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Managing Home Parenteral Nutrition during Pregnancy in Patient with Short Bowel Syndrome: Case Report of a Successful Experience in Sabah, Malaysia

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ABSTRACT

Short bowel syndrome (SBS) is one of the pathophysiological mechanisms of chronic intestinal failure (IF) which warrants long-term parenteral nutrition (PN). Despite the common use of PN, there has been few literature on PN use for IF patients since before pregnancy. Our patient was diagnosed with gangrenous bowel secondary to congenital malrotation in 2015. Extensive bowel resection resulted in SBS requiring lifelong home PN. In February 2020, patient discovered her pregnancy and PN pharmacists were informed. Energy from PN was increased with addition of multivitamins, trace elements (TEs) and electrolytes tailored to weekly blood results. At gestation week 34, 24-hour urine urea test showed positive nitrogen balance of +3g/day, demonstrating adequate supply from PN. At gestation week 37, a small-for-gestational-age female infant was delivered via spontaneous labour. Apgar score demonstrated excellent condition and the infant managed to thrive subsequently. Patient's pre-pregnancy underweight status and insufficient weight gain throughout pregnancy are believed to have put her at higher risk for low-birth-weight infant. Vitamins and TEs supplementation generally met the recommendation for pregnancy, which is however established for oral/enteral intake. This case report proved that successful pregnancy is possible in SBS patients on HPN with close monitoring and effective communication with patient.
Keywords: *Home Parenteral Nutrition, Nutritional Support, Pregnancy, Short Bowel Syndrome, Case Report*

Introduction

Chronic intestinal failure (IF) is a long-lasting decrease of gut absorptive function in metabolically stable patients who require intravenous supplementation in the form of home parenteral nutrition (HPN) over months or years (Pironi *et al*., 2015; Bond *et al*., 2017). Short bowel syndrome (SBS) is one of the main pathophysiological mechanisms of chronic IF, accounting for 75% of adult HPN in Europe (Pironi *et al*., 2015). SBS may be the consequence of congenital malformation or extensive surgical resection of malfunctioning or diseased bowel (Pironi *et al*., 2015). In patients with SBS, chronic IF reversibility and

weaning off from HPN are possible if intestinal adaptation takes place successfully, especially with colon continuity intact (Pironi *et al*., 2015). However, it is unlikely for patients with less than 60cm of small bowel remnant as well as removed ileum, ileocecal valve and colon to wean off from IVS (Medscape, 2021). In this case, lifelong HPN is usually warranted.

Pregnancy may involve multiple physiological changes especially in the form of increased nutritional requirements (Carrasco-Rojasa *et al*., 2016). PN use in pregnancy for conditions irrelevant to IF such as hyperemesis and anorexia nervosa has been commonly reported (Smith *et al*., 2022). However, there has been limited literature on the use of long-term PN throughout pregnancy for indications with IF involvement, including SBS (Carrasco-Rojasa *et al*., 2016). In Malaysia, while HPN program has been endorsed following increased knowledge and experience, HPN during pregnancy has not been reported thus far. This case report is illustrating the first successful pregnancy outcome of HPN in Sabah state from a nutritional perspective.

Case Presentation

Patient was a 24-year-old female when she was diagnosed with gangrenous bowel secondary to congenital malrotation back in December 2015, necessitating extensive bowel resection from duodenum D4 to proximal transverse colon. She has no family history of malignancy and bowel diseases. With only 15cm of small intestine remaining, SBS was confirmed requiring lifelong PN. HPN was started after being discharged and administered via chemoport, which was eventually changed to peripherally inserted central catheter (PICC). Simultaneously, oral intake was encouraged as tolerated. Her requirements were calculated based on ideal body weight (IBW), which was 50.1kg.

