

Dissertation

Netcoaching: A new and effective alternative to traditional onsite models for supervised tube weaning in infancy

Dissertation submitted by

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Declaration

I hereby declare that this dissertation is my own original work and that I have fully acknowledged by name all of those individuals and organisations that have contributed to the research for this dissertation. Due acknowledgement has been made in the text to all other material used. Throughout this dissertation and in all related publications I followed the guidelines of “Good Scientific Practice“.

Graz,

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Last but not least, my family and my friends deserve my deepest gratitude – thank you for everything!

Abbreviations and Definitions

NG tube	nasogastric tube
PEG	percutaneous endoscopic gastrostomy
ND tube	nasoduodenal tube
NJ tube	nasojejunal tube
MD	Median
SD	Standard deviation
ENS	Enteral nutrition support
GI tract	Gastrointestinal tract
GER-D	Gastroesophageal reflux disease
IA	Infantile Anorexia
TD	Tube dependency
HEN	Home enteral nutrition
HETF	Home enteral tube feeding

List of figures

Figure 1: Placement of a nasogastric tube. Derived from: http://www.macmillan.org.uk/Cancerinformation/Livingwithandaftercancer/Eatingwell/Nutritionalsupport/Nutritionalsupport.aspx	7
Figure 2: G-tube placement. Derived from: http://pedsurg.ucsf.edu/conditions--procedures/gastrostomy-tubes.aspx	8
Figure 3: Different Types of feeding tubes. Derived from https://alfa.saddleback.edu/data/enteral-feedings	9
Figure 4: Play Picnic. Source: Archives of the University Children's Hospital Graz with friendly permission of the children's parents.	17
Figure 5: Example for an „online medical chart“, derived from https://login.notube.com/user/login/	19
Figure 6: Extract of an intake protocol, derived from https://login.notube.com/user/login/	20
Figure 7: Development of number of patients in the treatment groups	28
Figure 8: Gender distribution in the Netcoaching program	29
Figure 9: Gender distribution in the onsite program	29
Figure 10: Countries of referral.....	31
Figure 11: Distribution of types of tubes in the two treatment options	32
Figure 12: Reaction to food in both treatment groups	33
Figure 13: Groups of diagnoses for both treatment options.....	41
Figure 14: Outcomes in both treatment options.....	43
Figure 15: Follow up outcomes	53
Figure 16: Comparison „Outcome after treatment – follow up“	56

List of tables

Table 1: Comparison of the two treatment options	23
Table 2: Statistical analysis of „Annual comparison of number of patients” in the two treatment groups.....	27
Table 3: Statistical analysis of „Gender distribution“ in both programs	28
Table 4: Statistical analysis of „age distribution“ in both treatment groups	30
Table 5: Distribution of types of tubes in the two treatment options	32
Table 6: Statistical analysis of „Reaction to food“ in both treatment groups	33
Table 7: Statistical analysis of „duration of tube feeding in months“ in both treatment options.....	34
Table 8: Statistical analysis of „percentage lifetime tube feeding“ in both treatment groups	34
Table 9: Statistical analysis of „groups of diagnoses“ in both treatment groups ...	42
Table 10: Statistical analysis of „outcome“ in both treatment groups	44
Table 11: Statistical analysis of underlying medical diagnosis and „outcome“	46
Table 12: Statistical analysis of „weight development“ in both treatment options.	47
Table 13: Statistical analysis of „Weight loss in %“ in both treatment groups.....	47
Table 14: Statistical analysis of „duration of treatment“ in both treatment groups	48
Table 15: Statistical analysis of the „Client Satisfaction Questionnaire“	50
Table 16: Statistical analysis of response rate to follow-up Questionnaire.....	51
Table 17: Statistical analysis of „Response rate over the years of treatment“	52
Table 18: Follow-up outcomes	53
Table 19: Follow-up outcome in the two treatment options	54
Table 20: Statistical analysis of comparison of „outcome after treatment – follow up“	56

Abstract german

Ziel der Studie: Das "Grazer Modell" zur Sondenentwöhnung erfreut sich seit fast 30 Jahren großer Bekannt- und Beliebtheit. PatientInnen werden aus dem In- und Ausland zugewiesen, um den Übergang von exklusiver Sondenernährung zu einer selbstständigen und nachhaltig ausreichenden oralen Ernährung zu erlernen. Neben der stationären und ambulanten Variante (onsite Programm) wurde im Jahr 2009 ein telemedizinisches Coachingprogramm (Netcoaching) entwickelt.

