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CLINICAL CASE

Pregnancy and short bowel syndrome treated with parenteral nutrition

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KEYWORDS

Parenteral nutrition;
Short bowel
syndrome;
Intestinal ischemia;
Pregnancy

Abstract

Background: Total parenteral nutrition has improved the survival of patients with short bowel syndrome; however, it has been used infrequently in pregnant patients.

Clinical case: We present the case of a 32-year-old woman who was 22 weeks pregnant at the time of admission and required a massive resection of 4 m of small intestine due to mesenteric torsion. The patient did not present surgical complications during the immediate post-operative period and there were no adverse effects on the foetus. Management with parenteral nutrition was carried out for 105 days, showing metabolic changes in the lipid profile of the mother without any other significant changes. The composition of solutions for enteral nutrition was managed with lipids as non-protein caloric intake.

The foetus was monitored by ultrasonography with no change in its development. The newborn presented arterial hypertension secondary to vesicoureteral reflux, which was successfully resolved with treatment.

Conclusions: The presentation of this case demonstrates the effectiveness of the management of short bowel syndrome in a pregnant patient without changes in gestation evolution.

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PALABRAS CLAVE

Nutrición parenteral;
Síndrome de intestino
corto;
Isquemia intestinal;
Embarazo

Embarazo y síndrome de intestino corto tratado con nutrición parenteral**Resumen**

Antecedentes: La nutrición parenteral total ha mejorado la supervivencia de los pacientes con síndrome de intestino corto; sin embargo, se ha empleado en pacientes con embarazo en pocas ocasiones.

Caso clínico: Se presenta el caso de una paciente de 32 años de edad, con embarazo de 22 semanas de gestación, que requirió resección masiva de 4 m de intestino delgado por torsión mesentérica. No presentó complicaciones quirúrgicas durante el postoperatorio inmediato ni efectos adversos sobre el producto. Se manejó con nutrición parenteral durante 105 días, con cambios metabólicos en el perfil de lípidos de la madre, sin otros cambios de importancia. La composición de las soluciones de nutrición parenteral se manejó con lípidos, como aporte calórico no proteico.

El producto fue controlado ultrasonográficamente sin cambios en su desarrollo. Al nacimiento, presentó hipertensión arterial secundaria a reflujo vesicoureteral, que se resolvió satisfactoriamente con tratamiento.

Conclusiones: La presentación de este caso demuestra la eficacia del manejo del síndrome de intestino corto en una paciente embarazada, sin alteraciones en su evolución gestacional ni afectación para el producto.

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Background

Pregnancy involves multiple physiological changes in women, which evolve depending on their health. Some can impact on the nutritional status and development of pregnancy^{1,2}. These changes are fragile when the patient has a condition that warrants an acute abdominal surgical procedure, especially when it is urgent such as a massive bowel resection. This situation poses serious challenges to both the health team involved (midwife) and the surgeon and to treatment planning and care processes in the immediate postoperative period and long term.

When an extensive intestinal resection is conducted, survival conditions for the mother and the foetus depend on the care with which they are provided, thus the main points to consider are as follows: avoid intraoperative and postoperative septic complications and maintain a stable haemodynamic status. The mother's nutrition will depend on any additional requirements of the pregnancy and the medications used to maintain adequate uteroplacental blood flow³.

A massive bowel resection in a pregnant patient is associated with high mortality, especially for the foetus⁴. Since its initiation, parenteral nutrition has shown favourable results during long periods in patients with short bowel syndrome⁵. There is currently limited experience in its involvement with pregnant women and short bowel syndrome and, because of the peculiarity of this situation, each case treated provides new insights into the management of this syndrome and offers new standards or guidelines to follow based on the results obtained⁶.

This is the case of a 32-year-old woman who was 22 weeks pregnant and required a massive small bowel resection of 4 m caused by an internal hernia with mesenteric torsion treated with parenteral nutrition until 35 weeks gestation.

The patient underwent a caesarean section resulting in a live birth.

Clinical case

Female patient, 32 years old, with a surgical history at age 15 for a small bowel tumour. A resection of 30 cm was carried out and there was no specific aetiology of the tumour. Gyneco-obstetric history: gravida 2 para 0, abortus 1, with a gestation of 22 weeks. She had experienced intermittent crampy abdominal pain for 4 days with no vomiting or stools. On the last day she presented data of systemic inflammatory response. Her tests included plain x-rays and abdominal ultrasound.; No computed tomography (CT) was performed.

