**Patient, parent and professional perception of the use of maintenance enteral nutrition in Paediatric Crohn’s Disease**

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Running head- Perception of maintenance nutrition in Crohn's

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Abbreviations: IBD; inflammatory bowel disease, CD; Crohn’s disease, MEN; maintenance enteral nutrition

**Abstract**

**Aims**-Maintenance enteral nutrition (MEN) is routinely used in Paediatric Crohn’s Disease (CD) to prolong remission although there is limited evidence for efficacy and alack of formal guidelines. This study surveyed patients’, parents’ and professional experience with MEN.

**Methods**-Two questionnaires were developed to survey experience of MEN; i) Patients/Parents (children >10 years of age aimed to complete independently) and ii) Dietitians. Questionnaires were sent to families prescribed MEN after exclusive enteral nutrition (EEN) between 2015-17 (n=77) and dietitians working in paediatric regional centres in UK (n=23).

**Results**- Response rate to the questionnaires was 53% patients, 62% parents and 83% dietitians. Patients/parents reported medical/dietetic advice to be the primary factor affecting compliance, 30% patients reported side effects. Fifty-six percent of patients/58% parents stated a preference for dietary advice rather than MEN. Dietetic responses indicated 79% used MEN after EEN as standard procedure and 79% did not have exit criteria for MEN. Sixty-eight percent perceived the taste was the primary factor affecting patient compliance.

**Conclusion**- Patients’ perception of the usefulness of MEN differs to professionals. This study highlights the extensive practice of MEN after EEN in clinical remission, which may not be nutritionally indicated. Patient preference is for dietary advice rather than MEN.

**Key notes**

* Maintenance enteral nutrition (MEN) is routinely used in Paediatric Crohn’s Disease (CD) during remission but there is little data on patients’, parents’ or professional experience of treatment.
* These data indicate MEN is widely used in paediatric regional centres to prolong remission, despite limited evidence.
* MEN side effects are widely reported and patient perceptions differed to professional opinion. Patient preference is for dietary advice in the management of CD during remission.

**Introduction**

Nutritional compromise and delayed linear growth is common in paediatric Crohn’s disease (CD) at presentation (1). Exclusive Enteral Nutrition (EEN) is widely used to induce remission with continued use of oral nutritional supplements as maintenance enteral nutrition (MEN), given alongside diet to enhance nutritional status and prolong remission(1-3). However, the evidence base for MEN prolonging remission in paediatric CD is limited and there is little consensus on the amount or duration of MEN required (2, 4-7). There are no randomised controlled trials using MEN versus normal diet to prolong remission in paediatric CD patients, with are a limited number of studies which are non-randomised, non blinded and retrospective (4, 5, 7-10) (Table 1). Quantitative pooling of the paediatric studies is not feasible due to the diversity of volumes and duration of MEN used. The most recent and largest study of 58 paediatric CD patients by Gavin *et al* (7) reported no significant difference in length of remission at 12 months between patients receiving MEN compared with normal diet.

MEN is also used clinically to maintain weight gain initiated by EEN (5). However results from our centre demonstrated there was no difference in BMISDS at 12 months between patients who received MEN and those on normal diet, although clearly in a heterogenous non-randomised population there are many potential confounders (7). There is also a paucity of studies on patient tolerance and acceptability of MEN amongst patients.

A Cochrane review reported that oral nutritional supplementation is not well tolerated in paediatric chronic disease due to physical side effects of nausea, bloating and food displacement (11). Adherence to a MEN regimen may therefore be affected, however adherence rates are only measured subjectively in paediatric CD studies on MEN and the impact of daily MEN upon patients’ quality of life is not assessed. To our knowledge, factors influencing patient adherence with MEN and the extent to which MEN is recommended in UK clinical practice to either prolong remission or improve nutritional status, have not been previously described.

As such, the aim of this study was to describe the impact of MEN on daily routine, appetite and factors affecting adherence and to identify patients’ preferred mode of nutritional support during inactive disease. The objectives were to; i) characterise separately, patient (hereafter “child”) and parent perception of the use of MEN and ii) investigate the extent of clinical practice of MEN in regional paediatric IBD centres.

**Methods**

Patients were recruited from the Southampton Children’s Hospital Paediatric IBD service covering the Wessex region.

