than three, and 5 had three or more side branch ectasia on S-MRCP images. According to the calculations by the Mann–Whitney  $U$  test, there was no significant difference in the WD between the groups with and without acinarization ( $p = 0.50$ ). According to the calculations by using the Kruskal–Wallis test, there was no significant difference in the diameter of Wirsungocele between the side branch ectasia groups ( $p = 0.90$ ). Correlation analysis was performed to assess the correlation between WD measured on MRCP and MPDD, side branch ectasia, and acinarization. There was a significant positive association between WD and MPDD ( $r = 0.66$ ,  $p < 0.05$ ) (Fig. 2). There was no significant correlation with the other variables (Table 3). In addition, results of the Pearson correlation indicated that there was a significant positive association between side branch ectasia and acinarization ( $r = 0.74$ ,  $p < 0.001$ ).

The mean of WD in the group without recurrent acute pancreatitis was  $4.09 \pm 1.70$  mm, whereas it was  $5.75 \pm 1.90$  mm in the group with recurrent acute pancreatitis. There was a significant difference in WD measured on MRCP between the patients with and without recurrent acute pancreatitis ( $p = 0.03$ ). There was no significant difference between the patients with and without abdominal pain, chronic pancreatitis, or hyperenzymemia ( $p > 0.05$ ) (Table 4).

## Discussion

In this study, we demonstrate that Wirsungocele has important clinical implications and MRCP is helpful in the diagnosis. This is the largest series of Wirsungocele evaluated by MRCP.

MRCP was reported to be highly sensitive and specific in the evaluation of pancreatic ductal anatomy in the previous studies [23, 24]. Therefore, we evaluated the diagnostic value of MRCP for the detection of Wirsungocele, using S-MRCP and ERCP as the reference methods and found high sensitivity rates. S-MRCP detected Wirsungoceles more frequently than did ERCP, which fits with findings from a previous study, which demonstrated that ERCP without secretin stimulation may underestimate the rate of Santorinicele and make ERCP less successful than MRCP in the detection of Santorinicele [1]. The difference in detection rates of Wirsungocele by ERCP and S-MRCP in our study may have resulted from the use of secretin in S-MRCP but not in ERCP or because the endoscopists were not focused on Wirsungocele while obtaining the images.

Secretin causes an increase in the amount of fluid in the pancreatic duct. Therefore, IV synthetic secretin administration during MRCP exam improves the visualization and evaluation of the pancreatic ductal anomalies and classification of chronic pancreatitis [12–17]. Likewise, in our study the mean WD measured on S-MRCP was larger than the mean WD measured on MRCP. This may improve the diagnostic confidence for detection of Wirsungocele.

There was a significant positive correlation between WD and MPDD. We think that the increase in the size of Wirsungocele may cause more congestion of the pancreatic juice in the duct and result in pancreatic duct dilatation.

Secretin is also used for the evaluation of the pancreatic secretory capacity. Duodenal filling beyond the genu inferior within 5–10 min is accepted as sufficient capacity [13]. Acinarization is the term used for defining the progressive increase in signal intensity of pancreas parenchyma on either side of the pancreatic duct on post-secretin images. It is assumed to occur because of increased pancreatic ductal pressure with adequate secretory capacity, so an insensitive but a specific finding of early chronic pancreatitis [25, 26]. We had 9 patients who showed the finding of acinarization. We did not observe a significant difference in the diameter of Wirsungocele between the acinarization groups. Also, all of the S-MRCP images exhibited grade 3 duodenal filling in our study. Therefore, we think that the Wirsungoceles in our study had not caused a significant impediment of papillary drainage. Acinarization showed a significant correlation only with the side branch ectasia, which are both signs of chronic pancreatitis.

Pancreas divisum and santorinicele are associated with chronic asymptomatic hyperenzymemia, chronic and recurrent acute pancreatitis. The mechanism may be the congestion of pancreatic juice because of the relatively decreased drainage through the smaller duct and papilla [3, 4, 8, 27]. Sphincterotomy at the minor papilla may be performed in these patients to prevent recurrent acute pancreatitis [28, 29]. There are a few case reports of Wirsungocele in the literature associated with acute pancreatitis, chronic abdominal pain,

and chronic asymptomatic pancreatic hyperenzymemia. However, the mechanism of these clinical findings has not been confirmed. It is still unclear whether Wirsungocele is among the causes of recurrent acute pancreatitis [6–10]. We were interested to find whether the increase in the size of Wirsungocele is associated with recurrent acute pancreatitis, chronic pancreatitis, or abdominal pain. Only groups with and without recurrent acute pancreatitis showed a significant difference in the diameter of Wirsungocele. We observed that patients with recurrent acute pancreatitis had larger Wirsungocele. It is possible that the size of Wirsungocele may also be a factor for the recurrence of acute pancreatitis. Further studies would be helpful to investigate this observation.

The limitations of our study were S-MRCP and ERCP was not performed for all of the patients and endoscopists were not focused to depict Wirsungocele for a study while obtaining the images.