A laparoscopy was performed and subsequent to that a laparotomy because she presented an internal hernia with torsion of the small bowel, with well established necrosis of 4 m on the small bowel, jejunum and ileum (fig. 1). The origin was a defect in the mesentery in the area where the tumour resection had been made, as reported in the surgical history.

Given the frank evidence of a well-established ischemic, a small bowel resection was performed with irreversible vascular lesion of 4 m. No distortion of the affected bowel was conducted. An anastomosis was performed in the remaining small bowel, 7 cm terminal ileum and 30 cm jejunum; the ileocecal valve and colon was preserved in its entirety (Figs. 2 and 3).

Postoperative evolution was satisfactory, with haemodynamic stability of the patient and the foetus was unchanged. The next day parenteral nutrition was initiated, which lasted until 35 weeks of pregnancy.

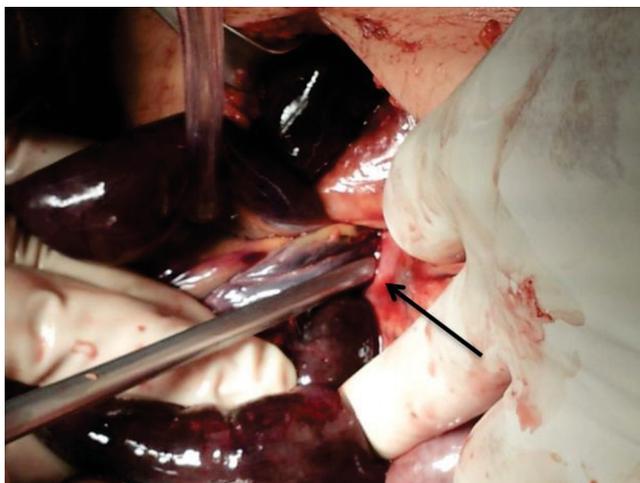


Figure 1 Ischemia of the small bowel (4 m) from a mesentery torsion due to internal hernia in the intestinal mesentery.

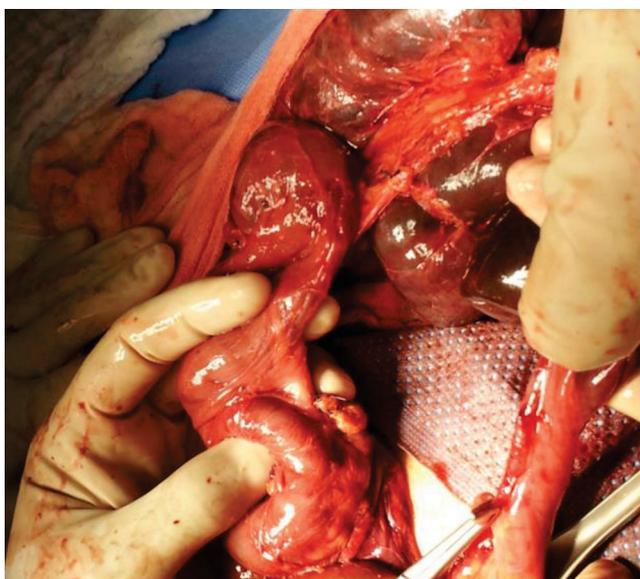
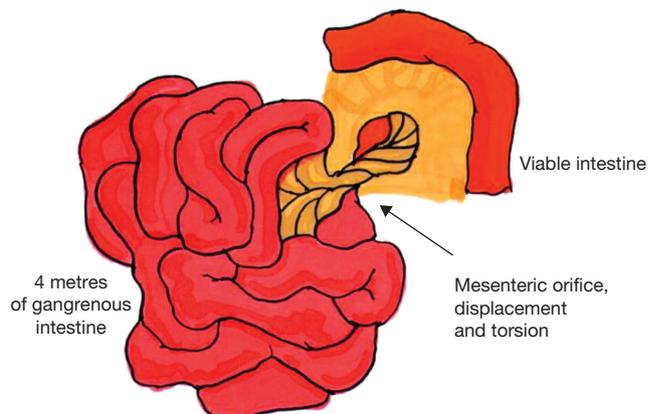
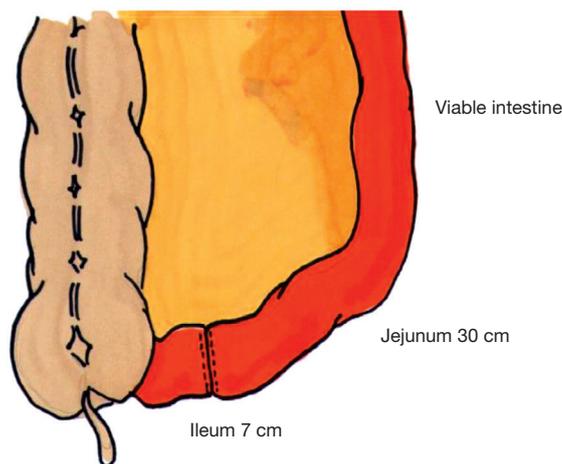