**1. Child and Parent Questionnaires;**Two separate questionnaires were developed based on a paediatric CD questionnaire by Svolvos (12), one for children and a similar copy modified for parents in partnership with five families of patients with CD at a public and patient involvement event to ensure questions reflected real life experience (Appendix 1 & 2). Study group members and children/parents checked the content validity and readability. Questionnaires were individually addressed and sent to children and their parents. The questionnaires were split into 4 themes (14):

1. Child demographics
2. Practical aspects of the MEN regime
3. Appetite for food since diagnosis
4. Child and parent perceptions of factors influencing adherence to MEN

**2. Paediatric Dietetic Questionnaire**

A health care professional questionnaire was developed based on a dietetic clinical practice questionnaire by Armstrong (13) (Appendix 3). This was distributed to dietitians listed as members of the British Society of Paediatric Gastroenterology and Nutrition (BSPGHAN) Associate Members Group. The dietitians represented the views of the majority of IBD paediatric regional centres in the UK. A reminder email was sent to all dietitians one month later to maximise response rate. The dietitian survey was split into four themes (13):

1. Dietitian’s role and length of experience with paediatric CD
2. Nutrition outcomes for MEN and assessment of estimated energy and protein requirements
3. Practical aspects of the MEN regimen
4. Dietitian perceptions of factors influencing adherence to MEN

Statistics

Summary statistics are presented in raw response counts and frequencies and as a percentage of total responses. Open ended comments were categorised as either positive or negative by the investigators.

Ethical approval

This study was approved and registered as a service evaluation by the University of Southampton Hospitals NHS Trust.

**Results**

**1. Child and Parent Questionnaires**

1. *Child demographics*

A total of 77 children were identified from medical and dietetic records who had been prescribed MEN after EEN between 2015-2017 and were in clinical remission, as defined by physician global assessment, all were sent a questionnaire for completion.. Forty-one children (53%) and forty-eight parents (62%) responded. The median age of the children was 14 years (range 9-18 years), thirty male (63%) and eighteen female (37%). All had been prescribed MEN within the last two years. Twenty-two children (54%) and twenty-eight parents (58%) were no longer taking MEN at the time of the survey.

1. *Practical aspects of the MEN regime*

Twenty-seven children (66%) reported they took Modulen IBD (1-1.5kcal/ml, Nestle) as a nutritional supplement drink alongside normal diet. All 27 children had used Modulen as EEN induction therapy. The remaining fourteen children (34%) switched to Fortijuice (1.5kcal/ml, Nutricia) or Fortisip Compact (2.4kcal/ml, Nutricia) ready-made, smaller volume nutritional supplement drinks. Seventeen children (42%) and thirty-one parents (65%) recalled being asked to take MEN for as long as possible. Sixteen children (39%) and twenty-three parents (48%) recalled being asked to take MEN as two drinks daily before school and before bed. No patients had MEN via nasogastric tube or gastrostomy tube.

1. *Appetite for food since diagnosis*

Ten children (24%) perceived their appetite for food in remission to be excellent (Figure 1+2). Children rated appetite scores higher than parents perceived. Twenty-three children (56%) perceived their appetite had changed since diagnosis citing that the “timing of meals” and “limiting the quantity of food eaten at a meal” was important to them. Thirty parents (63%) reported an increased interest in the quality of food they provided for their family, “making more nutritious home cooked food” and “relying less on processed foods” (Figure 3+4). Half of all children reported certain foods made them feel unwell (Figure 4).

1. *Child and parent perceptions of factors influencing adherence to MEN*

Twenty-six children (63%) and thirty-one parents (65%) reported daily adherence to the recommended quantity of MEN. Twelve children (52%) reported their adherence to MEN was primarily influenced by advice given by the medical and dietetic profession and secondly, “parental nagging” (43%) (Figure 4). Ten parents (36%) perceived that “nagging” was the primary influence on their child’s adherence to MEN, although 8 parents (29%) rated medical / dietetic advice and “feeling of fullness” to be important. In open ended questions, seven children (30%) described bloating and nausea after drinking nutritional supplements which affected their adherence. Seven children (30%) and seven parents (25%) recognised that the taste of the supplement drink was important but not the primary influence in the acceptability of MEN. Six parents (21%) were concerned about stopping MEN in case their child became unwell again, a concern shared by only two children (9%). Only two parents (7%) were concerned about excess weight gained on long term MEN, a concern not shared by children. Two children (9%) admitted forgetting to take MEN, a fact unrecognised by parents. Child and parent open ended responses describing their experience using MEN were recorded (Table 2).

