Int J Clin Exp Pathol 2013;6(5):994-997
www.ijcep.com /ISSN:1936-2625/IJCEP1302021

Case Report

Successful delivery after IVF-ET in an abdominal cocoon patient: case report and literature review

Dan Hu, Rui Wang, Ting Xiong, Han Wang Zhang

Reproductive Medicine Center, Tongji Hospital, Tongji Medical College, Huazhong University of Science and Technology, Wuhan, The People's Republic of China

Received February 17, 2013; Accepted March 23, 2013; Epub April 15, 2013; Published April 30, 2013

Abstract: Abdominal cocoon (AC) is a rare condition of uncertain etiology. We report the case of a 29-year-old infertile Chinese woman with AC, who successfully got twin pregnancy and delivery through in vitro fertilization (IVF) and embryo transfer (ET). And this review discusses the current concepts of its pathogenesis, diagnosis and treatments. AC might lead to tubal infertility and IVF-ET would be the most effective remedy for the patients desiring pregnancy.

Keywords: Abdominal cocoon, embryo transfer, in vitro fertilization, pregnancy

Introduction

Abdominal cocoon (AC), or sclerosing encapsulating peritonitis, is a rare condition of uncertain etiology and was first described and reported by Foo et al in 1978 [1]. It is characterized by partial or total encapsulation of abdominal organs by a fibrous membrane that looks like a cocoon. It is predominantly seen in young female, but a few cases of successful delivery with AC following in vitro fertilization (IVF) and embryo transfer (ET) have been reported. This article reports a case of twin pregnancy after IVF in an AC patient and reviews the current concepts of its pathogenesis, diagnosis and treatments, especially for primary infertility.

Case report

A 29-year-old Chinese woman attended our hospital with a 2-year history of primary infertility. Physical examination and lab tests were normal. Gynecological ultrasound identified a cyst in the right ovary, and the tumor biomarkers such as AFP, CEA, CA125 and CA19-9 were negative. Hysterosalpingography (HSG) presented left tubal obstruction and right tubal hydrosalpinx. In addition, the sperm analysis of her husband was normal.

Subsequently, she was treated with laparoscopic exploration for tubal factor. But the excessive pressure of intra abdomen handicapped operative procedure; the troca strayed into the intestinal lumen when the surgeon attempted to enter the peritoneal cavity forcibly. And then it was replaced by exploratory laparotomy for further management, peritoneum and greater omentum were not identified in the peritoneal cavity, part of intestine tubes were encased within an opaque, thick and dense fibrous membrane. Bowel perforation was explorated in the distal ileum, approximately 1 cm in diameter.

Tracing back her medicine history, the patient was a nonsmoker, nonalcoholic consumer, and without abdominal pain in peacetime; and there were no hepatopathy, tuberculosis, intraabdominal cavity administration, prolonged use of beta-blocker practolol and surgical history before. In addition, her lab tests and chest X-ray were normal. On the basis of the above findings, the diagnosis of AC was made.

Simple neoplasty was performed immediately. When the surgeon shaked uterine manipulator through the vagina, the fundus of uterus covered by membrane was only faintly visible, and bilateral adnexa revealed difficulty. Due to the severity of the adhesion, adhesiolysis became of little value. And IVF-ET was suggested to the patient for infertility therapy after the operation. Finally she got twin pregnancy after two IVF

cycles with gonadotropin-releasing hormone (GnRH) agonist protocols, and luteal support lasted up to the first trimester. It's also worth mentioning that 48 ml clear hydrosalpinx fluid was aspirated during the process of ovum pick-up in the first IVF trial. No gastrointestinal complications such as bowel obstruction occurred throughout pregnancy. There were no abnormal findings at each prenatal visit. At 35 weeks of gestation, the woman delivered by cesarean section because of the breech position and threatened premature labor, giving birth to two healthy baby girls who weighed 2140 g and 2530 g, respectively.

