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Management of pancreatic cysts in children

Postępowanie terapeutyczne u dzieci z torbielami trzustki

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S u m m a r y

Introduction. Pancreatic cyst is a pathological fluid collection located in the pancreas or directly adjacent to it. Pancreatic pseudocyst is the most common complication of acute or chronic pancreatitis. However, due to the rare occurrence of these entities in children, the knowledge about evolution and management of pancreatic cysts in paediatric patients is still very low.

Aim. To evaluate the management of pancreatic cysts in paediatric population.

Material and methods. Clinical data of 57 children with pancreatic cyst treated in a 10-year period were reviewed comprehensively: 13 with posttraumatic pancreatic pseudocyst, 13 with pancreatic cyst in the course of acute pancreatitis (AP) of other than traumatic etiology, 22 with pseudocysts as a complication of chronic pancreatitis (CP) and 9 patients with cysts of different etiology. The relationship between the etiological factor, treatment method and clinical outcome of the patients was analyzed.

Results. Among 26 paediatric patients with pancreatic cysts in the course of AP, both posttraumatic or not, 5 children (19.2%) were managed by conservative methods (antibiotics, nutritional therapy). Ten children (38.5%) underwent endoscopic treatment. Pseudocysts resolved after endoscopic cystogastrostomy or endoscopic retrograde cholangiopancreatography (ERCP) in 7 children (70%). There were no ERCP-related complications. Seventeen patients (65.4%) were treated by surgical drainage or resection. In 3 patients serious complications occurred, requiring relaparotomy or blood transfusion. Fifteen children (88.2%) after surgery had clinical and radiologic resolution of their cysts.

Since 2007, 22 of 215 children (10.7%) with CP were diagnosed with pancreatic pseudocyst. Twelve patients (52.2%) were treated by observation. Ten children (45.5%) underwent endoscopic cystogastrostomy or pancreatic duct stenting. Eight children after endoscopic treatment had clinical and radiologic resolution of their PP. Five patients (22.7%) were treated surgically. PP resolved after operation in all cases. There were no complications or failures related to surgical procedures. Some patients fell into more than one category.

Conclusions. 1. Endoscopic or surgical drainage of pancreatic cysts in children is a safe and effective procedure. 2. Asymptomatic cysts can be managed by a period of observation.

S t r e s z c z e n i e

Wstęp. Torbiel trzustki to patologiczny zbiornik płynowy zlokalizowany w trzustce lub bezpośrednio do niej przylegający. Torbiele rzekome są najczęstszym powikłaniem zarówno ostrego (OZT), jak i przewlekłego zapalenia trzustki (PZT). Ze względu na rzadkie występowanie tych jednostek chorobowych w populacji pediatrycznej, wiedza na temat leczenia torbieli trzustkowych u dzieci jest nadal bardzo wąska.

Cel pracy. Ocena sposobu leczenia torbieli trzustkowych w populacji pediatrycznej.

Materiał i metody. Analizie poddano dane 57 pacjentów z torbielami trzustki hospitalizowanych w latach 2007-2017: 13 z psudotorbielą pourazową, 13 z torbielą powstałą jako powikłanie OZT o innej etiologii niż uraz, 22 z torbielami trzustki powstałymi w przebiegu PZT i 9 pacjentów z torbielami o innej etiologii. Badano również związek między czynnikiem etiologicznym oraz metodą i wynikami leczenia.

Wyniki. Wśród 26 dzieci z torbielami trzustki powstałymi w przebiegu OZT zarówno o etiologii urazowej, jak i nie, 5 dzieci (19,2%) było leczonych zachowawczo (antybiotykoterapia, leczenie żywieniowe). Dziesięcioro pacjentów (38,5%) poddano zabiegom endoskopowym, z czego u 7 (70%) zaobserwowano ustąpienie torbieli. Nie było powikłań związanych z endoskopową cystogastrotomią lub endoskopową cholangiopancreatografią wsteczną (ECPW). Osiemnaście pacjentów (65,4%) wymagało zabiegu chirurgicznego: dre-

