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Original article

Pregnancy is possible on long-term home parenteral nutrition in patients with chronic intestinal failure: Results of a long term retrospective observational study

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SUMMARY

Background & aims: Home parenteral nutrition (HPN) improves survival and quality of life in patients with chronic intestinal failure (IF). Few cases of pregnancy on HPN have been published. The aim of this study was to report pregnancy cases on long-term HPN in benign IF.
Methods: This retrospective study included all pregnant patients on HPN from 4 HPN referral centers. Data on demographics, ongoing pathology, HPN type, maternal and newborn complications were collected.
Results: From 1984 to 2014, 21 pregnancies occurred in 15 patients (short bowel syndrome (n = 11), motility disorders (n = 3), mucosal disease (n = 1)) of whom 14 occurred after 2010. Median follow-up was 12 years. Median HPN duration before pregnancy was 8 years. HPN was adapted monthly during pregnancy, with close monitoring and supplementations. Energy intake was regularly increased and median maternal weight gain was 10 kg. Median age at the first pregnancy was 27 years. In 55% of cases, the newborn was preterm. Maternal complications occurred in 67% of cases (mainly due to underlying disease or HPN complications). There were 3 post-partum hemorrhages and 6 hypotrophic newborns. Eighteen infants were healthy and 2 chronic intestinal pseudo-obstruction (CIPO) were suspected.
Conclusion: Our series, the largest reported to date, shows that pregnancy is possible in HPN patients but the complication rate is high. A specific support is necessary, particularly in CIPO patients. As pregnancies have increased over the last 15 years, physicians practicing in HPN referral centers should be aware of the need for implementing a specific multidisciplinary monitoring in HPN patients considering pregnancy.

1. Introduction

Intestinal failure (IF) has been first described in 1981 by Fleming and Remington as “a reduction in the functioning gut mass below the minimal amount necessary for adequate digestion and absorption of food” [1]. Recently, IF definition has changed for “the reduction of gut function below the minimum necessary for the

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absorption of macronutrients and/or water and electrolytes, so that intravenous supplementation is required to maintain health and/or growth" [2]. IF may be classified into five major pathophysiological conditions, which may originate from various gastrointestinal or systemic diseases: short bowel syndrome (SBS), intestinal fistula, intestinal dysmotility, mechanical obstruction and extensive small bowel mucosal disease [2]. Various pathologies may lead to chronic IF.

The main treatment of chronic IF is parenteral nutrition (PN) which has appeared in the 1960's and home PN (HPN) has been developed a few years later. This nutritional support improves patient quality of life, and patients on HPN may make family projects [3,4].

In 1972, Lakoff et al. have described the first case of pregnancy in a patient with anorexia nervosa treated with PN [5]. In 1981, Cox et al. have reported a case treated with total HPN during the last trimester of pregnancy for Crohn's disease [6]. Since then, various publications have reported patients on PN during pregnancy with successful outcome [7], mostly for pancreatitis or hyperemesis gravidarum. However, only a few cases treated with PN for IF during pregnancy have been described. In a systematic literature review from 1972 to 2015, we identified 14 pregnancies in 12 IF women on HPN [8–21]. The main complication was prematurity and postpartum hemorrhage occurred in one patient.

In these women, the nutritional needs during pregnancy should integrate the needs related to the pregnancy and those related to the underlying disease itself. The main issue is the risk for macronutrient, and especially micronutrient, deficiencies. Indeed, in case of vitamin (vitamin B9, D, K and A), iron or zinc deficiencies, the consequences may be dramatic for the fetus.

Given the prolonged survival and improved quality of life on HPN, an increasing number of IF patients on HPN may plan a pregnancy. However, it remains difficulty to answer questions about the possibility of pregnancy, the optimal management of these pregnancies and the factors which should be closely monitored to avoid complications. The aim of this study was to report pregnancy cases in benign IF patients on long-term HPN from three referral centers to enhance our expertise.

