The challenges of managing and following-up a case of short bowel in eastern europe

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1. Introduction

We followed an individual case of short bowel syndrome (SBS) and tried to illustrate what children and parents had to go through in a developing country with a less financed healthcare system. These cases are rare, complex and often not considered a priority by healthcare planners in government. Medical records were gathered from 3 different hospitals in 2 different countries and this case was followed up from November 2013 to February 2016. Patient's weight and height were regularly monitored, while hypercaloric and hyperproteic diet were structured to improve nourishment and to gain weight. The patient initially was placed on total parenteral nutrition (TPN), then followed by parenteral nutrition (PN) combined with enteral feeding, and later progressed to just enteral feeding [1]. The complex and complicated nature of this case after enterectomy coupled with life threatening enterocolitis with salmonella infection 2 years after surgery were all part of the problems we encountered. Most of these patients die because of inadequate monitoring system due to the lack of healthcare funding. Parents had to seek extra medical help for their child from the West. In order to reduce hospital stay and cost, the parents were involved and trained on how to continue patient's healthcare man-

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Table 1
Laboratory test- short bowel syndrome (SBS).

| CBC | Post-operation I | Pre-operation II | Post-operation III | 1-day | 2-day | 3-day | 8-day | 12-day | 15-day | 19-day | 21-day | Normal values |
|-----|-----------------|-----------------|-------------------|-------|-------|-------|-------|--------|--------|--------|--------|----------------|
| WBC | 25.34 x 10⁹      | 30.12 x 10⁹     | 19.48             | 12.85 | 10.23 | 15.23 | 17.21 | –      | –      | –      | –      | 3.4–9.5 x 10⁹/L |
| RBC | 3.25 x 10¹²      | 3.56 x 10¹²     | 3.48 x 10¹²       | 3.38  | 3.78  | 3.95  | –     | –      | –      | –      | –      | 4.2–5.1 x 10¹²/L |
| HGB | 10.8             | 10.1            | 10.5              | 9.8   | 11.5  | 11.4  | 9.5   | 9.7    | 11.4   | 35.8–42.4 | 12.6–14.0 g/dL |
| HCT | 32.4             | 30.9            | 31.5              | 30.9  | 28.6  | 33.5  | 32.7  | 33.1   | 28.4   | –      | –      | 33.1–36.0% |
| PLT | 6,89,000         | 9,000,000       | 6,67,000          | 6,23,000 | 524,000 | 5,38,000 | 5,46,000 | –   | –      | –      | –      | 150–450 x 10⁹/L |
| Neut%| 91.2             | 83.9            | 91.7              | 84.1  | 84.2  | –     | 44.4  | –      | –      | –      | –      | 1.50–5.0 x 10⁹/L |
| Lymph%| 4.2            | 4.9             | 4.3               | 4.5   | 4.3   | –     | –    | 57     | 62     | –      | –      | 0.00–0.8 x 10⁹/L |
| Mono%| 1.9             | 1.8             | 1.6               | 1.5   | 1.4   | –     | 2.5   | –      | –      | –      | –      | 0.00–0.8 x 10⁹/L |
| Eos | 0.3             | 0.2             | 0.1               | 0.1   | 0.3   | –     | –    | –      | –      | –      | –      | 0.0–0.8 x 10³/L |
| CR | 113              | 112             | 112               | 114   | 113   | –     | 111   | 112    | –      | –      | –      | 112–108 mmol/L |
| Creatinine | 210          | 190             | 141               | 1.2   | 0.10  | 8     | 6     | 4      | N      | 1.21   | <0.10  | 0.0–0.7 mg/dL |
| Urea | 2.0             | 2.2             | 2.1               | 2.0   | 2.0   | 2.2   | 2.2   | N      | 2.3    | 2.2    | –      | 2.4–6.0 mg/dL |
| Amylase | –             | –               | –                 | –     | –     | –     | –     | 293    | 196    | –      | 78     | 54 |