nażu operacyjnego torbieli lub częściowej pankreatektomii. U 3 dzieci wystąpiły poważne powikłania, wymagające relaparotomii lub transfuzji krwi. U 15 pacjentów (88,2%) stwierdzono po zabiegu ustąpienie objawów klinicznych oraz reabsorpcję pseudotorbieli w badaniach obrazowych. W ciągu 10 lat retrospektywnej obserwacji u 22 spośród 215 (10,7%) dzieci z PZT rozpoznano torbiele rzekome trzustki. Dwunastu pacjentów (52,2%) było leczonych zachowawczo. Dziesięcioro dzieci (45,5%) poddano endoskopowej cystogastrotomii lub protezowaniu przewodu trzustkowego, z czego u ośmiorga zabieg był skuteczny. Pięcioro pacjentów z PZT (22,7%) było leczonych chirurgicznie. U żadnego z nich po operacji nie stwierdzono ponownego wypełnienia się pseudotorbieli. Podczas leczenia chirurgicznego nie obserwowano żadnych powikłań. Niektórzy pacjenci zostali przypisani do więcej niż jednej kategorii.

Wnioski. 1. Endoskopowy lub chirurgiczny drenaż torbieli trzustki u dzieci jest bezpieczną i skuteczną metodą terapeutyczną. 2. Bezobjawowe torbiele mogą być leczone zachowawczo.

INTRODUCTION

By the end of the 1970s, the spectrum of diagnosed cystic pancreatic lesions was very narrow and limited mainly to post-inflammatory pseudocysts and mucosal or serous tumors. With the development of imaging techniques, progress has been made in recognizing and classifying pancreatic cysts. The incidence of pancreatic cysts is estimated to be between 1.2 and 20%, with increasing prevalence with age. A pancreatic cyst is a fluid collection located in the pancreas or directly adjacent to it. Cysts are single or multiple, with varying diameters ranging from a few millimeters to several dozens of centimeters. Depending on the cushioning layer on the inside, they can be true – lined with epithelial and more often occurring – pseudocysts. Pancreatic pseudocysts are usually a result of pancreatic insult with ductal disruption and leakage of pancreatic enzymes into surrounding soft tissues. These encapsulated fluid collections lack a true epithelial lining and is walled off by fibrous tissue. Cysts may cause pain, nausea, abdominal fullness, vomiting and jaundice. In adults neoplastic cysts are dominant, which account for about 60% of all cysts (1). In 2015 American Gastroenterological Association (AGA) Institute Guidelines, and the accompanying AGA Technical Review, on the diagnosis and management of asymptomatic pancreatic cysts in adult patients were published (2, 3). There are also many studies on classification, diagnosis and treatment of pancreatic pseudocysts which occur in the setting of acute or chronic pancreatitis, postoperatively, or after pancreatic trauma (4-9). Until now, there is still very few data about evolution and management of pancreatic cysts in children (10-20).

AIM

The aim of our study was to assess the management of pancreatic cysts in paediatric population.

MATERIAL AND METHODS

We retrospectively reviewed the charts of all children with pancreatic cysts hospitalized in the Children's Memorial Health Institute between 2007 and 2017, inclusive. There were 57 patients, 33 males and 24 females, 13 children with posttraumatic pancreatic pseudo-

cyst, 13 children with pancreatic cyst in the course of acute pancreatitis (AP) of other than traumatic etiology, 22 patients with pseudocysts as a complication of chronic pancreatitis (CP), 3 – with polycystic pancreas, 4 – with asymptomatic, accidentally detected pancreatic cyst, 1 – with congenital pancreatic cyst and 1 child with cyst caused by *Echinococcus* infection. The relationship between the etiological factor, treatment method and clinical outcome was analyzed. The results of pancreatic cysts treatment were reviewed with a breakdown of acute and chronic conditions.

RESULTS

Among 26 paediatric patients with pancreatic cysts in the course of acute pancreatitis, both posttraumatic or not, 5 children (19.2%) were managed by conservative methods (antibiotics, nutritional therapy). Ten children (38.5%) underwent endoscopic treatment: six (26%) – endoscopic cystogastrotomy, four patients (15%) – pancreatic duct stenting. Pseudocysts resolved after ERCP in 7 children (70%), the treatment of remaining three was ineffective or during the ERCP the operative indications were established. There were no ERCP-related complications. Seventeen patients (65.4%) were treated by surgical drainage or resection. In three patients bleeding to the bottom of the pancreatic cyst occurred, requiring relaparotomy or blood transfusion. Fifteen children (88.2%) after operation had clinical and radiologic resolution of their cysts. Some patients fell into more than one category.