In conclusion, MRCP is a highly sensitive and non-invasive imaging tool for detection of Wirsungocele. Secretin causes increase in the WD which may improve detection. Increase in the size of Wirsungocele is associated with MPDD which may be caused by the congestion of pancreatic juice in the duct. Greater Wirsungocele diameter is associated with recurrent acute pancreatitis.

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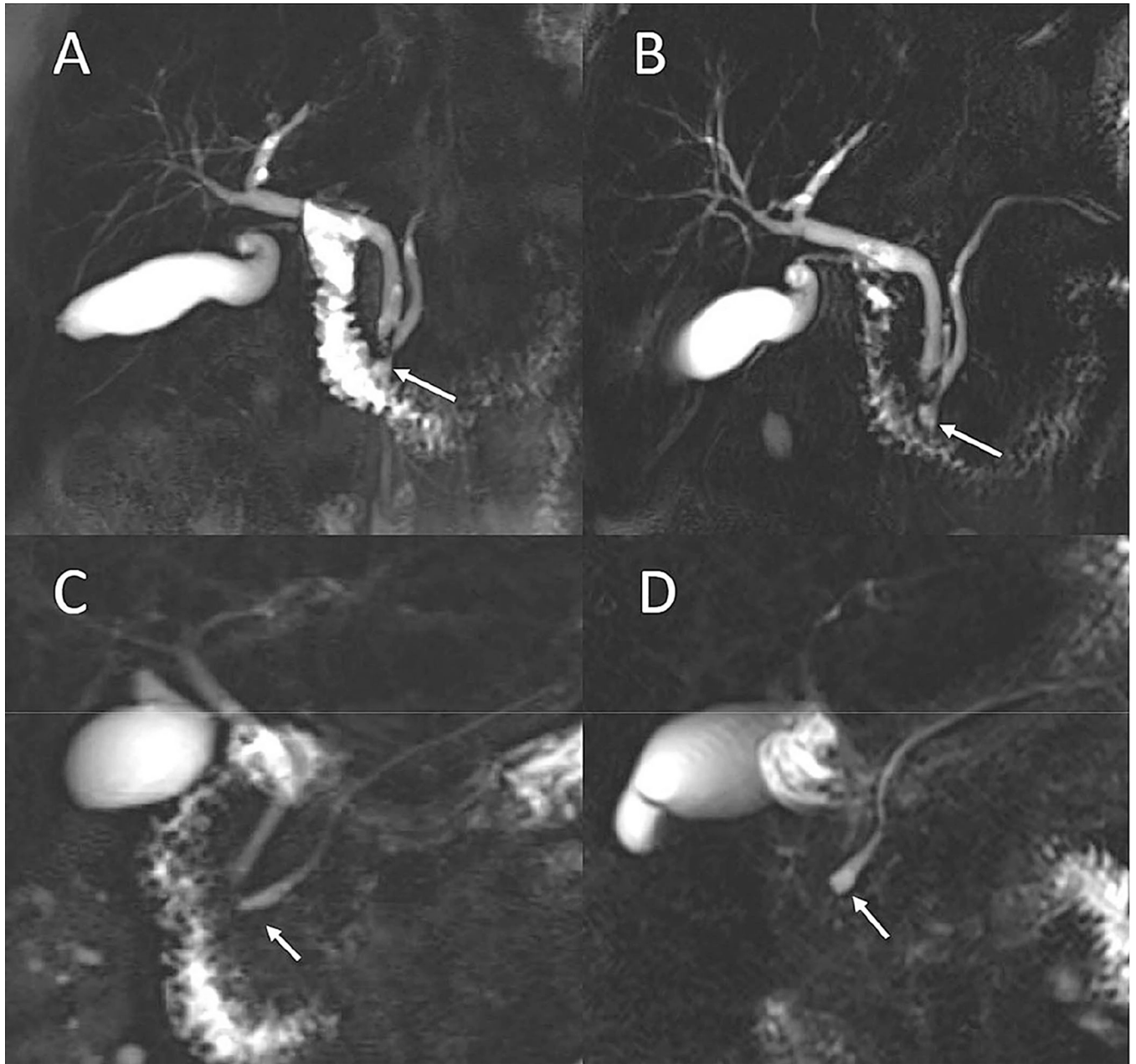
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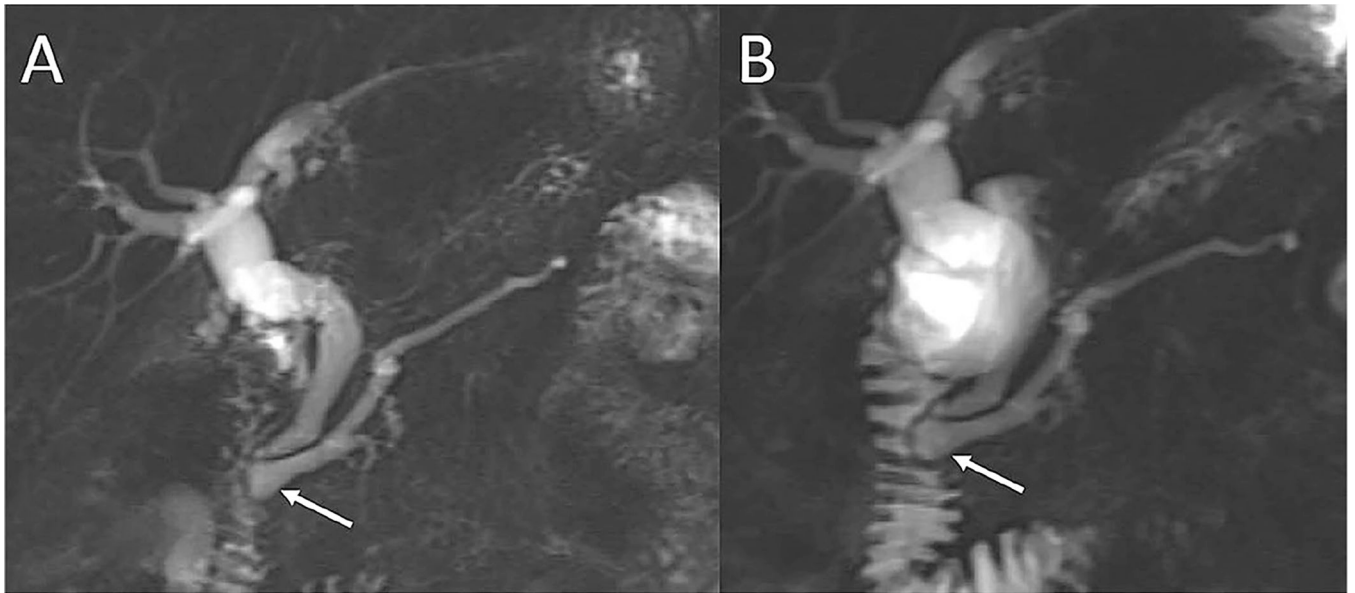
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**Fig. 1.** Wirsungocoele (arrow) was seen on pre-secretin MRCP image (a). There was mild increase in the diameter of the Wirsungocoele (arrow) on the post-secretin MRCP (b). An indeterminate case on pre-secretin image (c) gets cystic appearance (arrow) with significant increase in size after secretin enhancement (d)