| Lipase | –             | –               | –                 | –     | –     | –     | –     | 156    | 83     | –      | –      | 12–70 U/L |
| ALT | 45              | 58              | 108               | 99    | N     | 200   | 42    | 58     | 41     | 60     | 58     | 7 to 56 units per liter |
| AST | 26              | 39              | 43                | 41    | N     | 52    | N     | N      | 56     | 45     | –      | 10 to 40 units per liter |
| Blood sugar | 92           | 89              | 130               | 110   | 116   | 120   | 123   | N      | 134    | 127    | 90     | 100–125 mg/dL |
| Ferritin | –             | 58              | 314               | –     | –     | –     | –     | 58     | 27     | –      | 58     | 11 to 307 nanograms per milliliter |
| Transferrin saturation | – | 15 | – | – | – | – | – | 15 | 12 | – | 15 | 15–45% |
| CCT | 98              | 87              | 79                | 84    | N     | 80    | 112   | 109    | 67     | 60     | 52     | – |
| PCR | 58              | 65              | 1,83,83           | 150   | 86.6  | 120   | N     | 3      | 3.2    | 2.5    | 1.9    | 40–60 IU/mL |
| PCT | 0.74            | 0.32            | 9.43              | 4.77  | –     | –     | –     | –      | –      | –      | –      | <0.15 mg/ml |
| Serum protein | 74          | 60              | 22                | 4.5   | 1.66  | N     | N     | 6      | 2.3    | 2.5    | 1.9    | 64–83 g per liter (g/L) |
| Creatinine | 24           | 53              | 13                | 30    | 29    | 65    | 32    | 40     | 44     | 42     | 56     | 60–180 microg/dl |
| Cholesterol | –           | –               | –                 | –     | 15    | –     | N     | –      | –      | –      | –      | <200 mg/dl |
| Triglycerides | –          | –               | –                 | –     | 56    | –     | N     | –      | –      | –      | –      | <150 mg/dl |
| Thyroid values | N       | N               | –                 | –     | –     | –     | –     | –      | –      | –      | –      | – |
| Urine test | –          | –               | –                 | –     | –     | –     | –     | –      | –      | –      | –      | – |
| Urine specific gravity | –      | 1.033           | 1.010             | 1.010 | –     | 1.013 | –     | N      | –      | –      | –      | negative |
| Leukocyte | –           | –               | –                 | –     | –     | –     | 20    | –      | –      | –      | –      | – |
| Ph | 5.6             | 5               | 5                 | –     | –     | –     | –     | –      | –      | –      | –      | 4.5–8 |
| RBC (erythrocyte) | –      | –               | –                 | 5280/microl | –  | – | – | – | – | – | – | <3 HBC/hpf |
| Protein | –           | –               | –                 | –     | –     | –     | –     | –      | –      | –      | –      | >150 mg/dl |
| Stool test | –       | Pseudomonas aeruginosa | – | – | Klebsiella pneumonia | – | – | – | – | – | – | Enterococcus sp.+++ +E. Coli+++ Salmonella |
| Calprotectin | –     | –               | –                 | –     | –     | –     | –     | <100   | –      | –      | –      | Negative |
| Occult bleeding | –     | –               | –                 | –     | –     | –     | –     | +      | –      | +      | –      | <50 |
| Abdominal R-ray with barium | –     | –               | –                 | –     | –     | –     | –     | O short stoma area stenosis | – | – | – | negative |

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CT

- Free abdominal fluid in Douglas pouch and between the intestinal loops; Air fluid levels and intestinal distention present. No paraperitoneum.

Abdominal ultrasound

- Subileus/ileus with sufficient fluid between the intestinal loops. No intra-abdominal abscess

Irigoscopy (Barium enema)

- Retrograde transit within the transverse colon and the ileum, anastomotic stenosis was excluded

