

## Recurrent Acute Pancreatitis as A Manifestation of Sphincter of Oddi Dysfunction

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### **ABSTRACT**

*Recurrent acute pancreatitis (RAP) is defined as two or more occurrence of acute pancreatitis with no evidence of underlying chronic pancreatitis. Prevalence of RAP varied from 10-30%. One of the postulated mechanism of this condition is sphincter of Oddi dysfunction (SOD) which is a clinical biliary pain syndrome or acute pancreatitis (AP) due to pancreatobiliary obstruction at the level of sphincter of Oddi. We reported a 29-year-old female patient who came to Cipto Mangunkusumo Hospital regarding upper quadrant abdominal pain with previously well documented history of AP in the last six months before admission. Laboratory findings showed elevated pancreatic enzyme level which was consistent with AP. The patient underwent magnetic resonance cholangio-pancreatography (MRCP) and endoscopic ultrasound (EUS) examination and both of the results showed dilatation of pancreatic duct which suggested SOD. Due to the lack of further diagnostic modality, manometry was not performed on this patient. However, after excluding other possible etiology of SOD, the patient underwent endoscopic retrograde cholangio-pancreatography (ERCP) and sphincterotomy was performed. The signs and symptoms of AP was relieved after sphincterotomy and not yet recurred.*

**Keywords:** *recurrent acute pancreatitis, sphincter of Oddi dysfunction (SOD), endoscopic retrograde cholangio-pancreatography (ERCP), manometry, abdominal pain*

### **ABSTRAK**

*Pankreatitis akut rekuren didefinisikan sebagai 2 atau lebih kejadian pankreatitis akut tanpa bukti adanya pankreatitis kronik yang mendasari. Prevalensi pankreatitis akut rekuren berkisar antara 10-30%. Salah satu mekanisme yang mendasari keadaan ini adalah disfungsi sfingter Oddi yang merupakan sindrom klinik nyeri bilier atau pankreatitis akut karena adanya obstruksi pankreatobilier setinggi sfingter Oddi. Kami melaporkan perempuan usia 29 tahun yang datang ke rumah sakit (RS) Cipto Mangunkusumo dengan keluhan utama nyeri perut kanan atas dan riwayat pankreatitis akut berulang dalam 6 bulan terakhir sebelum masuk RS. Pemeriksaan laboratorium menunjukkan peningkatan enzim pankreas yang sesuai dengan pankreatitis akut. Pasien menjalani pemeriksaan magnetic resonance cholangio-pancreatography (MRCP) serta endoscopic ultrasound (EUS) dan hasil kedua pemeriksaan menunjukkan dilatasi duktus pankreatikus yang bisa ditemukan pada disfungsi sfingter Oddi. Manometri tidak dilakukan karena keterbatasan fasilitas. Setelah menyingkirkan penyebab pankreatitis*

*akut rekuren lainnya, pasien menjalani sfingter otomi melalui endoscopic retrograde cholangio-pamcreatograh (ERCP). Gejala dan tanda pancreatitis akut hilang dan tidak terjadi rekurensi.*

**Kata kunci:** *pankreatitis akut rekuren, sphincter of Oddi dysfunction (SOD), endoscopic retrograde cholangio-pamcreatograh (ERCP), manometri, nyeri perut*

## INTRODUCTION

Recurrent acute pancreatitis (RAP) is defined as two or more occurrence of acute pancreatitis with no evidence of underlying chronic pancreatitis. Idiopathic RAP covers 20-30% of all cases of acute pancreatitis.<sup>1</sup> The causes of idiopathic RAP range from mechanical, toxic-metabolic, to miscellaneous. Etiological evaluation of RAP includes detailed history of alcohol intake, smoking, medications, trauma, viral illness, exposure to toxins, and family history.<sup>1,2</sup> Initial investigations of RAP include serum biochemistry and abdominal ultrasound. Abnormal liver function test (LFT) suggest biliary etiology of acute pancreatitis episode. Microlithiasis can be strongly suspected as the etiology when abnormal LFT and absence of gallstones on imaging are found. Patient is labeled as having idiopathic pancreatitis if the etiology remains unclear after the first attack.