Eleven children (65%) and eighteen parents (82%) perceived that “smaller volumes” of nutritional supplement drinks would have improved adherence (Figure 5). Fourteen children (34%) used smaller volume energy dense drinks as MEN. Eight children (47%) perceived “different flavours” may have assisted adherence. Twenty-three children (56%) and twenty-eight parents (58%) reported they would prefer a nutritional supplement in the form of a food. Examples given by the children and parents were cereal/cereal bar, cake, pizza, “something where the flavour is more like food” or just “nice food”! Thirty-three children (81%) and thirty-three parents (69%) requested a recipe book to help manage CD during relapse and in remission.

**2. Dietitian questionnaire**

1. *Dietitian’s role and length of experience*

Questionnaires were distributed to 23 paediatric dietitians. Nineteen dietitians (83%) from regional IBD centres in the UK responded to the questionnaire, thirteen specialist paediatric gastroenterology dietitians (68%), fourteen had more than 5 years experience working with CD patients (74%).

1. *Nutrition outcomes for MEN and assessment of energy and protein requirements*

Dietetic reported nutrition outcomes for MEN were; growth (63%), weight gain (32%) and improvement of micronutrient status (32%). All dietitians measured weight, height, body mass index (BMI) and two dietitians (11%) measured mid upper arm circumference (MUAC). All dietitians used dietary reference values for energy(25) to estimate patients’ energy requirements. Seventeen dietitians (89%) recommended 25-30% of estimated daily energy requirement as MEN.

1. *Practical aspects of the MEN regimen*

Eighteen dietitians (95%) recommended MEN after EEN primary induction therapy. Fifteen dietitians (79%) reported MEN after EEN was standard procedure in their hospital. Thirteen dietitians (72%) advised patients to take MEN for as long as possible. Nine dietitians (47%) reported the primary indication for using MEN was to prolong remission and three dietitians (16%) used MEN primarily to treat persistent faltering growth. All dietitians used polymeric MEN as first line choice. If a patient with faltering growth refused to take nutritional supplement drinks, nine dietitians (47%) would provide alternative high protein and energy food based dietary advice and four (21%) would consider enteral feeding via NG tube.

Fifteen dietitians (79%) did not have exit criteria for MEN therapy. Four dietitians (21%) used the following exit criteria: “when maintenance medication takes effect, “when growth is consistent”, “growing appropriately” or “when ideal weight is achieved”. Responses to a free text question indicated there was concern regarding recommending MEN as standard practice commenting “there needs to be more robust evidence for MEN in patients who are not nutritionally compromised” and “better evidence for ongoing use of MEN for all patients is required.”

*iv. Dietetic perceptions of factors influencing* adherence *to MEN*

Eight dietitians (42%) estimated that 60% of their patients complied daily with MEN. Thirteen dietitians (68%) perceived the “taste” of the nutritional supplement drink was the primary factor affecting adherence although twelve dietitians (63%) recognised “parental nagging” as important whilst nine dietitians perceived bloating and nausea was influential (47%) (Figure 4). Six dietitians (32%) perceived that child and parent concern about excessive weight gain whilst taking MEN was an issue affecting compliance. Four dietitians (21%) perceived their advice had a significant impact on patient compliance.

Eleven dietitians (58%) assessed adherence to MEN subjectively, using either a cross checking method (42%) or a food diary (26%). Four dietitians (21%) checked with the GP to confirm whether repeat prescriptions had been collected by the family. Twelve dietitians (63%) perceived that “different flavours” of nutritional supplement drinks, including savoury, would have improved adherence the most, whilst five dietitians (26%) perceived frequent reminders from the dietitian would have assisted adherence (Figure 4). Four dietitians (21%) perceived that smaller volumes of MEN would be influential. Two dietitians (11%) suggested that setting an expectation with families at diagnosis that MEN was a long term treatment may improve compliance. One dietitian (5%) perceived the use of recipes for nutrient dense food and drinks would be a useful alternative practice to oral nutritional supplements.

**Discussion**

This study characterised different perceptions between children (young people), parents and dietitians relating to their experience of MEN. Children who responded to the survey were representative of the general paediatric CD population, predominantly male and teenage. We have shown that during inactive disease, many families would prefer dietary advice in preference to MEN as a mode of nutritional support. This is similar to the findings of Svolvos *et al.* (12) who reported families preferred dietary advice to EEN during active disease as it enabled the patients to feel more “normal and part of the family”.