Discussion

AC is a rare condition in which total or partial encapsulation of intra-abdominal organs by a dense fibrous sac, which might lead to acute, subacute or chronic intestinal obstruction. As yet, the underlying etiology is still uncertain, and is thought to result from a single or complex interaction of multiple factors. There are many hypotheses of the etiology: 1. Congenital development malformation: the abnormal membrane was derived from the lining of the extraembryonic celom that has entered the abdomen with the intestine during the 12th week of gestation [2]; 2. Eyewinker stimulation: numerous possible etiological factors such as dialysate solution acidity, plasticizers and particulate matter in ambulatory dialysis could cause membrane formation secondary to peritonitis [3]; 3. Age and gender factor: retrograde menstruation in adolescent girls may induce fibrous capsule formation [1]; 4. Medicines affection: beta-blockers including practolol [4], propranolol [5], and metoprolol [6] have been known to cause peritoneal membrane; 5. Bacterial and viral infection: Staphylococcus aureus and Streptococcus infections were commonest in bacterial peritonitis [3]; ECHO viruses [7], Coxsackie B virus or adenovirus [8] have been incriminated as a cause of primary peritonitis; besides, tuberculous abdominal cocoon was being reported more frequently [9]. Even so, more work has to be done to elucidate the exact etiology of AC. Our patient was a young woman without abdominal pain in peacetime; and had no hepatopathy, tuberculosis, intraabdominal cavity administration, long-term use of beta blockers and surgical history before. So we assumed that inflammatory changes such as pelvic adhesion and hydrosalpinx might be involved in the process of peritoneal encapsulation.

Because the clinical manifestations are non-specific and vary from individuals, and hence the diagnosis is rarely made preoperatively, it is often diagnosed at the time of laparotomy or autopsy accidently. The condition is usually asymptomatic, a small percent of patients' symptoms are non-specific, such as abdominal pain, nausea, abdominal fullness, vomiting, an abdominal mass and bowel obstruction, but also shows primary infertility in female, which is usually misdiagnosised as chronic appendicitis, incomplete intestinal obstruction, ovarian cyst torsion and so on. Given this backdrop, there is a general sense that AC could be diagnosed when part or the entire abdominal organs are encased within a fibrous membrane, which couldn't be explained by any other diseases simultaneously. The diagnosis of AC in our patient was confirmed at the time of laparotomy, which revealed the characteristic uneven thickness of the membranes which encased some small intestine in a cocoon of opaque tissue [10].

However, imaging examinations play an important role in preoperative diagnosis of the disease. Barium meal follow-through (BMFT) described the radiological sign as small bowel clumped together in a serpiginous manner, giving the appearance of a cauliflower [11]. Contrast-enhanced computed tomography (CECT) showed that clusters of intestinal loops were enclosed within a membrane-like sac [12, 13]. Multi-detector row CT (MDCT) can clearly display the profile of the saclike membrane and provide more details to the surgeon [14]. Magnetic resonance imaging (MRI) displayed tortuous bowels with air-liquid matter, and peritoneal adhesion in the abdominal cavity directly [15]. Ultrasound presented a thick-walled mass containing bowel loops, loculated ascites and fibrous adhesions [3, 10, 12, 16]. Explorative laparotomy and laparoscopy may improve the diagnostic accuracy [13, 17]. The histological examination of the membranes presented proliferation of fibrous connective tissue with nonspecific chronic inflammatory reaction [16]. Unfortunately, there were no imaging results, and the characteristics mentioned above weren't found during ultrasound test, because our patient was asymptomatic and encountered incidentally during surgical

exploration. The reasons for which the disease was not paid attention to can be concluded as follows: the lack of knowledge due to its rarity and the untypical clinical symptoms and imaging examinations. Therefore, a better awareness of this disease and the combination of clinic and radiology may be facilitated in preoperative diagnosis.

Surgery is the first choice of symptomatic AC in almost all literatures. The principle is simple freeing the adhesions and excising the covering fibrous membrane on the small intestine carefully as much as possible. AC with infertility belongs to tubal infertility; we think the cause might be that fallopian tube and ovary were encapsulated within fibrous membranes, which limited the activity of fallopian tube and its function of picking up eggs or conveying gametes. The clinical symptoms of these patients are dormant without any self-conscious symptoms, but pelvic adhesion are severe, so any attempts to detach adhesion and recover the anatomic structure of the organs in the pelvic cavity would be futile. For this reason, the best treatment option for infertility is IVF and ET, and there is no difference in cycle regimens when the functions of uterus and ovary are normal. In our case, we did not perform any further surgical intervention for infertility after confirming AC during the operation, and IVF-ET was directly suggested for the treatment of infertility. Finally, she successfully got twin pregnancy and delivery after two IVF-ET attempts.