Diese Arbeit bietet erste Daten einer weltweit einzigartigen Stichprobe (N=473) von PatientInnen, welche zwischen dem 1.1.2009 und dem 31.12.2013 entweder an der ambulanten/stationären Therapie (N=228) *oder* am Netcoachingprogramm (N=245) teilgenommen haben. Evaluiert wurde neben biografischen (Alter, Herkunftsland, Geschlecht) sowie biometrischen und medizinischen (Diagnosen, Gewichtsentwicklung, Sondenart, Dauer der Sondenernährung, Dauer der Behandlung) und psychologischen (Reaktion auf Nahrungsmittel, Zufriedenheit der Eltern) Variablen vor allem die Erfolgsrate der beiden Programme. Ebenso wurde auch die Nachhaltigkeit der erfolgten gezielten Intervention analysiert.

Methode: Als Zielvariable wurde der erreichte Erfolg der Sondenentwöhnung auf einer vierstufigen Skala (vollständige Entwöhnung, Teilentwöhnung, Entwöhnungsversuch ohne Erfolg, Abbruch der Behandlung) gemessen. Die beiden Behandlungsoptionen wurden hierfür verglichen, statistische Kennwerte wurden berechnet.

Ergebnisse: Es zeigt sich, dass 220/245 (89.8%) PatientInnen im telemedizinischen online Programm (Netcoaching) sowie 184/228 (80.7%) PatientInnen im onsite Programm vollständig sondenentwöhnt werden konnten. Im Weiteren wurde im Netcoachingprogramm kein Kind mit "Entwöhnungsversuch ohne Erfolg" klassifiziert, im onsite Programm betraf dies 6/228 Kinder (2.6%). Beide Ergebnisse zeigten keine signifikanten Unterschiede zwischen dem angewandten Beratungskonzept und den Behandlungsformen.

Signifikante Unterschiede ergaben sich bezüglich der Ergebnisse “Teilentwöhnung” – 12/245 (4.9%) PatientInnen im Netcoaching sowie 37/228 (16.2%) PatientInnen im onsite Programm konnten partiell entwöhnt werden – sowie “Abbruch der Behandlung”: 13/245 PatientInnen (5.3%) brachen das Netcoaching-Programm vorzeitig ab, nur 1/228 (0.4%) PatientInnen verließ das onsite Programm vorzeitig.

Fazit: In dieser ersten Evaluation einer großen Population von Kindern die zur Sondenentwöhnung nach dem “Grazer Modell” behandelt wurden, zeigt sich, dass die Erfolgsquote der telemedizinischen Behandlung gegenüber der onsite Behandlung ähnlich gute bis tendenziell sogar bessere Ergebnisse vorweist. Vor dem Hintergrund der Ökonomie (keine Infektionsgefährdung, keine Spitalsbelegung, geringere Kosten, kein Reiseaufwand, keine Wartezeiten) ist das Netcoaching-Programm durchaus als zeitgemässe, kindgerechte, adäquate und attraktive Alternative zu der traditionellen onsite Behandlung zu sehen.

Abstract English

Aim of the study: The „Graz model“ of tube weaning has been internationally recognised since nearly 30 years. Patients from all over the world participate in the program. Besides the in- and outpatient treatment option (onsite program), a new telemedical program (online program called Netcoaching) has been developed in 2009. This thesis offers data of a unique and large sample of patients (N=473) who participated either in the onsite program (N=228) or in the telemedical online program „Netcoaching“ (N=245) between Jan 1st, 2009 and December 31st, 2013. Biographic data (age, country of referral, gender), biometrical and medical data (diagnoses, weight development, type of tube, duration of tube feeding) as well as psychological (reaction to food, client satisfaction) aspects were collected, analysed and evaluated. Furthermore, the sustainability of the tube weaning intervention corresponding to the long-term success rate has been analysed.

Method: The outcome of the tube weaning program was declared as the target variable. A four-step scale (totally weaned, partially weaned, weaning trial without success, interruption of treatment) has been used. Both treatment options were compared, statistical values were calculated.