In February 2020, patient discovered her third pregnancy around gestation week 8, after her first uneventful pregnancy back in 2013 and an episode of induced abortion following her diagnosis in 2015 (obstetric history acronym G3P1A1). Upon presenting for her first prenatal visit, she was underweight at 41.8kg with body mass index (BMI) 17.4kg/m2. Energy provision from PN was then increased from 27.4kcal/kg/day to a maximum of 37.6kcal/kg/day in volume ranging from 1.5 to 2L (Table 1). Multivitamins, trace elements (TEs) and electrolytes were added into each PN bag, with amount (Table 2) tailoring to weekly blood investigation results including serum electrolytes and liver function tests (Fig. 1-3). In addition to parenteral infusion, patient was compliant to her daily oral folic acid 5mg, ferrous fumarate 200-350mg, ascorbic acid 100mg and one tablet of Vitamin B complex throughout pregnancy. HPN was administered 6 days a week with one PN-free day for better quality of life.

Table 1: Daily energy, macronutrients and lipid:carbohydrate (L:C) energy ratio during each trimester, in comparison with European Society of Parenteral and Enteral Nutrition (ESPEN).

Trimester	Energy (kcal/kg/day)	Protein (g/kg/day)	Carbohydrate (g/kg/day)	Lipid (g/kg/day)	L:C Energy Ratio
ESPEN Guideline for Adult HPN (not accounting for pregnancy)					
N/A	20 - 35	0.8 – 1.0	3 - 6	≤1	40:60
Patient's intake from HPN					
1	27.4 - 37.6	1.2 - 1.7	3.2 - 4.3	1.0 - 1.3	42:58 or 46:54
2	37.6	1.7	4.3	1.3	42:58
3*	25.1 - 37.6	1.1 - 1.5	2.9 - 4.3	0.9 - 1.3	42:58 or 46:54
*Provision in third trimester decreased towards delivery due to sepsis and catheter-related complications.					

Table 2: Comparison between Recommended Dietary Allowance (RDA) for pregnancy (Medscape 2018) and patient's daily intake from parenteral sources.

	RDA for pregnancy	Patient's daily intake from parenteral sources		
		Vitamins	Trace Elements	Parenteral Nutrition
Fat-soluble Vitamins				
Vitamin A	**770mcg**	1050mcg		
Vitamin D	**5mcg**	5.5 mcg		
Vitamin E	**15mg**	10.2mg		
Vitamin K	**90mcg**	N/A		
Water-soluble Vitamins				
Vitamin C (ascorbic acid)	**85mg**	125mg		
Vitamin B1 (thiamine)	**1.4mg**	3.51mg		
Vitamin B2 (riboflavin)	**1.4mg**	4.14mg		
Vitamin B3 (niacin)	**18mg**	46mg		
Vitamin B5 (panthotenic acid)	**6mg**	17.25mg		
Vitamin B6 (pyridoxine)	**1.9mg**	4.53mg		
Vitamin B7 (biotin)	**N/A**	69mcg		
Vitamin B9 (folate)	**400-600mcg**	414mcg		
Vitamin B12 (cobalamin)	**2.6mcg**	6mcg		
Trace Elements (TEs)				
Calcium	**1000mg**			3200-3700mg
Phosphorus	**700mg**			700-1160mg
Iron	**27mg**		0.55mg	
Zinc	**11mg**		2.5mg	9.9-13.2mg