The majority of paediatric IBD regional centres around the UK were represented, with 3/4 dietitians reported MEN to be standard practice after primary induction with EEN. Half stated the principal aim of prescribing MEN was to prolong remission although the evidence underlying the efficacy for this role of MEN in paediatric CD is limited. Several retrospective studies reported no difference in length of remission between children prescribed MEN and those on a normal diet (4, 5, 7). ESPEN guidelines do not support the use of MEN for paediatric CD to prolong remission (6) and ESPGHAN (14) recommend that MEN should be limited to a select group of paediatric patients with mild disease who are not on maintenance medication, limiting use of MEN to a small minority of paediatric CD patients.

Better disease management of paediatric CD in recent years has been fundamental to improving paediatric patients’ nutritional status (15, 16). The incidence of obesity in paediatric CD is now equivalent to that reported in the general paediatric population (17). Although children with CD have a lower lean body mass than their healthy counterparts, this may be more related to the physical inactivity associated with chronic disease than a prolonged poor nutritional intake (18). This study reported that children regained their appetite to prediagnosis levels once in disease remission, therefore may be more capable of maintaining their weight gain and growth through diet alone than previously perceived by healthcare professionals. Recommending MEN to prolong remission when nutritional rehabilitation may already have been achieved with EEN and without anthropometrical entry or exit criteria or assessment of disease severity, may therefore potentially increase the risk of adiposity as seen in other chronic diseases(19). Increased adiposity has been associated with faster disease progression in patients with CD and an increased risk of surgery (20, 21).

This study demonstrated that the side effects of MEN such as bloating and nausea created difficulties with compliance. Families stated a preference for nutritional support as dietary advice instead of MEN. Young people and parents reported an increased interest in their diet after diagnosis, similar to previous findings (22, 23). Engaging young people with CD in the co-design of studies on healthy diet and lifestyle choices may positively impact clinical and nutritional outcomes in adulthood and as a patient led practice may improve quality of life (24).

This study does have limitations. Questionnaires were only distributed to families whose children were prescribed MEN and did not include opinions from children and parents who had not received MEN who may have views on the use of nutritional supplements. One child surveyed was less than 10 years old but all children were deemed suitable for independent survey completion by their parents. Appetite assessment was subjectively measured only, potentially incurring a risk of recall bias. Although local patient perspective on MEN cannot be directly compared with national perspective, nor with dietetic perception, the experience of families in our study is similar to that reported by other patients with chronic disease consuming oral nutritional supplements and by clinical dietitians internationally (11, 25). A lower response rate in our study (53% children and 62% parents) compared with a similar patient questionnaire by Svolvos (71% response rate) may relate to the longer study period of up to 2 years between MEN treatment and questionnaire, compared to 1 year post EEN in his study (12). Half the patients in our study were no longer taking supplements at the time of the questionnaire therefore providing their opinion from memory. It is possible that families most keen to use diet as a means of managing their disease in remission will have returned questionnaires more commonly than those who felt it was not useful.

**Conclusion**

This study reports perspectives of young people and parents on the use of MEN after primary induction with EEN and the personal experience/clinical practice of UK dietitians using MEN. We identified a different perspective between patients, families and professionals on the use of MEN. MEN after EEN was standard practice in the majority of paediatric IBD regional centres with the primary aim of prolonging remission despite little evidence to support this claim. Young people and parents reported difficulties with adherence to MEN and stated a preference for dietary advice. This study advocates that the extensive use of MEN in clinical practice is limited to comply with ESPGHAN recommendations. Patient led care promotes the use of dietary advice as a mode of nutritional support during inactive disease.

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Figure 1. Patient and parent perceptions about appetite in remission

Figure 2. Patient and parent perception of the way they think about food since diagnosis

Figure 3. Patient and parent perception of foods that make them feel unwell

Figure 4. Patient, parent and professional perceptions of factors affecting adherence of MEN

Figure 5. Patient, parent and professional perceptions of strategies to improve adherence to MEN

Table 1. Summary of papers using MEN alongside normal diet to prolong length of remission

Table 2. Child and parent open text comments about their experience using MEN

Appendix 1. Crohn’s Disease and Nutritional Supplements Young Person Survey

Appendix 2. Crohn’s Disease and Nutritional Supplements: Parent Survey

Appendix 3. [The use of oral nutritional supplements in children with Crohn's Disease](https://docs.google.com/forms/d/e/1FAIpQLSfoHZVqKwh3ronJODw_DFPMQ7nUEOiAT_xl29eN8UMqtnwptw/viewform?c=0&w=1&usp=mail_form_link)

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The authors declare no competing interests

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