Results: Complete weaning was achieved in 220/245 (89.8%) patients opting for the online program as well as in 184/228 patients (80.7%) in the onsite program. In the online program, no child was labelled with the outcome “weaning trial without success” whereas 6/228 children (2.6%) in the onsite program have been labelled with this outcome. Both results show no significant differences between the treatment groups.

Significant results are shown in the outcome groups “partially weaned” – 12(245 (4.9%) in the Netcoaching group versus 37/228 (16.2%) patients in the onsite group could be weaned partially – and “weaning trial without success”:

13/245 patients (5.3%) interrupted the Netcoaching program, and only 1/228 (0.4%) patient left the onsite program before completion.

Conclusion: In this first evaluation of a large sample of children participating in the “Graz model”-concept for tube weaning, similar efficacy of the newly invented telemedical online option compared with onsite program has been demonstrated. Based on economical aspects (less hospital admission, lower risk of infections, less expensive, no travel costs, no waiting list), the Netcoaching program can be considered as an adequate and attractive alternative for the goal of effective and sustainable transition from enteral to sufficient oral intake to the traditional onsite treatment options.

Table of contents

DECLARATION	I
ACKNOWLEDGEMENTS	II
ABBREVIATIONS AND DEFINITIONS	III
LIST OF FIGURES.....	IV
LIST OF TABLES	V
ABSTRACT GERMAN.....	VI
ABSTRACT ENGLISH.....	VIII
TABLE OF CONTENTS.....	X
1. INTRODUCTION/THEORETICAL BACKGROUND	1
1.1 TUBE FEEDING IN PAEDIATRICS.....	1
1.1.1 INDICATIONS.....	1
1.1.2 CONTRAINDICATIONS	6
1.1.3 DURATION OF TUBE FEEDING	6
1.1.4 TYPES OF FEEDING TUBES.....	7
1.1.4.1 Transnasal feeding tubes.....	7
1.1.4.2 Percutaneous feeding tubes.....	8
1.1.5 COMPLICATIONS OF TUBE FEEDING	10
1.1.5.1 Complications associated with the feeding tube	10
1.1.5.2 Gastrointestinal complications.....	10
1.1.5.3 Tube dependence	11
1.1.6 HOME ENTERAL NUTRITION (HEN)/HOME ENTERAL TUBE FEEDING (HETF)	12
1.1.7 PSYCHOLOGICAL ASPECTS OF TUBE FEEDING	13
1.1.8 TUBE WEANING.....	14
1.1.8.1 Tube weaning programs/strategies	14
1.1.8.2 The Graz model of tube weaning	16
1.1.8.3 Comparison of the two treatment options	22
1.2 TELEMEDICINE.....	24
1.2.1 TELEMEDICINE IN PEDIATRICS.....	24

1.2.2	USE OF TELEMEDICINE FOR THE TREATMENT OF EATING DISORDERS	24
2.	MATERIAL AND METHODS.....	26
2.1	PARTICIPANTS.....	26
2.1.1	INCLUSION CRITERIA	26
2.1.2	EXCLUSION CRITERIA	26
2.1.3	ANNUAL COMPARISON.....	27
2.1.4	GENDER.....	28
2.1.5	AGE.....	29
2.1.6	COUNTRIES OF REFERRAL	30
2.1.8	TYPES OF TUBES	32
2.1.9	REACTION TO FOOD	33
2.1.10	DURATION OF TUBE FEEDING	34
2.1.11	UNDERLYING MEDICAL DIAGNOSES.....	35
3.	RESULTS	43
3.1	PRIMARY OBJECTIVE: SUCCESS OF TREATMENT.....	43
3.2	FURTHER VARIABLES.....	45
3.2.1	DOES THE UNDERLYING MEDICAL DIAGNOSIS INFLUENCE THE OUTCOME?.....	45
3.2.2	WEIGHT DEVELOPMENT DURING TREATMENT	47
3.2.3	DURATION OF TREATMENT.....	48
3.2.4	CLIENT SATISFACTION.....	49
3.2.4.1	The Client Satisfaction Questionnaire (CSQ-8)	49
3.2.4.2	Outcomes of the CSQ-8.....	49
3.2.5	LONG-TERM OUTCOMES	50
4.	DISCUSSION	57
5.	BIBLIOGRAPHY	62
6.	APPENDIX	69
6.1	PUBLICATIONS.....	69
6.2	CONFERENCE PROCEEDINGS.....	71
6.3	MEDICAL QUESTIONNAIRE – ENGLISH VERSION.....	72

1. Introduction/Theoretical background

1.1 Tube feeding in paediatrics

Tube feeding is used in many different areas of paediatrics to influence the nutritional state of a child if the child is not able or allowed to cover its nutritional requirements itself. Enteral nutrition support (ENS) is generally defined as „nutrition via the GI tract“ (1) but the term is mostly used as a synonym for tube feeding.