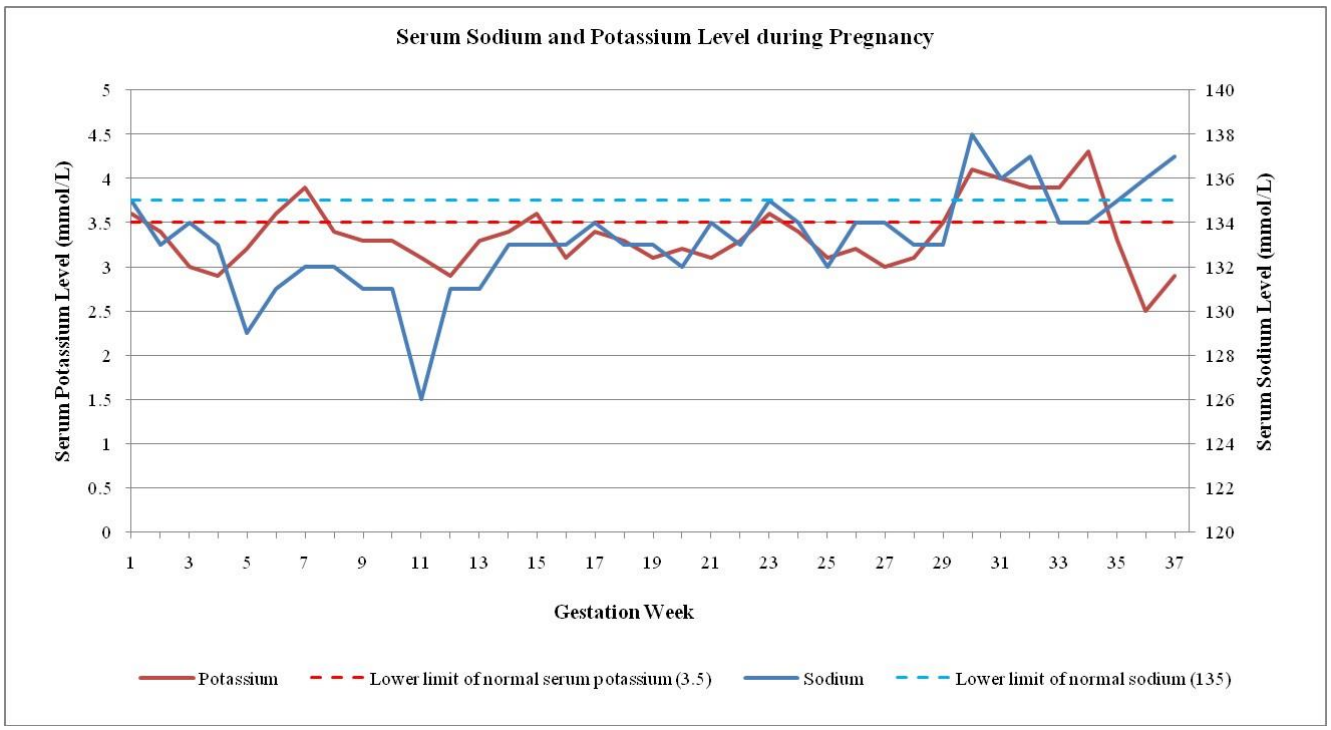

Figure 1: Patient's serum sodium and potassium level during pregnancy.