Soft tissue ultrasound/Doppler venous ultrasound of the neck and subclavian vein

- Very well visualized jugular veins and without obstruction

Blod gas

| Ph  | pO2  | pCO2 | Na  | K   | Ct+ | Mg+ | Phosphate | Ca  | HCO3 | BE  | Lac | pO2  | 1/L  | 1/L | 1/L | 1/L | 1/L |
|-----|------|------|-----|-----|-----|-----|-----------|-----|------|-----|-----|------|------|-----|-----|-----|-----|-----|
| 7.24| 46   | 38   | 140 | 6.3 | –   | –   | 7.32      | 0.91| 19.7 | 7.7 | 2.5 | 7.32 | 7.31 | 7.36 | 7.24 | 7.35–7.45 |
| 7.30| 45   | 67   | 133 | 5.6 | 4.1 | 0.45 | –         | 1.28| 15   | 6.4 | 2.6 | –    | –    | –   | –   | –   |
| 7.30| 45   | 67   | 133 | 5.6 | 4.1 | 0.45 | –         | 1.28| 15   | 6.4 | 2.6 | –    | –    | –   | –   | –   |
| 7.41| 47   | 65   | 133 | 4.1 | 0.45 | –    | –         | 1.28| 15   | 6.4 | 2.6 | –    | –    | –   | –   | –   |
| 7.32| 7.31| 7.36 | 7.24| 7.35–7.45| 38–42 mmHg | 75–100 mmHg | 135–144 mg/dl | 3.6–5.2 mg/dl | 97–106 mg/dl | 1.1–2.3 mg/dl | 3.2–5.7 mg/dl | 8.8–10.4 mg/dl | 21–28 mmol/L | 0–50 mg/L | 0.5–2.2 mmol/L | 13.5 to 17.5 g/dL | 0–50 mg/L | 0–50 mg/L | 200–900 picograms per milliliter (pg/mL).
Short, medium and long time prognosis are correlated with the underlined complications; local or systemic.

### 2. Case presentation

A 6 years old female patient arrived at our emergency unit in a serious condition: abdominal pain, fever, nausea, bilious vomiting, no passage of stool, abdominal distention, tachycardia and no history of surgery. Abdominal ultrasound and CT scan showed signs of intestinal obstruction. Emergency surgery was performed while common mesenteriy, intestinal obstruction due to volvulus with extensive ischemic necrosis of the ileoceccolic intestinal loop was discovered. Intestinal resection followed by side to side ileocolic anastomosis was performed with just 80 cm of the intestine left and without ileocele valv. The post-operation status of patient was not encouraging. Pathology result: An ulcerative ileojejuncolic mucosa (Table 3). Blood pressure: 70/50 mmHg, urine output: 400 ml/day, gastric tube: 300 ml of blood strained fluid, central venous catheter was placed. Post-operation lab test; acidosis and anemia (Table 1). Parent requested transfer to a regional hospital after 24 h. Upon arrival the patient continued with total parental nutrition, antibiotics and antifungal agents. The patient remained in a severe catabolic state, metabolic acidosis, severe malabsorption and loss of nutrients, water and electrolytes through diarrhea (more than 10 stools/day), refusal of enteral feeding and signs of intestinal obstruction (seen on ultrasound and CT scan). Medial laparotomy was performed 7 days after, and ischemic necrosis of the anastomatic joint was discovered and resected followed by end to end anastomosis with just 70 cm of the intestine left. Meanwhile the increased bilious secretion was not encouraging. Despite all the effort to correct electrolyte and acid base imbalance, the patient developed intestinal failure, circulatory, renal and respiratory insufficiency. The antibiotics regimen were changed. The patient continued to show signs of paralytic ileus for several days, maldigestion, malabsorption, diarrhea, hydric-electrolyte imbalance and abdominal distention, and repeated ultrasound and CT scan showed signs of intestinal obstruction. Exploratory laparotomy was performed seven days after the second surgery. Intestinal adhesion was discovered and lysed; the anastomotic joint was intact, however, the patient’s postoperative status did not improve. The patient remained febrile; the bilious aspirate (1000 ml/day), watery diarrhea (7–10 stools/day), metabolic imbalance, blood culture and peritoneal fluid culture were negative, stool culture was positive for pseudomonas aeruginosa, the intestinal failure continued. The pathology report after the second surgery demonstrated inflammatory changes, villous blunting (Table 4). Laboratory analysis showed anemia, reactive thrombocytes, inflammatory infectious syndrome (Table 1). The parents requested overseas transfer. The patient arrived overseas in a catabolic state (bilious vomiting, metabolic acidosis, pyrexia, confusion, speechlessness). Vitamin A, D, and E deficiency, selenium deficiency and axial hiatal hernia, gastro-esophageal reflux and hepatomegaly were also present. An upper GI study showed good contrast passage with 2 suspicious areas of possible stenosis. Swab showed multi-resistant germ (Klebsiella pneumonia) requiring antibiotics. PN and balancing of the electrolyte levels, fresh frozen plasma and albumin helped achieve anabolism; The patient was mobilized with the help of physical therapy. However, the patient continued to lose large amount of bilious fluid requiring further investigation by upper and lower endoscopy (gastroduodenoscopy and sigmoidoscopy) and biopsy. Visceral hernia and intestinal adhesion were solved through a medial laparotomy followed by peritoneal lavage (for colon and abdominal wall acesis), while two stomas (jejunostomy and colostomy) were placed at the stenotic sites to improve nutrition. The stomas were closed after 6 weeks. Correlated therapeutic management helped to reduce gastric secretion, improve digestive efficiency (against diarrhea, antibiotics, eubiotics etc). Bicarbonate, potassium, calcium and vitamin D deficiencies were corrected. Enteral feeding was gradually introduced and tolerated (dietary food with low lactose and fructose content). The parent’s were trained to continue PN at home especially at night, smokkafien peripheral emulsion infusion for 18 months. Patient’s condition improved and stool frequency reduced.