Figure 2 Massive resection of the small bowel with ileus-jejunum anastomosis with 7 cm of terminal ileum and 30 cm of residual jejunum.



For the remainder of her pregnancy the patient presented with small amounts of liquid stools, on an average of 3-4 times a day. She reduced oral intake to fluids and an elemental diet at low doses.

Her daily parenteral nutrition was calculated with a ratio of protein/kg of weight of 2 g; fat/kg of 1.5 g at onset and subsequently 0.75 g; the calorie/nitrogen relationship was 100:1. In addition: glutamine 30 g, zinc 15 mg, folic acid 15 mg, trace elements and vitamins. The remaining nutritional elements were administered as needed during pregnancy. To maintain the stability of the patient, her nutrition was managed with minimal changes.

The pregnancy was monitored with foetal ultrasound every week, with an appropriate growth of the foetus and placenta, without presenting anatomical or functional abnormalities until 35 weeks gestation when the caesarean birth was performed.

Parenteral nutrition was administered with a double lumen catheter via percutaneous infraclavicular route for a period of 94 days. It was managed with the centre's technique and there were no infectious or mechanical catheter complications.

The patient had clinical and obstetric daily monitoring during the course of pregnancy. Her weight curve developed normally according to the evolution of the pregnancy without presenting clinical manifestations of any nutrient deficiency and with suitable nutrition tolerance.

The patient's total serum lipid values, cholesterol and high and low density lipoprotein increased from the fifth week when the contribution of lipids were reduced to 0.75 g/kg; they were reduced again after 6 weeks and kept moderately elevated (Table 1). Serum glucose values remained at normal levels throughout the entire pregnancy.

The ponderal weight curve depended on the stage of gestation. No surgical clinical complications were observed or



Figure 3 Oesophagus-gastro-duodenal series showing the small bowel residual with preservation of the ileocecal valve.

complications of the short bowel syndrome pathology caused by the massive small bowel resection.

At 35 weeks gestation, with clinical and ultrasound monitoring, it was decided that a caesarean section would be performed, which was successful and resulted in the birth of a female infant whose weight was 2,200 g and an Apgar score of 9/9. The patient evolved without postoperative complications. The decision to interrupt the pregnancy was taken because the foetus was mature.

At 15 days, hypertension was identified in the foetus, which disappeared, and her lipid profiles were normal; the vesicoureteral reflux that was identified was corrected. The infant did not present any other alterations. Physical, intellectual and motor development were normal.

The mother continued with parenteral nutrition at home, following a low-residue diet, with a gradual reduction in parenteral nutrition during 2 years from the beginning of treatment.

Discussion

The case is regarding a patient with bowel syndrome caused by intestinal ischemia, occurring at 22 weeks gestation.

There are several considerations to take into account in this case. The mother's pregnancy was normal until 22 weeks. With a history of a previous intestinal resection due to an unspecified intestinal tumour, her profile showed intermittent abdominal pain shortly before surgical treatment, with evidence of systemic inflammatory response that did not correlate with the severity of the surgical findings. Radiology studies were plain x-rays and ultrasound or her abdominal area; no abdominal CT was performed. The use of CT in cases of acute abdomen with pregnancy has increased to 25%, with no apparent impact to the foetus. The diagnostic imaging guides suggest that, during pregnancy, in cases such as the one presented in which single studies were inconclusive, a low-dose CT should be performed⁷. In this case, delay of the study facilitated the establishment of such severe intestinal necrosis that was totally irreversible. There are acute abdominal conditions associated with pregnancy in which the diagnosis is made difficult by the pregnancy itself, such as in acute appendicitis, trauma or intestinal occlusion.

In a review, Boland et al.⁴ reported cases of small bowel resections with maximum extensions of 160 cm, with the death of the foetus in cases of 22 weeks gestation, like the one reported herein.