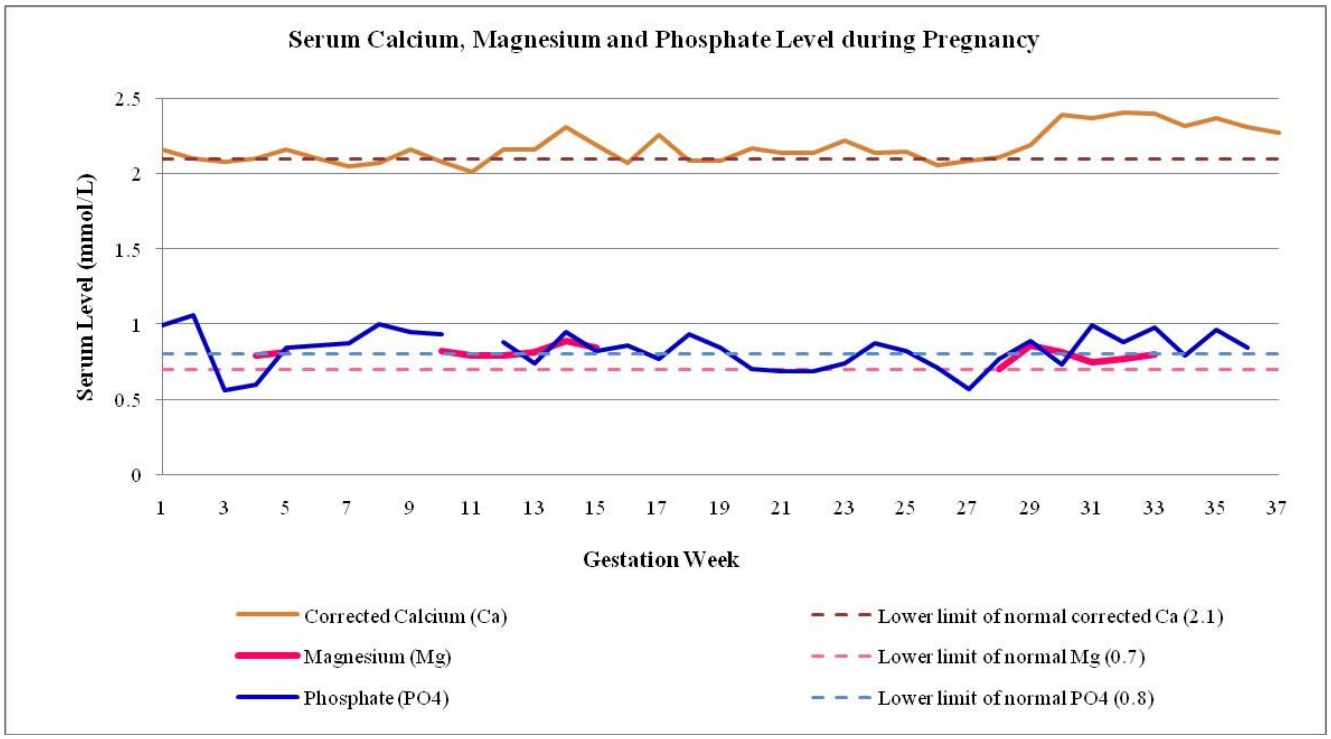

Figure 2: Patient's corrected calcium, serum magnesium and phosphate level during pregnancy. Calcium and magnesium level were in range most of the time. Multiple asymptomatic hypophosphatemia resolved without complications following addition of phosphate into PN bags up to permissible maximum.