1.1.1 Indications

Generally, feeding tubes are used if people are not able to sustain themselves by oral passage and is limited to patients who have at least a partially functioning gut (2, 3). Many medical or even psychological circumstances can lead to the situation that a person is malnourished or not able to cover its nutritional needs.

Pearce (4) defines indications for enteral feeding as followed:

- Medical (e.g. inflammatory bowel disease, renal failure...)
- Neurological (e.g. cerebrovascular accident, brain tumour...)
- Surgical (pre-/postoperative, Sepsis, gastrointestinal tract surgery...)
- Orthopaedic (e.g. after trauma)
- Psychiatric (eg. Anorexia nervosa)
- Paediatric (e.g. Cystic fibrosis)
- Miscellaneous (Intensive care patients, Ileus...).

In the field of paediatrics, commonly accepted criteria for nutritional intervention are not evidence based, so that the objectives and indication for the EN depend on the clinical condition of the child (5, 6). Some children need enteral nutrition since they were born (e.g. because of extreme prematurity, perinatal asphyxia, congenital malformations), others might need tube feeding after months or even years for a large range and diversity of pediatric reasons (e.g. severe injuries, cancer, infantile anorexia...).

The ESPHGAN (5) lists conditions for paediatric patients requiring enteral nutrition:

- Inadequate oral intake (disorders of sucking and swallowing, tumors, food aversion, anorexia, neurological impairment, prematurity...)
- Disorders of digestion and absorption (Cystic fibrosis, short bowel syndrome, malabsorption due to food allergies, immunodeficiency, chronic liver disease, intestinal fistula...)
- Disorders of gastrointestinal motility (Hirschsprung disease, chronic pseudo-obstruction..)
- Increased nutritional requirements and losses (Chronic solid-organ diseases, inflammatory bowel diseases, multiple trauma...)
- Growth failure or chronic malnutrition (Anorexia nervosa, nonorganic growth faltering)
- Crohn disease
- Metabolic diseases.

The next part of this thesis presents some typical and frequent examples of medical circumstances leading to the need for partial or complete tube feeding in infants and children:

- **Preterm birth**

The CSPEN guidelines (1) recommend tube feeding in preterm infants with gestational age <32-34 weeks, children who cannot be fed orally due to swallow or sucking dysfunction, illness or medical condition and as a supplement of inadequate oral nutrition intake. The feeding plan in preterm infants should follow a strict plan (1, 2, 3). Enteral nutrition should be introduced as early as possible if their general medical state allows gut feeding. However, the goal for preterm infants is the transition to oral feeding as early as possible, which can often be achieved with careful feeding training in medically stable preterms. Breast feeding is recommended (1).

- **Cerebral palsy**

Children who suffer from CP often have problems regarding swallowing, sucking and chewing. The issues range from sensory to neurological ones so that in some cases, complete or partial tube feeding is necessary. Nevertheless, no randomised control trials about the comparison of tube vs oral feeding in this diagnostic entity of children have been published (4).

- **Oncology**

Tube feeding is indicated for all patients who suffer from malnutrition or patients who are expected not to be able to eat for at least 7 days or not to eat sufficiently (<60% of daily requirement) for more than 10 days (7). During the phase of intensive chemotherapy, tube feeding is often necessary in paediatric patients. It could be shown that patients whose parents agreed to enteral nutrition before cycle 2 of chemotherapy showed less weight loss and improved nutritional status (5).

- **Congenital heart diseases**

Children who suffer from congenital heart diseases requiring open-heart surgery, will often need nutritional support through a feeding tube. Nearly half of the infants with univentricular heart defects require supplementation with nasogastric or gastrostomy tube at discharge from neonatal surgery (8). As a unintended side effect, some children stay dependent on their feeding tubes after medical stabilization (9) has been achieved and no medical reason demands further ENS.

