Renal Replacement Therapy

CASE REPORT

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Two cases of obturator hernia in patients undergoing hemodialysis: case report and literature review

Kosei Yamaguchi[1,2], Mineaki Kitamura[1,2*], Junichiroh Hashiguchi[2], Takashi Harada[2], Satoshi Funakoshi[2], Susumu Eguchi[3], Hiroshi Mukae[4] and Tomoya Nishino[1]

Abstract

Background: Obturator hernia (OH) is an extremely rare abdominal wall hernia with risk factors including aging, female sex, emaciation, and increased abdominal pressure. Its symptoms are nonspecific, and diagnosis is often delayed; however, this delay can lead to a fatal course. Therefore, early diagnosis and surgical intervention are necessary to reduce the mortality rate associated with OH. Considering the risk factors for OH, patients currently undergoing hemodialysis (HD) may be particularly vulnerable to OH. Here, we report two cases of OH in patients undergoing HD along with a review of the relevant literature.

Case presentation: Case 1 included a 76-year-old female undergoing HD due to autosomal dominant polycystic kidney disease. She was hospitalized for upper abdominal pain, vomiting, and diarrhea. On the day of hospitalization, she was diagnosed with OH using computed tomography and underwent emergency surgery. Case 2 included a 90-year-old emaciated female who was admitted to our hospital for projectile vomiting while undergoing HD. She was diagnosed with OH and shock, but surgery was not performed due to shock. Nonetheless, her blood pressure gradually increased, and she completely recovered. Spontaneous reduction in OH was confirmed on the third day of hospitalization. Both patients recovered well.

Conclusions: The symptoms of OH are non-specific, and certain symptoms such as vomiting and anorexia are often overlooked because they are common in patients undergoing HD. It is important to include OH in the differential diagnosis of digestive organ symptoms in patients undergoing HD, especially in those with risk factors for OH, such as elderly female patients on HD due to autosomal dominant polycystic kidney disease.

Keywords: Aging, Autosomal dominant polycystic kidney disease, Hemodialysis, Intestinal obstruction, Obturator hernia

Background

Obturator hernia (OH) is a rare disorder defined as the protrusion of the abdominal contents through the obturator foramen [1]. OH is more likely to develop in females; the elderly; multiparous, emaciated people; and those with increased intra-abdominal pressure due to complications [2]. OH is often undiagnosed or unsuspected because it is not apparent externally or palpably and is associated with nonspecific symptoms and signs [2]. Moreover, OH has serious consequences as delayed diagnosis often results in a severe disease course [3]. Thus, patients with risk factors for OH should be examined carefully.

*Correspondence: minekitamura@nagasaki-u.ac.jp

[1] Department of Nephrology, Nagasaki University Graduate School of Biomedical Sciences, Nagasaki, Japan
Full list of author information is available at the end of the article

Patients undergoing hemodialysis (HD) tend to be elderly [4], emaciated [5], and can have an increased intra-abdominal pressure if they suffer from autosomal dominant polycystic kidney disease (ADPKD) [6]. Despite being a rare complication, the incidence of OH in patients undergoing HD may be higher than that in the general population because of the characteristics of these patients. We report two cases of OH in patients undergoing HD along with a review of the literature.

Case presentation

Case 1

A 76-year-old female with ADPKD had undergone HD for two years. Her body mass index was 15.7 (weight, 38.2 kg; height, 156 cm), and she had previously delivered two children. Eight months previously, she had intermittent pain radiating from the right hip to the right thigh. She was diagnosed with disk herniation (L5/S1) by orthopedics and administered analgesics, but her pain worsened. She was admitted to our hospital for an intensive examination and treatment of upper abdominal pain, vomiting, and diarrhea lasting one day. On arrival, her body temperature was 36.9 °C, blood pressure was 174/93 mmHg, and heart rate was 92 beats per minute. A distended abdomen without tenderness or muscle guarding and hypoactive bowel sounds were noted on physical examination. There was no rebound pain or mass in the abdomen. The results of blood examinations were normal (Table 1).