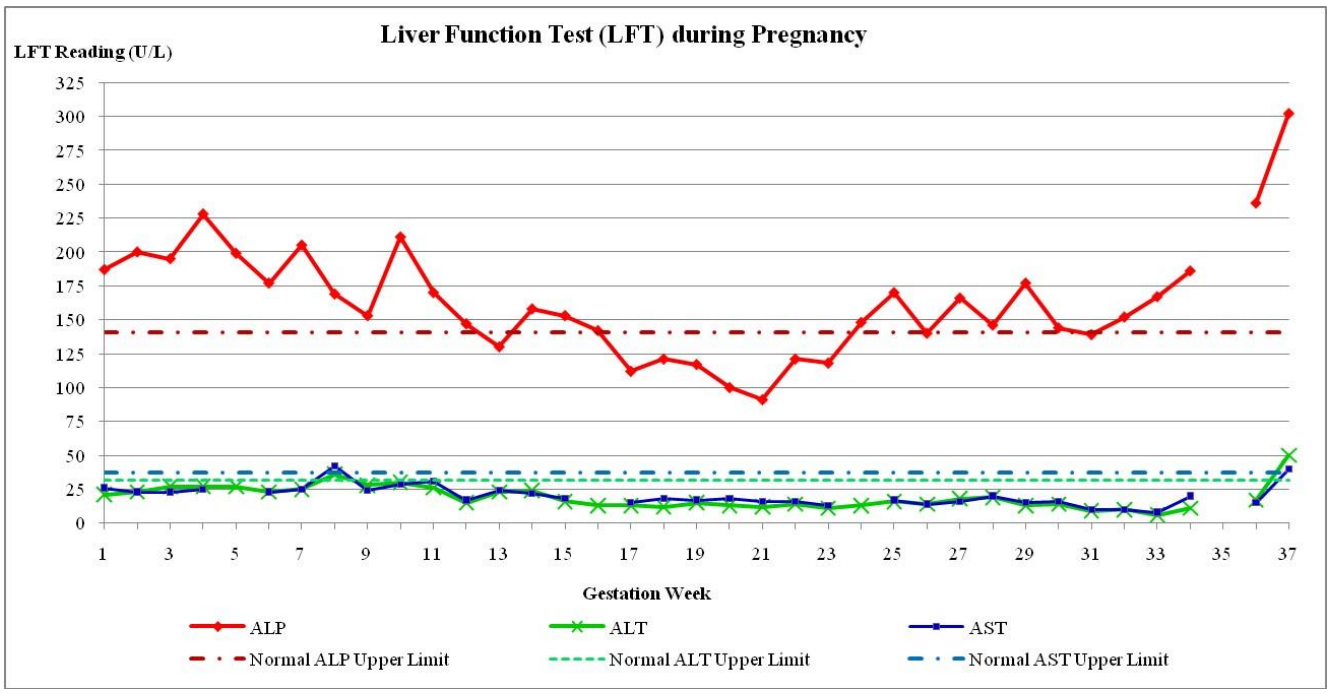

Figure 3: Patient's liver function test (LFT) during pregnancy. ALP: Alkaline Phosphatase; ALT: Alanine Transaminase; AST: Aspartate Aminotransferase. ALT and AST were in range while ALP was above upper limit (UL) most of the time, and peaked at 302U/L right before delivery.

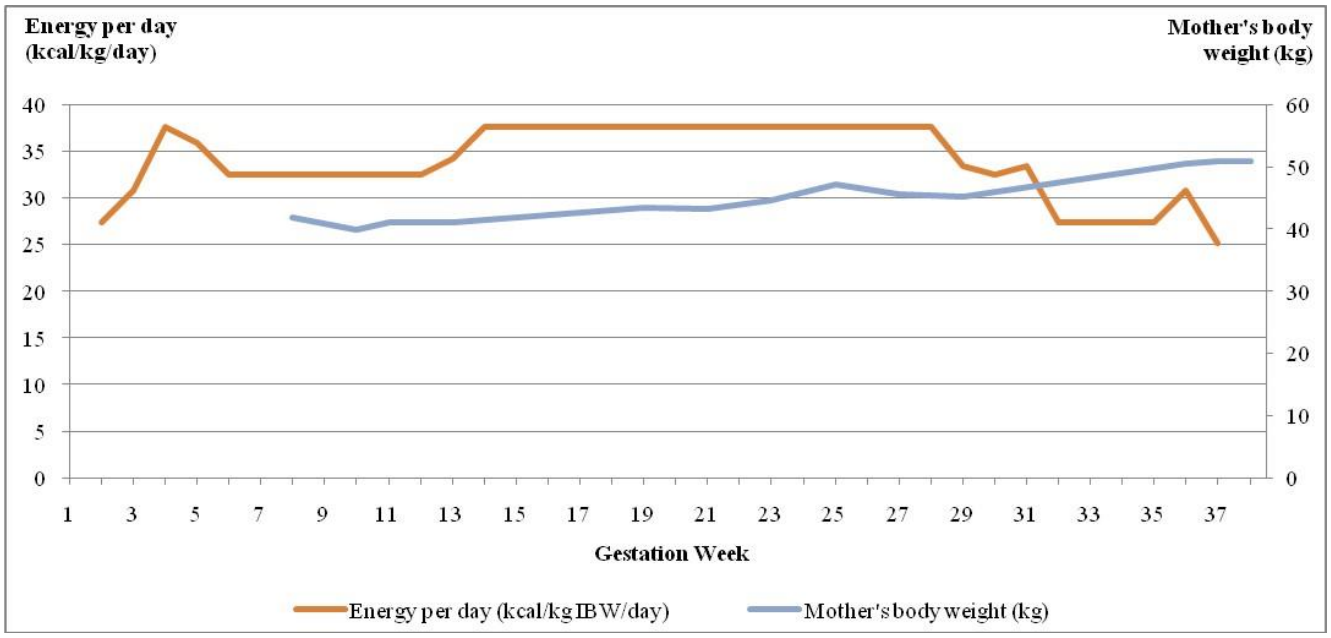

Figure 4: Energy provision and patient's weight changes throughout pregnancy.

Her pregnancy progressed normally until gestation week 30 when patient developed sepsis due to catheter infection. She was admitted for one month and her calorie provision was adjusted according to concurrent medication and fluid input in ward. Despite the complication, she still managed to gain 9.2kg and achieved maximum 51kg close to delivery (Fig. 4). At gestation week 37, she delivered a small-

for-gestational-age (SGA) female infant via spontaneous labour, with weight 2.35kg (1.7th centile), length 44cm (0.3th centile) and head circumference 31cm (0.8th centile). Nevertheless, Apgar score at 1 minute was 9 out of 10, indicating excellent condition without complication. The infant developed normally and managed to catch up on growth, with weight, height and head circumference on the 87.7th, 9.5th and 54.8th centile respectively at 9 months old.