- **Transplant medicine**

Infants with severe and progressive hepatic, renal or cardiac dysfunction needing a transplant, enteral nutrition is started before, during or after the intervention of organ or bone marrow transplant and immunosuppression. This option has clearly increased the longterm outcome and has become a state of the art intervention in the care of these fragile patients.

- **Metabolic disorders**

Some metabolic disorders in infancy require complete or partial tube feeding, examples are given below.

- *Persistent hyperinsulinemic hypoglycemia of infancy (PHHI)*

Children with PHHI require feeding tubes due to the fact that they need short and regular feeding intervals and a special dietary composition. Even in absence of other medical reasons not to eat orally, it is known that many children with PHHI suffer from oral aversion and food refusal (10).

- *Progressive metabolic disorders (e.g. Tay-Sachs disease, Krabbe disease...)/neurodegenerative diseases (e.g ALS, SMA...)*

Children with progressive neurodegenerative or metabolic diseases will often need enteral nutrition due to severe muscular hypotonia, progressing swallowing difficulties and severe feeding problems. However, the issue of enteral nutrition and especially the effect of its unintended negative side effects can sometimes also be perceived as a burden for the child itself and the families quality of life (11).

Goldman (1998) states: „In general, nutritional goals aimed at restoring health are secondary to comfort and enjoyment, although assisted feeding, via a nasogastric tube or gastrostomy, may be appropriate for those with slowly progressive disease“ (12).

- **Malformations/diseases of the gastrointestinal tract**

- *Gastroesophageal reflux disease (GER-D)*

Children with severe GER-D often show feeding problems and food refusal. This can lead to the necessity of enteral nutrition (13). In most cases, after ineffective attempts of propulsive and antacid medication and other unspecific changes of food consistency and positioning, a fundoplication might be recommended prior to tube placement. However, as nasogastric tube feeding might increase the symptom and does not prevent reflux, a PEG insertion is mostly recommended (14). Generally, enteral nutrition for children with GER-D should only be transitional and should only be performed with strict clinical indication.

- *Oesophageal atresia*

Children who are born with this malformation are candidates for early surgery and must be fed by enteral nutrition shortly after birth. After surgery and the complete healing of the oesophagus, children may start to eat orally, sometimes with limitations on texture (only pureed food due to recurrent oesophageal stenoses), nevertheless, many children stay dependent on the feeding tube or suffer from severe feeding/eating problems (15).

• **Mental and behavioural disorders**

As eating disorders of adolescents and adults are detected easily and the need of intermediate tube feeding is clearly accepted, only little evidence is given for mental and behavioural disorders of infants and toddlers requiring enteral nutrition.

- *Infantile Anorexia*

Infantile Anorexia is defined as an eating disorder characterized by food refusal and failure to thrive, the onset takes place during the developmental stage of separation and individuation (6– 36 months). The infant refuses to eat in order to increase its autonomy, it comes to a „battle of power and wills“ between the child and the primary caregiver, mostly the mother (16). In severe cases, short-term tube feeding is required. An effective exit-strategy for the feeding tube is crucial.

- *Autism spectrum disorders*

Children with Autism often suffer from feeding disorders, ranging from picky eating up to long-lasting food refusal (17). In specific cases, tube feeding is required to sustain the child's nutritional needs.

All in all, there are numerous indications for tube feeding in infancy and childhood. As in some cases the indication is clear (e.g. intensive care patients, children with malformations preventing oral feeding), there are cases when it is up to the physician on to decide whether a feeding tube should be placed (18).

1.1.2 Contraindications

The ESPGHAN Guidelines (5) list absolute contraindications for tube feeding: paralytic or mechanical ileus, intestinal obstruction, perforation and necrotising enterocolitis as well as relative contraindications: intestinal dysmotility, toxic megacolon, peritonitis, gastrointestinal bleeding, high-output enteric fistula, severe vomiting, intractable diarrhea.

1.1.3 Duration of tube feeding

- **Permanent vs. temporary tube feeding**

Whereas some conditions require permanent/life-long tube feeding (inability to swallow, progressive diseases), other patients need nutritional support by a feeding tube for a few days, weeks, months or even years. In many cases, the timeframe is not clearly defined which makes a transition (back) to oral feeding complicated.

Pearce (4) mentions, that the „majority of patients requiring nutritional support will need it for less than one month“.