Since 2007, 215 children with chronic pancreatitis were hospitalized in our institute. Twenty-two of them (10.7%) were diagnosed with pancreatic pseudocyst. Twelve patients (52.2%) were treated by observation. Three children (13%) underwent endoscopic cystogastrotomy. Cysts resolved after drainage. In seven patients (30.4%) ERCP with pancreatic duct stenting was performed. Five children after endoscopic stenting had clinical and radiologic resolution of their PP. Four patients (17.4%) were treated by surgical drainage or resection. One child underwent Jurasz's operation. PP resolved after operation in all cases. There were no complications or failures related to surgical procedures. Some patients fell into more than one category.

Three of four patients with asymptomatic pancreatic cyst were managed by observation. In one child a pseudocyst, size 3 x 3 cm, was removed during laparotomy. A procedure was complicated by acute pancreatitis with large post-inflammatory cyst which required endoscopic cystogastrostomy. In the boy with congenital pancreatic cyst, due to the large size and clinical symptoms, the abdominal mass was removed. Children with polycystic pancreas were managed by a period observation. In the two of them polycystic kidney disease coexisted. Patient with pancreatic cyst caused by *Echinococcus* infection received antiparasitic treatment.

DISCUSSION

Pseudocysts are uncommon lesions in paediatric population. In the 1970s, Cooney and Grosfeld reviewed the literature and identified worldwide only 75 well-documented paediatric cases (21). Despite the almost fifty years have elapsed, this number has only increased by several dozens of described children with pancreatic cysts (10-20). When in adults predominate neoplastic pancreatic cysts, in children the main causes include abdominal trauma, acute pancreatitis and chronic pancreatitis. Rarely, pancreatic cysts may be idiopathic, congenital or associated with infection, such as *Echinococcus* infection or tuberculosis. They may be also present in patients with renal polycystic kidney disease, medullary cystic kidneys, cystic fibrosis, and in other congenital morbidities such as Ivesmark syndrome, trisomy 13 or 15 chromosome, Elejalde syndrome, chondrodysplasia or familial fibrocystic pancreatic atrophy (1). Pancreatic cysts can be managed by conservative methods, percutaneous drainage, endoscopic drainage or by operative intervention. Treatment of pancreatic pseudocysts has changed over the years and the old teaching that the presence of cyst of more than 6 cm in diameter for 4-6 weeks should be treated surgically is no longer true (22).

The indications for drainage are now the presence of clinical symptoms, enlargement of cysts size, and complications (hemorrhage, infection, rupture or pancreatic/biliary ducts obstruction). The outcome of pseudocysts, which either can spontaneously resolve or require operation, depends also on the etiology of pancreatitis. Our experience shows children with cysts occurred due to abdominal trauma, require usually endoscopic or surgical intervention. Pseudocysts which are a result of non-traumatic acute or chronic pancreatitis, especially small, asymptomatic lesions, are more likely to be managed by a period of observation. Completely different conclusions drawn Teh et al., who noticed pseudocysts from nontraumatic etiologies more often require and benefit from surgical interventions, whereas pseudocysts from traumatic etiology are more amenable to conservative management (traumatic etiology – need for intervention in 45% vs nontraumatic – 92%) (10). As our research shows, as well as previously published reports, endoscopic cystogastrostomy is a safe and effective alternative to laparoscopic or open cystogastrostomy or drainage for the minimally invasive management of pancreatic pseudocysts in the pediatric population (11, 12, 20). However, it must be remembered that not all cysts are anatomically accessible. The failure of endoscopic intervention was even as high as 25% in large adult series (23). Similar to previous reports, no complications related to pancreatic cysts in children with polycystic pancreas were recorded (24, 25). Congenital pancreatic cysts or those with infectious etiology such as *Echinococcus* infection belong to casuistry of medicine and require an individual approach.

CONCLUSIONS

Endoscopic or surgical drainage of pancreatic cysts in children with CP is a safe and effective procedure. Asymptomatic cysts can be managed by a period of observation.

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