Discussion

As per Table 1, PN provisions to our patient were similar to guideline. Since the recommendations do not account for increased needs of pregnancy, our provisions above the upper limits are justifiable. Calories during pregnancy should be increased starting second trimester, which aligned to our practice (Medscape, 2021). Despite the reduced energy intake following complications in third trimester, 24-hour urine urea test at gestation week 34 showed positive nitrogen balance of +3g/day (normal: -5 to +5g/day) with constant weight gain (Dickerson, 2016). However, her 9.2kg gain was still below the recommendation of the National Academy of Medicine (12.5-18.0kg) to reduce risk of low-birth-weight (LBW) infant (Medscape, 2018). Greater increment would have been anticipated if calories were not reduced. Besides, pre-pregnancy BMI may also affect birth weight independent of pregnancy weight gain (Medscape, 2018). Mothers underweight at baseline including our patient are at increased risk for LBW and SGA babies. Nevertheless, it is a relief that SGA status does not affect the infant's subsequent growth in life. Although HPN has been associated with increased likelihood of preterm deliveries (Caruso *et al*., 1998), our patient had a term delivery at gestation week 37, suggesting its possible link with the underlying disease instead of PN insufficiency.

Absorption of electrolytes takes place in all parts of small and large intestine (Pironi *et al*., 2015). Substantial bowel removal such as our case leads to inefficient absorption, as reflected by the dynamic fluctuations of serum levels in Fig. 2 and Fig. 3. This warrants frequent monitoring for appropriate electrolyte addition into PN according to the latest blood results, as well as timely management in case of abnormally low readings.

Most of the vitamin supplementation met the pregnancy recommended dietary allowance (RDA). Folic acid provision was especially highlighted due to the concern of neural tube defect (NTD) in newborns. The absence of NTD in our case demonstrated sufficient folic acid provision throughout and since before conception. However, it is noteworthy that these RDA recommendations are established for oral/enteral intake generally with less than 100% bioavailability. For patients given intravenous supplementation, parenteral absorption occurs at a greater extent (Sriram and Lonchyna, 2009). Due to the peculiarity of such cases, RDA for parenteral intake of vitamins has not been established.

Iron is the TE of interest in pregnancy, as inadequacy may cause anemia and detrimental consequences on infants. Since only about 10% iron is absorbed orally, pregnancy RDA targets final 3mg absorption (Medscape, 2018). Parenterally, our patient acquired complete 0.55mg daily. Orally, ferrous fumarate 200-350mg (elemental iron 66-115mg) was equivalent to 6.6-11.5mg absorption in normal subjects. However, as duodenum is the primary absorption site, absorption for our patient with only half duodenum remaining is questionable (Gulec *et al*., 2014). Contradictorily, it is postulated that iron loss via exfoliation of intestinal mucosa is reduced in SBS patients due to shorter intestine (Gulec *et al*., 2014). Accounting for the reduced absorption coupled with reduced loss, iron supplementation for our patient was possibly sufficient as reflected by the continuously optimal serum haemoglobin between 11.5 and 13.0g/dL (normal: 12-15g/dL), normal serum ferritin of 81.9ng/mL (normal: 4.63-204.44ng/mL) and the absence of both maternal and foetal complications.

The strength of this case report is that it provides an insight into a successful management of HPN to cater for the increased needs of pregnancy, which is currently lacking established guidelines due to the paucity of similar incidences. This case report therefore contributes to the repertoire of successful pregnancy cases dependent on PN. However, due to the limitation of test reagents available in our setting, more detailed tests for serum levels of vitamins and TEs were unable to be performed. This made monitoring and adjustment of these additives difficult.

Conclusion

This case report proves that successful pregnancy is possible in SBS patients on HPN, with proactive close monitoring and careful planning on the PN supplementation tailoring to oral intake. It is also prudent to ensure that patient can easily access to the Nutrition Therapy Team (NTT) including pharmacists, especially in the event of any complication to allow timely intervention.

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