Plain abdominal radiography revealed a dilated loop of the small intestine. Computed tomography (CT) revealed a dilated small bowel and ascites (Fig. 1a). A low-density mass was noted between the pectineus and right external obturator muscles (Fig. 1b), and the patient was diagnosed with incarcerated OH. She was transferred to another hospital and underwent immediate emergency laparotomy, the findings of which revealed that a loop of the ileum, approximately 50 cm from the ileocecal valve, had herniated into the right obturator canal. A

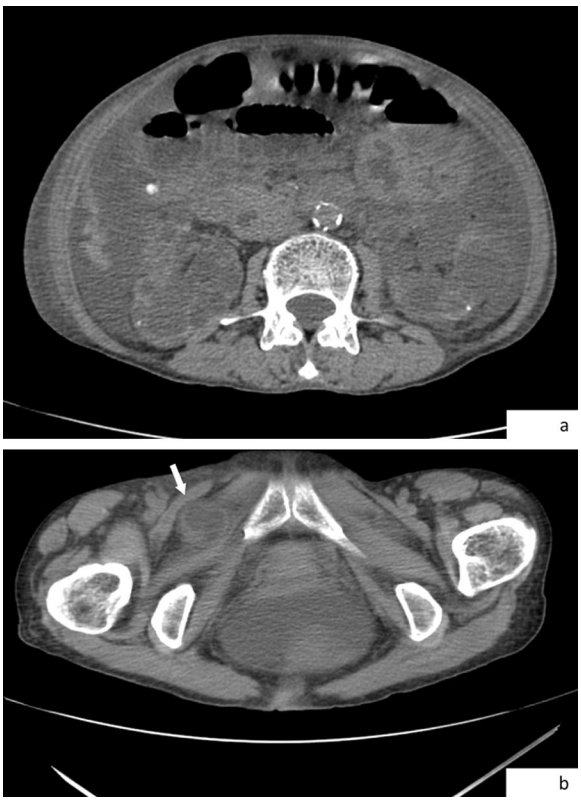

Fig. 1 Abdominal axial computed tomography images (case 1). **a** Dilated small bowel and ascites. **b** A low-density mass with a clear border located between the pectineus and right external obturator muscles (arrow)

Table 1 Laboratory data on the admission of case 1

Blood count			Serum chemistry					
WBC	5500	/μL	TP	5.4	g/dL	AST	26	IU/L
RBC	404×10^3	/μL	Alb	3.4	g/dL	ALT	17	IU/L
Hb	12.3	g/dl	BUN	66.3	mg/dL	T-Bil	0.5	mg/dL
Hct	37.0	%	Cr	10.4	mg/dL	ALP	245	IU/L
Plt	11.5×10^3	/μL	Na	137	mEq/L	γ-GTP	90	IU/L
			K	6.5	mEq/L	Amy	108	IU/L
pH	7.454		Cl	101	mEq/L	ChE	186	IU/L
HCO_3^-	21.7	mEq/L	Ca	8.8	mg/dL	LDH	303	IU/L
BE	− 1.2	mEq/L	P	6.9	mg/dL	CK	108	IU/L
						CRP	0.9	mg/dL

WBC white blood cell, *RBC* red blood cell, *Hb* hemoglobin, *Hct* hematocrit, *Plt* platelet, HCO_3^- hydrogen bicarbonate, *BE* base excess, *TP* total protein, *Alb* albumin, *BUN* blood urea nitrogen, *Cr* creatinine, *Na* sodium, *K* potassium, *Cl* chloride, *Ca* calcium, *P* phosphorus, *AST* aspartate aminotransferase, *ALT* alanine aminotransferase, *T-Bil* total bilirubin, *ALP* alkaline phosphatase, γ-*GTP* γ-glutamyl transpeptidase, *Amy* amylase, *ChE* cholinesterase, *LDH* lactate dehydrogenase, *CK* creatine kinase, *CRP* C-reactive protein

small perforation was noted on the incarcerated piece. Five centimeters of the perforated ileal segment were resected, and the defect at the site of the hernia was closed using non-absorbable interrupted sutures. The operation duration was 98 min. The patient recovered gradually and was discharged 10 days after surgery. The right hip pain resolved after surgery.