### Table 2

| Complication of SBS (case report) | PRIMARY ANATOMOSIS |
|----------------------------------|---------------------|
| 1. Peristomal skin excoriation    | Wound infection (local acesis) |
| 2. Granulomatosis                 | Intestinal obstruction (+) |
| 3. Stomal retraction              | Anastomotic leak, |
| 4. Prolapse                       | Anastomotic stenosis (+) |
| 5. Wound dehiscence              | Anastomotic ischemia/necrosis (+) |
| 6. Wound infection (local acesis) | Wound dehiscence |
| II. Systemic (intestinal stoma & primary anastomosis) | |

### Table 3

| Pathology result (First Surgery). | MACROSCOPIC ASPECT | MICROSCOPIC ASPECT |
|----------------------------------|--------------------|--------------------|
| COLOR: Dark pinkish jejum, ileum, hemorrhagic serosa and leucorhea (greenish intraluminal bowel content). | An ulcerative ileojejuncolic mucosa, with intestinal villi and ulcerative lesions. | Lymphoid follicles, lymphocytic infiltration of submucosa and mucosa, intestinal dilatation, thrombotic vessels of the submucosa, and intestinal wall lesions mimicking enterocolitis follicles overlapping the ischemic lesions. |
| Dark pinkish subileal ganglion group. | | |
| SIZE: 80 cm ileum and jejum length, 4/3 cm lymphatic ganglion at ileocolic angle. | Approximately 16 mesenteric lymphatic ganglia included in the block had the aspect of chronic lymphadenitis and with areas of hemorrhagic and thrombotic patches. | Also present was thrombotic mesenteric vessels and an omentum without polymorphic modification. |
| Subileal ganglion group of 0.5–2 cm. | | |
| Appendix of 10 cm and an omentum of about 12/15 cm. | | |

### Table 4

| Pathology result (Second Surgery). | MACROSCOPIC ASPECT | MICROSCOPIC ASPECT |
|-----------------------------------|--------------------|--------------------|
| COLOR: Intestinal lumen; ash color with brownish elastic areas. | Increased villous height, crypt depth, intestinal epithelial hyperplasia, intestinal fragments with large surface of thrombotic extravasation. | |
| SIZE: Intestinal tubular fragment of about 8.3 cm. | Hyperemic vessels with different stages of thrombus and highly infiltrated polymorphic inflammation (Ischemic modification). | |
Discussion