McMahon et al. (19) provide a decision algorithm for long-term tube feeding, including patient's/caregiver's wishes, medical evaluation and provides help on which type of feeding tube should be chosen.

After placing a feeding tube, a specialised aftercare treatment by medical professionals is inevitable (20) including regular check-ups, growth monitoring and frequent evaluation about the benefits and disadvantages of the enteral nutrition.

Generally, there is lack of evidence-based guidelines about the duration of enteral nutrition and required exit strategies.

1.1.4 Types of feeding tubes

There are many types of feeding tubes, they differ regarding the site of food delivery (gastric vs postpyloric) and the route (transnasal vs percutaneous tubes). The choice of the route is influenced by the likely duration of tube feeding as well as the integrity of the upper gastrointestinal tract (5).

1.1.4.1 Transnasal feeding tubes

Transnasal feeding tubes are placed via the nasopharynx into the stomach, duodenum or jejunum.

- **Nasogastric tubes**

NG tubes are polyurethane or silicone tubes of various sizes passed through the nasal passages via the oesophagus into the stomach. A correct insertion on the tube is vital. After placement, the position has to be verified. Generally, the tube is placed by medical staff, in some cases, parents are trained to insert the tube themselves. Nasogastric tubes should be used when the medical staff suspects the patient to need enteral nutrition support for less than 4-6 weeks (5, 21). Nasogastric tubes are not considered to be ideal for long-term enteral nutrition (4).

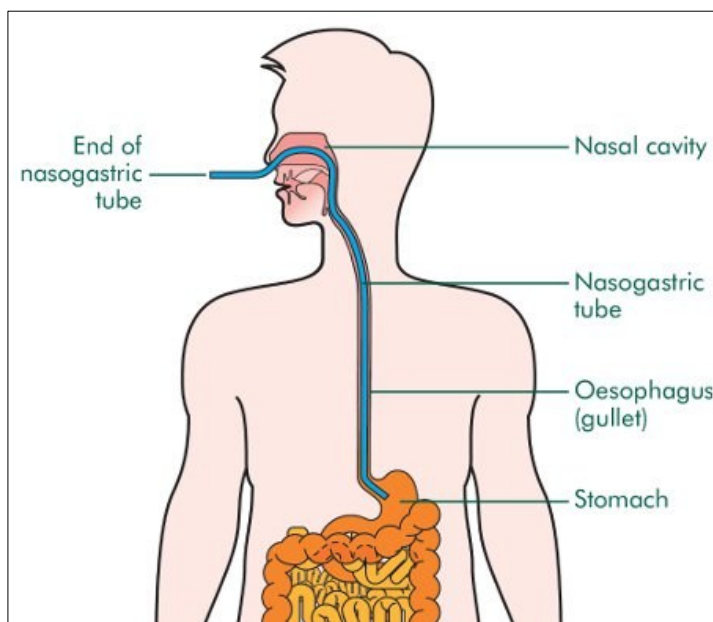


Figure 1: Placement of a nasogastric tube. Derived from: <http://www.macmillan.org.uk/Cancerinformation/Livingwithandaftercancer/Eatingwell/Nutritionalsupport/Nutritionalsupport.aspx>

- **Nasoduodenal/nasojejunal tubes**

ND or NJ tubes are placed transnasal into the duodenum or jejunum. The placement can be difficult, it is administered by endoscopy, fluoroscopy or “self propelling” feeding tubes (4). Duodenal tube feeding improves weight gain of infants with GER who need treatment for CHD-associated heart failure (22).

1.1.4.2 Percutaneous feeding tubes

- **Gastrostomy**

The percutaneous endoscopic gastrostomy (PEG) is inserted into the stomach of the patient using endoscopy. In few cases, a gastrostomy has to be inserted surgically (4). In case of long-term tube feeding (>4 weeks), feeding via a gastrostomy is the preferred route (4, 5, 21, 23) but contraindications have to be kept in mind. Gastrostomy feeding devices are either tubes or buttons, mostly a PEG tube is inserted in the first place and then replaced by a button. G-tubes and buttons are held by a balloon or bumper.