Case 2

A 90-year-old female had been undergoing HD for 24 years due to chronic nephritis. Her body mass index was 18.2 (weight, 38.3 kg; height, 145 cm), and she had previously delivered one child. She was previously diagnosed with anorexia (which lasted one month) and right thigh pain (which lasted for two weeks). She experienced a sudden attack of projectile vomiting during HD, following which the vomiting occurred repeatedly. On admission to our hospital, her body temperature was 38.2 °C, blood pressure was 197/81 mmHg, and heart rate was 83 beats per minute. Physical examination revealed hypoactive bowel sounds without distension, tenderness, or muscle guarding, and the laboratory results were unremarkable (Table 2). Plain abdominal radiography revealed a dilated loop in the small bowel. CT revealed dilation of the small bowel and ascites. A low-density mass was also noted between the pectineus and right external obturator muscles (Fig. 2a); however, this finding was overlooked at that time. She was hospitalized with a diagnosis of enteroparesis, and monitored continuously. Eight hours after hospitalization, her blood pressure gradually declined to 67/25 mmHg, and she developed shock. One day after hospitalization, a radiologist commented that the etiology of the ileus was OH based on radiological findings. We consulted a surgical team; however, emergency surgery was avoided because the patient was not expected to recover from the surgery due to the long duration of dialysis and her advanced age. However, two days after admission, her blood pressure gradually increased. Three days after admission, follow-up CT confirmed that the OH had spontaneously reduced (Fig. 2b). The vomiting and right hip pain resolved completely. The patient was discharged 11 days after admission. Moreover, her

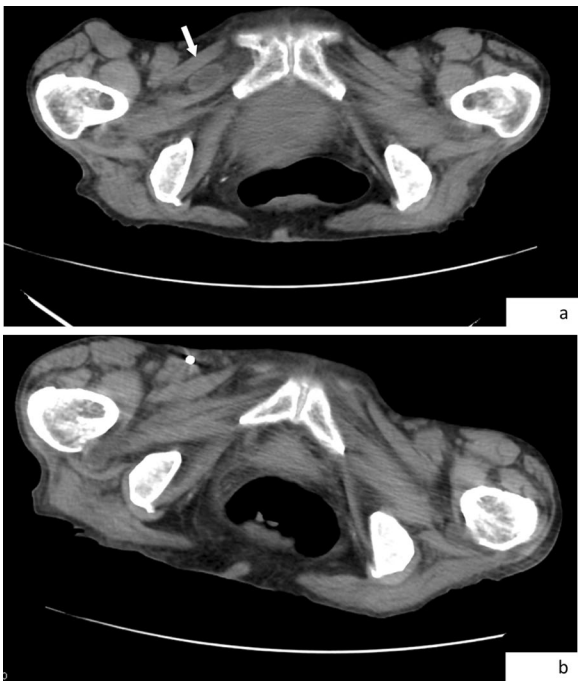

Fig. 2 Abdominal axial computed tomography images (case 2). **a** A low-density mass with a clear border located between the pectineus and right external obturator muscles (arrow). **b** The low-density mass disappeared