Decisions taken during the surgery of this patient with an extensive, severe and irreversible lesion because of the affected bowel conditions were ischemia caused by sliding of the intestine in a mesenteric window of the resected bowel. It was decided to not rotate the gangrenous intestine. A resection was performed from the ischemia free zone; a derivative process was not carried out to try to preserve the ileocecal valve. This allowed the survival of the foetus and the absence of maternal complications. The distortion of the affected bowel of 4 m would have caused a release of inflammatory mediators and bacteria that would lead to the death of the foetus and probably the mother. A bowel resection was performed and an

Table 1 Lipid profile during pregnancy, treated with parenteral nutrition

Weeks of gestation	22	23	24	25	26	27	28	29	30	31	32	33	34	35
Cholesterol (mg/dl)	235	109	236	232	285	289	338	357	391	330	254	180	181	208
TG (mg/dl)	62	181	307	243	296	298	320	325	363	229	151	117	186	237
Total lipids (mg/dl)	312	522	976	900	1045	1055	1184	1227	1356	1006	729	535	662	802
HDL (mg/dl)	25.5	17	29.6	31	23.4	21.4	24.9	27.8	27.8	28.8	28.4	30.7	23.9	34.4
Atherogenic index (mg/dl)	3.5	6.43	7.9	8.3	12.1	13.4	13.5	12.8	14	11.4	8.9	5.8	7.5	6
LDL (mg/dl)	47	56	145	181	202	208	261	267	297	256	195	126	120	126
VLDL (mg/dl)	16.7	36.2	61.3	47.9	59.2	59.5	64	64.9	72.5	45.7	30.2	23.3	37.2	47.5

HDL: high-density lipoprotein; LDL: low density lipoprotein; TG: triglycerides; VLSL: very low-density lipoprotein.

anastomosis of the remaining intestine, 7 cm of the ileum and 30 cm of the jejunum was conducted, despite the high risk of dehiscence in order to preserve the ileocecal valve and colon in their entirety. It was decided to not perform a jejunostomy and avoid the risk of an anastomosis because of the length of the residual intestine, 30 cm of jejunum, which would leave the patient with an ostomy that would be very difficult to manage with certain catastrophic results, nutritional, jejunostomy control and certainly to the pregnancy.

Foetal mortality of up to 100% has been reported when there is an association between intestinal occlusion and massive small bowel resection in cases of pregnancy of 20-25 weeks of gestation. Morbidity and mortality of massive bowel resection of the magnitude presented by this patient is high due to clinical conditions resulting from diarrhoea and electrolyte changes. There were no respiratory complications or postoperative anastomosis in the patient and she regained her intestinal integrity with the limitations of the residual intestine.

Knowledge of the basics of nutrition in patients with short bowel syndrome is essential because there are multiple metabolic changes that occur in the process of adaptation of the remaining bowel, which was minimal in this case, especially the ileum, as the resection was almost in its entirety. Once the post-surgical process had been successful where the metabolic response to trauma was controlled by closely watching the foetus, which was unchanged. It was decided to establish a programme based primarily on parenteral nutrition as the main nutritional support. In order to maintain minimum stimulus of the remaining intestine, avoid hypoperfusion and the possibility of anastomotic leakage as well as electrolyte abnormalities due to excessive loss of the remnant digestive tract. Parenteral nutrition was opted for because of the affected gastrointestinal tract, which started the day following surgery to reduce the hypoperfusion lesion and damage to distant organs.

The use of total parenteral nutrition in pregnant patients was analysed by Landon et al.⁸ who reported the importance of the mother's good nutritional condition to avoid prematurity, low birth weight and intrauterine growth retardation. The patient had an excellent nutritional status prior to the surgical event and parenteral nutrition was initiated early, anticipating the possibility of haemodynamic and nutritional disorders, first from the metabolic response to trauma because of the extent of resection and second from changes that would occur in the absence of most of the small bowel.

The patient was in the second trimester of pregnancy, a period in which the human placental lactogen (hPL) rises as well as other anti-insulin hormones, which are synthesised by the placenta and modify maternal glucose and amino acid usage; hPL stimulates lipolysis and fatty acids and glycerol becomes the main energy substrate for foetal growth.

Other metabolic changes to consider during pregnancy are increased mobilisation of glycogen with an increase of basal insulin production as carbohydrates and lipids are the main energy substrates during pregnancy. Throughout the treatment with parenteral nutrition the patient was administered combined carbohydrates and lipids as major energy substrates, depending on the mother's metabolic response to trauma and serum lipid values.