The term short bowel syndrome is a malabsorption state that occurs after the resection of a large portion of the small intestine [2]. This can also mean the need for prolonged PN as a result of intestinal failure. These patients require long term hospitalization and PN [3]. The degree of malnutrition depends on the remaining intestinal length, which is crucial in determining bowel functional capacity. With just 70 cm of the intestine left and no ileocecal sphincter capacity, severe malnutrition, watery diarrhea and metabolic acidosis in the acute phase of the disease was life threatening. This was followed by the adaptation phase from 2 to 4 days after bowel resection, which lasted for months. The second surgery is debatable as some experts argue that this is a paralytic ileus state often seen in the adaptation phase, some surgeons may prefer intestinal stoma instead of anastomosis at this stage. Stoma versus primary anastomosis is also debatable, though intestinal stoma had a better result in this case. Strategies that slow intestinal transit, improve peristaltic function, or enhance mucosal absorption function each has application in the management of SBS [4]. The last phase is the maintenance phase, here the absorption capacity of the intestine is at its maximum. The optimal goal in the management of SBS is to gain full enteral autonomy at the optimal time [5]. Long term TPN can lead to intestinal failure associated liver disease [6]. As enteral feeding was tolerated TPN was gradually weaned. Fluid and electrolyte imbalance were corrected during this process of bowel adaptation. To avoid metabolic bone disease calcium, vitamin D and alkaline phosphate levels were periodically checked. Soluble vitamins (A, D, E, K), and vitamin B12 were monitored closely for deficiency. Food with less osmotic load such as protein and fat helped provide additional stimulant for intestinal adaptation. Adequate PN and later enteral nutrition helped to reduce diarrhea from 10 to 3 stools per day. For maintenance of a good hydration and electrolyte balance normal or half saline, potassium, sodium bicarbonate were supplemented. Continuous sip of oral rehydration solution (ORS) through the day helped maintain a positive fluid balance. Extra intravenous fluid was needed when the enteral route was unable to meet the patient’s nutritional need. Small bowel biopsy in SBS always demonstrate inflammatory changes, villous blunting, while the present of adherent or intracellular bacteria proves the presence of short bowel bacterial overgrowth [7]. Catheter blood stream infection is another source of infection in SBS [8]. Bacterial translocation has been noted in animal models, but data supporting its occurrence in human is limited [9]. Bacterial overgrowth and impaired mucosal immunity puts SBS patients at risk of bacterial translocation [7]. Intestinal bacterial growth is controlled by many mechanisms such as: gastric acidity, pancreatic enzyme activity, enterocyte turnover, normal peristaltic activity and the presence of ileocecal sphincter [10]. These factors are altered in SBS Patients, so bowel dilatation with reduced peristalsis may develop adaptation mechanism to improve enteral adaptation, these factors on the other hand may favor bacterial overgrowth by reducing bowel ability to expel microorganisms. Intestinal endotoxin increases in children without ileocecal valve and can also impair liver function by decreasing body's bactericidal defense mechanism [7]. Elevated d-lactic is responsible for acidosis, this condition is associated with confusion, speech disturbance, and severe metabolic acidosis seen in this case. Growth retardation is as a result of severe metabolic disturbances and impaired immune system.

4. Conclusion

The high cost of managing SBS patients in the developing countries and the complicated nature of this disease limits parent's access to quality healthcare for their children. The main goal in SBS is to achieve intestinal autonomy at the optimal time because prolonged time of intestinal failure can lead to more complications or death. Reduce hospital stay and avoid hospital acquired infections. Reduce hospital cost through parental home healthcare management training.

Compliance with Ethical Standards

This article does not contain any studies with human participants or animals performed by any of the authors. The authors declare that they have no sources of funding. Informed consent was obtained from parents for this article's publication. This work has been reported in line with the CARE criteria and the paper above meets the CARE guidelines: consensus-based clinical case report guideline development [11].
Conflicts of interest
Authors declares no conflict of interest.

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Ethical approval
There is no ethical approval needed for this case.

Consent
Informed consent was obtained from all individual participants (parents) included in the study.

Author contribution
Prof. Dr Eugen Boia. Coordinator.
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