Table 2 Laboratory data on the admission of case 2

Blood count			Serum chemistry					
WBC	3730	/μL	TP	5.8	g/dL	AST	33	IU/L
RBC	391 × 10^3	/μL	Alb	2.8	g/dL	ALT	17	IU/L
Hb	12.7	g/dl	BUN	11.8	mg/dL	T-Bil	0.3	mg/dL
Hct	37.6	%	Cr	1.49	mg/dL	ALP	62	IU/L
Plt	14.6	/μL	Na	140	mEq/L	γ-GTP	18	IU/L
			K	3.9	mEq/L	Amy	107	IU/L
pH	7.496		Cl	101	mEq/L	ChE	288	IU/L
HCO_3^-	27.5	mEq/L	Ca	9.8	mg/dL	LDH	244	IU/L
BE	4.4	mEq/L	P	2.6	mg/dL	CK	90	IU/L
						CRP	0.23	mg/dL

WBC white blood cell, *RBC* red blood cell, *Hb* hemoglobin, *Hct* hematocrit, *Plt* platelet, HCO_3^- hydrogen bicarbonate, *BE* base excess, *TP* total protein, *Alb* albumin, *BUN* blood urea nitrogen, *Cr* creatinine, *Na* sodium, *K* potassium, *Cl* chloride, *Ca* calcium, *P* phosphorus, *AST* aspartate aminotransferase, *ALT* alanine aminotransferase, *T-Bil* total bilirubin, *ALP* alkaline phosphatase, γ-*GTP* γ-glutamyl transpeptidase, *Amy* amylase, *ChE* cholinesterase, *LDH* lactate dehydrogenase, *CK* creatine kinase, *CRP* C-reactive protein

appetite significantly improved after discharge. Elective surgical repair was considered. However, the patient and her family declined the surgery because of her advanced age, even though there was a possibility of recurrence. At nine-month follow-up, she was doing well and had no recurrence of the obturator hernia.

Discussion and conclusions

OH is a type of abdominal wall hernia that occurs when the internal organs of the abdominal cavity protrude from the obturator canal through which the obturator nerves and vessels pass [7]. OH has a higher incidence in females due to the greater width of the pelvis and larger obturator canal [8]. Right-sided OH is more common than left-sided ones since the left obturator foramen may be covered by the sigmoid colon [7]. The prevalence of bilateral OH was reported in only 6% of cases [9]. The protruding organ is usually the small intestine and intestinal obstruction is a clinical presentation [3]. OH is a rare disease, accounting for 0.07–1% of all abdominal wall hernias [10, 11], and is responsible for 0.4% of all patients with mechanical bowel obstruction [2]. However, it has a mortality rate of 12–70%, the highest of all abdominal wall hernias [12, 13]. The high mortality rate is attributed to the hernial orifice of the obturator foramen, which is narrow; once OH develops, it can easily develop into a strangulated hernia that obstructs blood flow and progresses to intestinal necrosis and perforation of the intestinal tract [11, 13, 14].

Since OH is a rare type of hernia [2], there are not enough reports to indicate the prevalence of OH in patients undergoing HD. However, various facts indicate that patients undergoing HD may be more likely to develop OH. Preperitoneal fat and lymphatic tissue in the obturator canal normally form a cushion around the neurovascular bundle to prevent herniation. When this fatty tissue is lost due to malnutrition, the risk of OH increases [15]. Malnutrition is a significant problem in patients undergoing HD due to uremia and amino acid loss from dialysis membranes [5]. Moreover, ADPKD increases intra-abdominal pressure and induces OH [16], although ADPKD accounts for just 4% of the etiologies of end-stage renal disease [4, 6].