Amato and Quercia⁹ conducted a review of the safety of lipid emulsions during pregnancy. The mother and the de-

veloping foetus require fatty acids and prostaglandin synthesis. Some of the effects produced by lipid derivatives of cottonseed oil were fat deposits in the placenta, premature labour induction from stimulation of the uterus and foetal abnormalities. The solutions used today, derived from soybean oil and sunflower seed, do not generate these problems^{10,11}. Physiological hyperlipidaemia is present during pregnancy because the administration of lipids in parenteral nutrition can result in excessive levels of triglycerides and cetonemia, which can be harmful to the foetus. Changes in the placenta can also occur such as heart attacks and fat deposits. The patient presented was administered lipid-based soybean oil and medium-chain triglycerides throughout pregnancy with the aforementioned adjustments depending on serum values. Neither the placenta nor the foetus suffered adverse effects from the administration of lipids.

Protein requirements during pregnancy are not accurate. An increase of 25-30 g/day is calculated; 2 g of protein per kg was administered in the case presented taking into account the trauma caused by the large intestinal resection and protein loss by diarrhoea. During the nutrition period, 30 g of glutamine was administered to cover, in addition to foetus requirements and the proper functioning of the placenta, the requirements of losses by the intestine and the need to maintain a proper intestinal barrier to prevent bacterial translocation.

Zinc has a vital role in a variety of metabolic, immunological and enzymatic functions. Reyes et al.¹² identified serum zinc and copper deficiency in patients with parenteral nutrition with clinical manifestations compatible with this deficiency. The necessary requirements were identified in patients with high enterocutaneous fistulae output and short bowel syndrome. The requirements for these patients were 15-20 mg of zinc/day.

Oligoelements or trace elements are essential components in the administration of parenteral nutrition. Btaiche et al.¹³ conducted a review of the concentrations of these elements in 26 patients with parenteral nutrition at their home, with and without short bowel syndrome. The total days of nutrition at home was 40,493. Serum determinations of the oligoelements were made once a year. When the authors compared doses of oligoelements proposed by the clinical guidelines and ranges they obtained, they concluded that higher doses of zinc should be administered in patients with prolonged nutrition and higher increase of intestinal losses.. They suggested low doses of copper, manganese and chromium to prevent long-term accumulation.

Considering the massive small bowel resection and losses due to diarrhoea, the patients were administered 15 mg of zinc, 3.38 mg of copper and 7.62 mg manganese daily. During treatment, the patient had no clinical signs of deficit and serum determinations were normal.

Baker et al.¹⁴ conducted a study on the role of the placenta in maternal-foetal vitamin transfer in humans. Serum determinations of B₁₂, B₆, biotin, folate, thiamin, riboflavin, pantothenate and nicotinate vitamins were conducted in maternal and foetal serum as well as in placental tissue in normovitaminemic and hypovitaminemic mothers who ingested vitamin supplements during pregnancy. They found no biotin deficiency. Intramuscular administration of ribofla-

vin, nicotinate and pantothenate is required for increases in maternal and foetal blood and placental tissue.

Velázquez et al.¹⁵ demonstrated that patients with short bowel syndrome who do not receive adequate doses of biotin presented clinical manifestations that interfere with cell metabolism. Our patient received daily administration through a multivitamin containing biotin and other elements of the complex B.

Electrolyte requirements were managed according to losses and taking the growth of the foetus into account.

Kirby et al.¹⁶ conducted a literature review of the experience of the use of parenteral nutrition in patients during pregnancy. Most items are single cases where the most frequent indications were from hyperemesis gravidarum, pancreatitis, inflammatory bowel diseases (chronic ulcerative colitis and unspecified Crohn's) and intestinal occlusion. The average treatment time was 4.8 weeks.

Conclusions

Parenteral nutrition in pregnant patients has been performed for short periods. There are some publications in cases of Crohn's disease or chronic intestinal pseudo-obstruction where it has been observed that nutrition can be done with few complications. There are several lessons about its management that can be learned from the case reported: the diagnosis of intestinal obstruction was late because a CT was not performed, intraoperative management was successful as the morbidity and mortality of the mother and the foetus were zero; parenteral nutrition therapy was performed without complications at initiation because of good surgical response, and parenteral nutrition was managed with lipids as caloric intake with changes in the patient's serum determinations that did not alter evolution.

Foetal development was normal with gynaecological monitoring by serial ultrasound. Parenteral nutrition allowed a satisfactory evolution of both the mother and the foetus.

Conflict of interest

The authors declare no conflict of interest.

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