**Fig. 2.** Wirsungocele (arrow) concurrent with main pancreatic duct dilatation was seen on pre- (a) and post- (b) secretin MRCP images

**Table 1**

The detection rates of Wirsungocele by MRCP, S-MRCP, and ERCP

Wirsungocele	S-MRCP			ERCP		
	Negative	Positive	Total	Negative	Positive	Total
MRCP						
Negative	0	6	6	3	0	3
Positive	0	20	20	10	3	13
Total	0	26	26	13	3	16
Sensitivity (95% CI)	76.9% (56.35–91.03%)			100% (29.24–100%)		

*S-MRCP* secretin-enhanced MRCP, *CI* confidence interval

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**Table 2**

Inter-reader agreement analysis for the detection of Wirsungocele on MRCP, diameter of Wirsungocele on MRCP and S-MRCP

	ICC	95% CI	<i>p</i> *
MRCP-W	0.79	0.58–0.89	< 0.001
MRCP-WD	0.89	0.74–0.95	< 0.001
S-MRCP-WD	0.80	0.54–0.91	< 0.001

*MRCP-W* detection of Wirsungocele on MRCP, *MRCP-WD* diameter of Wirsungocele on MRCP, *S-MRCP-WD* diameter of Wirsungocele on S-MRCP, *CI* Confidence Interval

\* Significant at the 0.01 level

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**Table 3**

The correlation analysis of diameter of Wirsungocele, main pancreatic duct dilatation, side branch ectasia, and acinarization

Pearson correlation	MPDD	SBE	Acinarization
WD ( <i>N</i> = 34)			
Correlation coefficient	0.66*	0.125	0.169
Sig.(2-tailed)	0.000	0.509	0.410
<i>N</i>	34	30	26

WD diameter of Wirsungocele, MPDD main pancreatic duct dilatation, SBE side branch ectasia

\*Correlation is significant at the 0.01 level (2-tailed)

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**Table 4**

Statistical analysis of the association between diameter of Wirsungocele and clinical-laboratory findings

<b>WD</b>	<b>N</b>	<b>Mean ± SD</b>	<b><i>p</i><sup>a</sup></b>
RAP			
Negative	26	4.09 ± 1.70	0.03*
Positive	8	5.75 ± 1.90	
CP			
Negative	25	4.47 ± 1.67	0.82
Positive	9	4.51 ± 2.45	
Hyperenzymemia			
Negative	12	4.53 ± 1.95	0.40
Positive	8	5.15 ± 1.76	
Abdominal pain			
Negative	30	4.66 ± 1.88	0.11
Positive	4	3.18 ± 1.20	

WD diameter of Wirsungocele, RAP recurrent acute pancreatitis, CP chronic pancreatitis

<sup>a</sup>Mann–Whitney *U* test

\*Significant at the 0.05 level