A delayed diagnosis along with subsequent delayed surgical treatment in OH cases may cause intestinal strangulation, which compromises intestinal viability; this adversely affects the morbidity and mortality rates [2]. Therefore, patients with suspected OH should be confirmed early by abdominopelvic CT. Identification of a mass-like lesion between the obturator externus and pectineus muscles on CT is the gold standard for the diagnosis of OH [8, 17]. However, it is difficult to diagnose OH for several reasons. First, contrary to other abdominal wall hernias, a palpable groin mass is not noticeable in OH because the hernial mass is usually hidden beneath the pectineus muscle [10]. Second, most patients with OH present with non-specific abdominal symptoms resulting from mechanical bowel obstruction such as nausea, vomiting, and pain associated with abdominal cramps [18]. Such non-specific abdominal symptoms are often considered to be due to other benign etiologies. Third, even signs specific to OH are difficult to identify. The Howship–Romberg sign is a definitive indicator of OH and is caused by the compression of the obturator nerve by the hernia sac, resulting in pain on the medial side of the ipsilateral thigh during internal rotation of the hip joint [15]. However, this sign is observed only in 15–50% of all OH cases [3]. Furthermore, it is often confused with the symptoms of orthopedic diseases, such as arthritis and trauma [3]. The Hannington-Kiff sign is characterized by the loss of the thigh adductor reflex despite a positive patellar reflex [19]. This sign is also attributed to compression of the obturator nerve and is considered even more specific than the Howship–Romberg sign [3]. However, this sign is often not noticed unless physicians examine neurological findings carefully. Moreover, muscle reflexes in the lower extremities are not usually identified on physical examination for abdominal symptoms [20].

Diagnosing OH may be more difficult in patients undergoing HD due to non-specific abdominal symptoms, which are commonly observed due to the high prevalence of gastroparesis (delayed gastric emptying) and constipation in these patients compared with the general population [21, 22]. In a previous report, a patient undergoing HD with nausea and vomiting was misdiagnosed with reflux esophagitis and ileus until the autopsy revealed OH [23]. Furthermore, lower extremity pain such as the Howship–Romberg sign have also been commonly reported in patients undergoing HD [23, 24] because the mean age of these patients has increased due to the advanced age of these patients [4]. There are a few reports of OH in patients undergoing HD with the Howship–Romberg sign; however, in these reports, the sign did not lead to the diagnosis of OH [23, 24]. In Case 1, lower extremity pain was considered a symptom of disk herniation. Abdominopelvic CT taken eight months later finally led to a diagnosis of OH. We should have suspected OH based on the abovementioned non-specific and common symptoms and performed a CT scan. However, it should be noted that even CT findings are often overlooked, as seen in Case 2 and the previous report [23].

OH often has already progressed to a strangulated hernia at diagnosis [11, 14]. Therefore, the basic treatment for OH comprises urgent surgical repair of the

Table 3 Review of the literature on obturator hernia in hemodialysis patients

Age	Sex	BMI	Causes of RF	Period of HD (year)	Chief complaints	Howship–Romberg sign	Duration of symptom onset to admission (day)	Duration from admission to surgery (day)	Outcome	References
58	Male	N/A	CGN	–	Abdominal pain	+	14	1	Dead on the fourth hospital day	[29]
82	Female	N/A	ADPKD	14	Vomiting	+	2	–	Undiagnosed and died 11 days after admission	[23]
63	Female	19.0	ADPKD	–	Vomiting	+	1	1	Discharged 5 days after surgery	[24]
75	Male	15.4	CGN	5	Abdominal distension	–	1	1	Alive	[30]
67	Female	N/A	ADPKD	20	Abdominal pain, vomiting	–	7	1	Alive	[16]
76	Female	15.7	ADPKD	2	Abdominal pain, vomiting, diarrhea	+	2	2	Discharged 10 days after surgery	Present case
90	Female	18.2	CGN	24	Vomiting	+	1	–	Discharged 11 days after admission	Present case

RF renal failure, *N/A* not available, *CGN* chronic glomerular nephritis, *ADPKD* autosomal dominant polycystic kidney disease

incarceration and surgical closure of the hernia defect [25]. A previous study investigating 86 cases of OH showed that the mortality rate after 30 days was 5.5% in patients who underwent surgery (n = 73, including emergency procedures in 59 cases) and 46.1% in patients who did not receive surgical treatment due to their comorbidities, poor general status, and high operation risk (n = 13) [15], which confirms that surgery is the optimal treatment. As an option other than emergency surgery, the usefulness of manual reduction or ultrasound-guided reduction has been reported [26, 27]. This noninvasive retraction is considered to be performed in all cases; however, if it the lesion is not repaired, emergency surgery should be performed [26]. It is also stated that a standby closure surgery of the hernia defect should be performed after noninvasive reduction [26]. There are a few report of OH cases that spontaneously reduced, such as Case 2, and the course after reduction has not been fully elucidated [28]. However, spontaneous reduction cases may recur through the same hernia defect and require surgical repair [28], as in noninvasive retraction cases. In any case, surgical repair should be considered as a definitive treatment of obturator hernia.