2. Materials and methods

This retrospective observational study was conducted in four adult HPN referral centers: Clichy (Beaujon Hospital), Rouen, Lyon and Bordeaux in France. All pregnant patients treated with HPN for benign chronic IF between 1984 and 2014 were identified. The patients had a multidisciplinary monitoring with gastroenterologists, nutritionists, gynecologists and a paramedical team (nurses, dieticians, pharmacists…). Inclusion criteria were: long-term HPN (defined as a treatment duration >6 months) for benign chronic IF from 1984 to 2014 in one of the four referral centers and occurrence of a pregnancy over this period. The patients with history of hydroelectrolytic supplementation and having initiated HPN at the beginning of the pregnancy were also included. Data on demographics, comorbidities, nutritional support characteristics, and possible maternal or fetal complications and outcome were collected in 2014 from patient medical records. In case of missing data, the patients or referring physicians were contacted by phone and questioned.

3. Results

3.1. Referral center characteristics

Three of the four HPN centers (Clichy, Lyon and Rouen) were created in 1984. The third one (Bordeaux) was created a few years later in 1993. Since their opening, almost 1800 patients treated with HPN were followed, including about 761 women of whom 304 of childbearing age (18–45 years old).

3.2. Patient demographics

Fifteen patients corresponding to a total of 21 pregnancies were included over the study period. Nine patients were followed in Clichy, three in Rouen, two in Lyon and one in Bordeaux. Patient demographics are summarized in Table 1.

The median age at HPN initiation was 19 years (IQR: 12 years). One patient (P2) died in 2008 due to complications of the underlying disease. All remaining patients were still treated with HPN at the end of the study.

Seventy three percent (n = 11) of patients were totally autonomous for HPN care, and only 13% (n = 2) had no autonomy at all patients were considered autonomous if they were able to carry out PN connection and disconnection by themselves.

IF etiologies are also reported in Table 1: 73% (n = 11) of patients had SBS (2 had terminal enterostomy, 3 had jejuno-colic anastomosis and 6 had jejuno-ileal anastomosis) and 27% (n = 4) of patients had ostomies.

Regarding previous HPN complications, 53% (n = 8) of patients had liver abnormalities, 73% (n = 11) had history of catheter-related infections and 47% (n = 7) history of catheter-related thrombosis.

3.3. Pregnancy characteristics

Among the 15 patients, there were 21 pregnancies over the study period. Most patients (n = 10, 67%) had only one pregnancy, 4 patients (27%) had 2 pregnancies, and only one patient (6%) had 3 pregnancies. The first pregnancy in our cohort occurred in 1992. In subsequent years, the number of pregnancies increased gradually. Only 2 pregnancies occurred before 2000, then 5 occurred between 2000 and 2010, and since 2010, 14 pregnancies were observed. None of the patients had had a pregnancy before IF diagnosis. As seen in Table 1, the median age at the occurrence of the pregnancy was 27 years, and the median HPN duration before pregnancy was of 8 years, with a maximum of 28 years. Two of the pregnancies occurred after *in-vitro* fertilization.

Delivery occurred after a median amenorrhea duration of 37 weeks (33–41 weeks, IQR: 2), and 55% of deliveries occurred prematurely. The main type of delivery was cesarean section (n = 14, 67%), among them, 9 were programmed (3 before 37 weeks of amenorrhea) for anoperineal lesions, uterine malformation of scarred uterus, and 5 were carried out in emergency (all during labor, 2 before 37 weeks of amenorrhea). One pregnancy was interrupted at 36 weeks of amenorrhea for intra-uterine death.

3.4. Maternal complications during pregnancy

Maternal complications were experienced by 67% of patients (n = 10) during pregnancy, corresponding to 67% of pregnancies. These complications are reported in Table 2.

The 2 postpartum hemorrhages occurred in patients with chronic intestinal pseudo-obstruction (CIPO). All disease-related complications corresponded to the appearance of a subocclusive syndrome in patients with CIPO or Crohn's disease. Among HPN-related complications, 5 were catheter-related infections (1 complicated by endocarditis and 1 by catheter-related thrombosis), one patient had cholestatic hepatopathy and one had ascites.

Two CIPO patients had particularly complicated pregnancies. The first patient had multiple severe sepsis (pneumonitis, pyelonephritis and vulva cellulitis) leading to hospitalizations in intensive care unit and a pseudo-occlusive syndrome treated medically.

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