In general, AC is rare and the diagnosis is usually difficult before the surgery, especially for obstetricians and gynaecologists. Therefore, except for imaging examinations mentioned above, the disease would be highly considered for primary infertility when we encounter the following situations: 1. there are no abnormalities with the development of uterus and ovulation; 2. HSG presents that tubal are obstructive or passable, sometimes hydrosalpinx is suggested in the image; 3. intra abdominal pressure was abnormally high under laparoscope. As far as infertility treatment is concerned, bowel stimulation and damage would be avoided as much as possible when the diagnosis is confirmed during the operation, because operation stimulation might aggravate the illness and finally give rise to postoperative intestinal obstruction. At this moment, IVF-ET would be the most effective remedy for the patients desiring pregnancy.

Acknowledgements

The authors thank L.F. (Obstetrics, Tongji Hospital, Tongji Medical College, Huazhong University of Science and Technology, China) for providing the photographic documentation of caesarean section.

Disclosure of interests

The authors have no conflicts of interest to declare.

Address correspondence to: Dr. Han Wang Zhang, Reproductive Medicine Center, Tongji Hospital, Tongji Medical College, Huazhong University of Science and Technology, 1095 JieFang Avenue, Wuhan 430030, The People's Republic of China. Phone: +862783662591; Fax: +862783662534; E-mail: hanwangzhang@yahoo.com.cn

References

[1] Foo KT, Ng KC, Rauff A, Foong WC, Sinniah R. Unusual small intestinal obstruction in adolescent girls: the abdominal cocoon. Br J Surg 1978; 65: 427-30.

[2] Sahoo SP, Gangopadhyay AN, Gupta DK, Gopal SC, Sharma SP, Dash RN. Abdominal cocoon in children: a report of four cases. J Pediatr Surg 1996; 31: 987-8.

[3] Holland P. Sclerosing encapsulating peritonitis in chronic ambulatory peritoneal dialysis. Clin Radiol 1990; 41: 19-23.

[4] Eltringham WK, Espiner HJ, Windsor CW, Griffiths DA, Davies JD, Baddeley H, Read AE, Blunt RJ. Sclerosing peritonitis due to practolol: a report on 9 cases and their surgical management. Br J Surg 1977; 64: 229-35.

[5] Harty RF. Sclerosing peritonitis and propranolol. Arch Intern Med 1978; 138: 1424-6.

[6] Clark CV, Terris R. Sclerosing peritonitis associated with metoprolol. Lancet 1983; 1: 937.

[7] Fowler R. Primary peritonitis: changing aspects 1956-1970. Aust Paediatr J 1971; 7: 73-83.

[8] Tobe T. Inapparent Virus Infection as a Trigger of Appendicitis. Lancet 1965; 1: 1343-6.

[9] Gadodia A, Sharma R, Jeyaseelan N. Tuberculous abdominal cocoon. Am J Trop Med Hyg 2011; 84: 1-2.

[10] Nakamoto H. Encapsulating peritoneal sclerosis-a clinician's approach to diagnosis and medical treatment. Perit Dial Int 2005; 25 Suppl 4: S30-8.

[11] Navani S, Shah P, Pandya S, Doctor N. Abdominal cocoon--the cauliflower sign on barium small bowel series. Indian J Gastroenterol 1995; 14: 19.
[12] Mohanty D, Jain BK, Agrawal J, Gupta A, Agrawal V. Abdominal cocoon: clinical presentation, diagnosis, and management. J Gastrointest Surg 2009; 13: 1160-2.
[13] Qasaimeh GR, Amarin Z, Rawshdeh BN, El-Radaideh KM. Laparoscopic diagnosis and management of an abdominal cocoon: a case report and literature review. Surg Laparosc Endosc Percutan Tech 2010; 20: e169-71.
[14] Wang Q, Wang D. Abdominal cocoon: multi-detector row CT with multiplanar reformation and review of literatures. Abdom Imaging 2010; 35: 92-4.
[15] Prokesch RW, Schima W, Schober E, Vychytil A, Fabrizii V, Bader TR. Complications of continuous ambulatory peritoneal dialysis: findings on MR peritoneography. AJR Am J Roentgenol 2000; 174: 987-91.
[16] Jain P, Nijhawan S. Tuberculous abdominal cocoon: a case report and review of the literature. Am J Gastroenterol 2008; 103: 1577-8.
[17] Ertem M, Ozben V, Gok H, Aksu E. An unusual case in surgical emergency: Abdominal cocoon and its laparoscopic management. J Minim Access Surg 2011; 7: 184-6.