Further, we conducted a literature review regarding OH in patients with HD. We searched medical databases, including MEDLINE and Google Scholar, and found only five cases reported previously (Table 3) [16, 23, 24, 29, 30]. Similar to the reports of patients not undergoing dialysis, there were more reports of elderly, emaciated females. Patients undergoing HD for a long time are reported more frequently; this is probably because they are prone to sarcopenia [31], which may cause OH. The etiology of end-stage renal disease in half of the reported cases was ADPKD. ADPKD may be a risk factor for OH as it is associated with an increase in intra-abdominal pressure. The mean time elapsed between the onset of abdominal symptoms and treatment was 1–2 days, which was shorter than that in previous reports (4–6 days) [3]. Patients undergoing HD may be diagnosed earlier because their frequency of visiting a medical facility for HD is approximately three times a week. However, we found the report of one case that was diagnosed at autopsy after death [23]. Patients undergoing HD tend to have multiple risk factors for OH and abdominal symptoms. Moreover, the Howship–Romberg sign tends to be misunderstood as being caused by benign complications and is thus overlooked in these patients. Therefore, cases of OH in patients undergoing HD could be underreported, and in some cases, death might have occurred because OH was undiagnosed.

This report has a few limitations. We could not count unreported cases of patients who had died, in whom OH was undiagnosed, or who had an unfortunate clinical outcome. Therefore, it is impossible to discuss the prognosis of patients undergoing HD with OH from the limited number of cases.

Conclusion

Herein, we report two cases of OH in patients undergoing HD. OH is infrequent, difficult to diagnose, and has a serious course. Considering that patients undergoing HD tend to have multiple risk factors for OH, the incidence of OH in these patients may be higher than that in the general population. The symptoms of OH are non-specific and can be confused with symptoms of benign conditions commonly observed in patients undergoing HD. This report aims to emphasize and highlight the importance of including OH in the differential diagnosis of patients undergoing HD with abdominal symptoms, especially those with risk factors for OH, such as advanced age, emaciation, and ADPKD, in order to reduce the mortality rate associated with OH in these patients.

Abbreviations

OH: Obturator hernia; HD: Hemodialysis; ADPKD: Autosomal dominant polycystic kidney disease; CT: Computed tomography.

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Author contributions

KY and MK wrote the initial drafts. KY, MK, JH, TH, SF, and SE participated in the discussion and treatment of the patients. HM and TN reviewed and revised the manuscript. All authors read and approved the manuscript and agreed with its submission to this journal.

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Availability of data and materials

The data and materials relevant to this case report are all included in this manuscript.

Declarations

Ethics approval and consent to participate

The present report and all procedures described in this report have been carried out in accordance with the 1964 Declaration of Helsinki and its later amendments. This study was approved by the Ethics Committee of Nagasaki Renal Center (Nagasaki, Japan) (21007).

Consent for publication

Written informed consents were obtained from the patients for the publication of this case report.

Competing interests

The authors declare that they have no competing interests.

Author details

[1]Department of Nephrology, Nagasaki University Graduate School of Biomedical Sciences, Nagasaki, Japan. [2]Nagasaki Renal Center, Nagasaki, Japan. [3]Department of Surgery, Nagasaki University Graduate School of Biomedical Sciences, Nagasaki, Nagasaki, Japan. [4]Department of Respiratory Medicine, Nagasaki University Graduate School of Biomedical Sciences, Nagasaki, Japan.

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