Further investigations are needed to establish the cause of RAP include tests for lupus, hypereosinophilia, pancreatic tumor, pancreatic divisum, and sphincter of Oddi dysfunction (SOD).<sup>2</sup> Endoscopic ultrasound (EUS) has commonly replaced endoscopic retrograde cholangiopancreatography (ERCP) for diagnosing microlithiasis, biliary tumors, and congenital anomaly of pancreas which can contribute to the occurrence of RAP.<sup>2</sup> Whilst widely being replaced by EUS, ERCP still plays important role in diagnosing pancreatic divisum, SOD, and stones of common bile duct (CBD). Another leading benefit of ERCP that EUS does not have is its capacity to directly give therapeutic measures such as extraction of stones and sphincterotomy.<sup>2</sup> Managements of RAP focus on establishing the etiology while treating the episode of acute pancreatitis itself.<sup>1,2</sup>

Here, we present a case of 28-year-old female patient with clinical and laboratory findings suitable for acute pancreatitis. The patient had experienced the same attack for three times in the last 6 months. Microlithiasis, lupus, pancreatic tumor were ruled out as the cause of these attacks and therefore SOD was considered. Due to unavailability of manometry ye SOD was strongly suggested, the patient underwent ERCP and sphincterotomy was performed. After the

procedure, the patient did not experienced any serious complication and never had another attack of acute pancreatitis.

## CASE ILLUSTRATION

A 28 year old female patient came to Emergency Unit Cipto Mangunkusumo Hospital regarding worsening abdominal pain since four days before admission. The pain was dominantly felt at the right upper quadrant and referred to the left upper quadrant and the back. The pain was constant and the degree were high and intense. This complaint was accompanied by nausea, vomitus, and slight fever. The patient had experienced pain with the same characteristic twice accompanied with increasing level of pancreatic enzyme and was told having acute pancreatitis. Due to the etiology of RAP was not yet established, the patient was advised to go to Cipto Mangunkusumo Hospital.

Physical examination of the patient revealed that the patient was fully alert, looked moderately ill, had normal vital signs and nutritional status. Abnormal finding was found in abdominal area where the patient had palpable pain in the right upper quadrant, epigastrium, and umbilical area. Cullen's sign and Grey Turner's sign for acute pancreatitis were absent in this patient. There was also painless reddish plaque in the right forefoot. Other physical findings were unremarkable.

From laboratory findings, it was obtained that hemoglobin (Hb) level was 12.2 g/dL, leukocyte 10.700/mm<sup>3</sup>, eosinofil 12%, d-dimer 1.3, amilase 572 U/L, lipase 585 U/L. Anti nuclear antibody (ANA), anti ds-DNA, gamma glutamiltranspeptidase, and alkaline phosphatase (ALP) were all within normal limit. The differential count for eosinofil of the patient remained high after repetition and therefore the patient underwent examination to look into the possibility of thrombosis condition. The results of total IgE and total eosinofil were increased while level of beta 2 glycoprotein and anticardiolipin were within normal limit. In Figure 1, the magnetic resonance cholangiopancreatography (MRCP) showed widen caliber of pancreatic duct and other biliary tract showed no abnormality. After supportive treatment for acute pancreatitis, the amilase

and lipase level of the patient were decreased to below 2 x upper normal limit. The patient then underwent EUS examination and the result revealed edematous pancreas and mild dilatation of pancreatic duct which suggest an SOD (Figure 2).

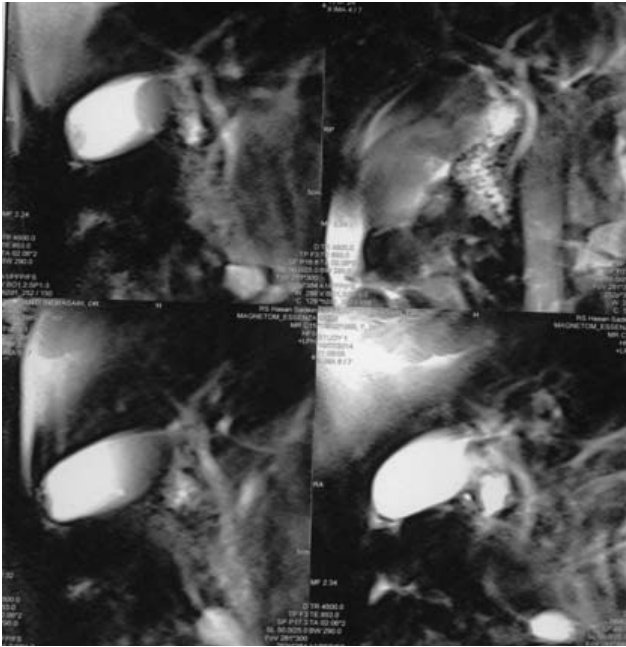


Figure 1. MRCP showed enlarged pancreas and dilated pancreatic duct

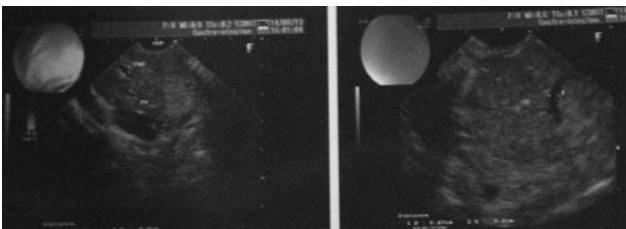


Figure 2. EUS showed edematous pancreas and dilated pancreatic duct

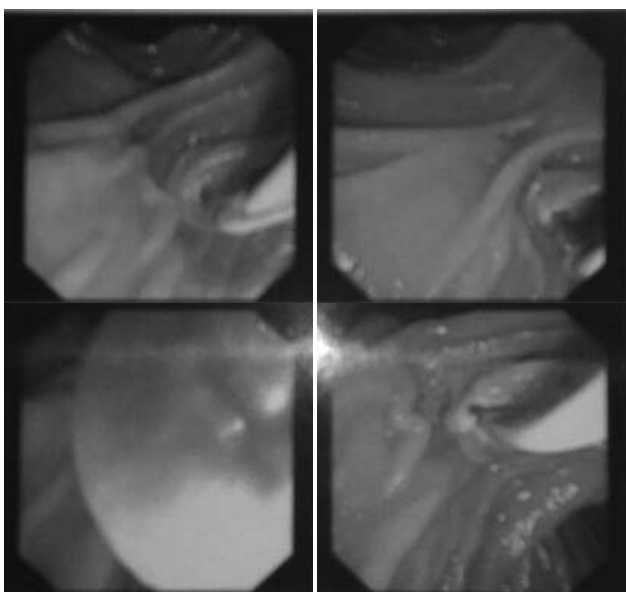


Figure 3. This figure showed sphincterotomy

Sphincter of Oddi dysfunction was considered the etiology of RAP in this patient. For manometry as the gold standard for diagnosing SOD was not available at that time, the patient was directly underwent sphincterotomy through ERCP (Figure 3). After ERCP the patient was stable and did not experience any significant complication. The patient was then discharged.

## DISCUSSION

In this case, the diagnosis of recurrent acute pancreatitis was established based on clinical signs and symptoms, elevated pancreatic enzyme, and documented history of previous acute pancreatitis. Following the resolved acute pancreatitis condition in this patient, the etiology of recurrent attack must be identified. The suspected cause of RAP in this patients included Sphincter of Oddi dysfunction, hypereosinophilic pancreatitis, systemic lupus erythematosus, and other autoinflammatory conditions. Laboratory findings did not support the diagnosis for any systemic condition whilst the imaging taken from EUS and MRCP both showed dilated pancreatic duct in which suggested SOD.

Sphincter of Oddi dysfunction itself is a structural and functional abnormality of SO that causes obstruction and primarily signed by abdominal pain both pancreatic or biliary.<sup>3</sup> Generally, this condition is formed by 2 different entities which are dyskinesia and stenosis. Seventy five percent of SOD's patients are young adult women eventhough all range of ages can also be affected. The Rome II diagnostic criteria are still used to help differentiating SOD symptoms from other biliary pain.<sup>3,4</sup> The latest classification divides SOD into four types based on pain, liver enzyme, pancreatic enzyme, and bile duct distention.<sup>5</sup> The gold standard of SOD is through endoscopic manometry in which the pressure of SO will be elevated.<sup>6</sup> Sphincterotomy is considered the most frequent approach to treat SOD other than surgery or pharmacologic therapy.

Manometry was not possible to be performed due to limitation of the facility. However, since other possible causes of RAP was not proven and findings of imaging showed dilated pancreatic duct, we performed sphincterotomy to the patient. The patient did not experience any significant complication from the procedure. Recent study showed that 55-59% of SOD patients have clinical improvement after sphincterotomy.<sup>7,8</sup> The most concerned complication of ERCP is pancreatitis. Other

less significant complications include abdominal pain and gastrointestinal complaints such as nausea and vomiting.<sup>9,10</sup>

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