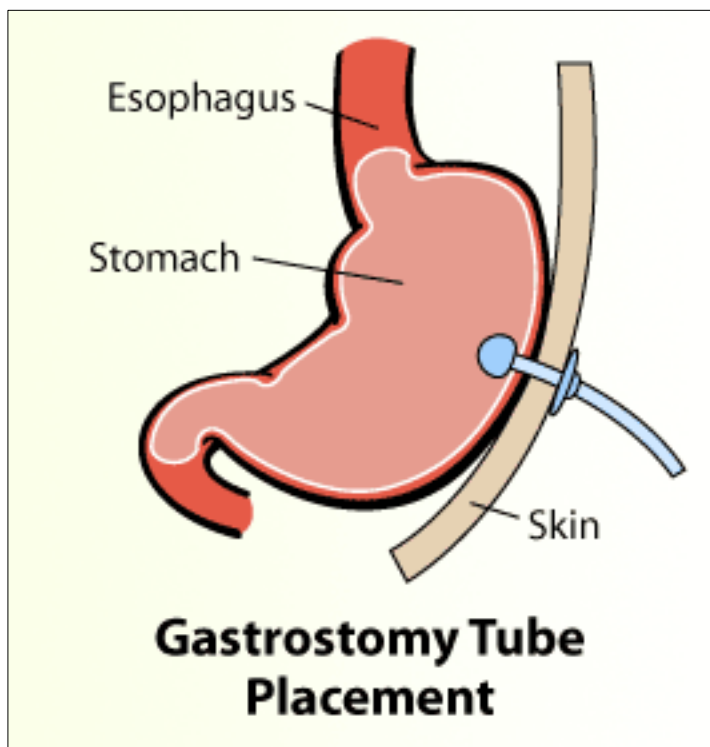


Figure 2: G-tube placement. Derived from: <http://pedsurg.ucsf.edu/conditions--procedures/gastrostomy-tubes.aspx>

- **Jejunal feeding tubes**

Percutaneous jejunal feeding tubes are often used in patients with unsuitable stomach access or with severe side effects of nasogastric/PEG feeding like massive vomiting.

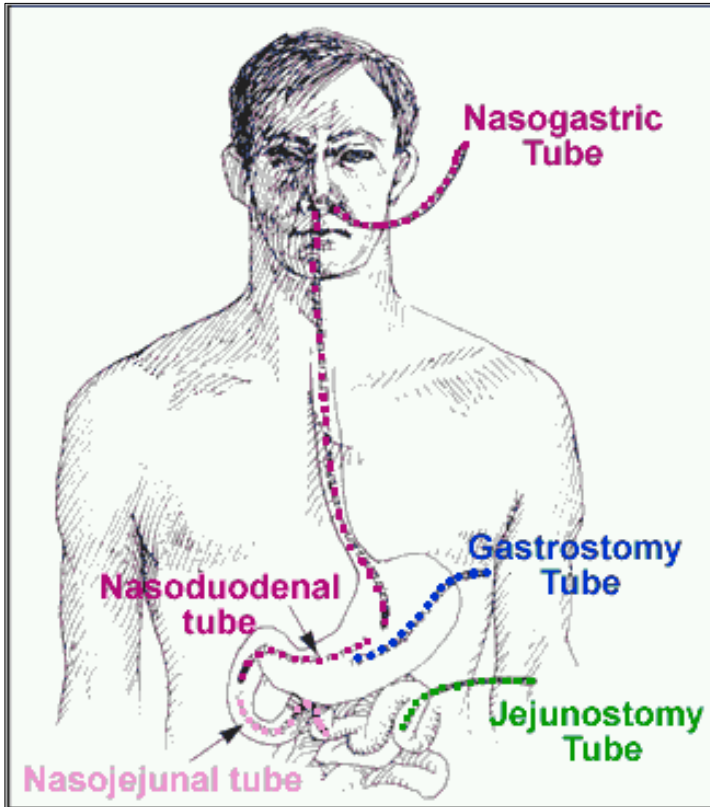


Figure 3: Different Types of feeding tubes. Derived from <https://alfa.saddleback.edu/data/enteral-feedings>

1.1.5 Complications of tube feeding

Beside the positive and often life-sustaining advantages of enteral nutrition, there are many reports on complications associated with tube feeding.

1.1.5.1 Complications associated with the feeding tube

Complications associated with the feeding tube itself may occur during tube placement but also during the phase of tube maintenance.

Complications specifically related to transnasal feeding tubes are: clogging, risk of misplacement, oesophageal or pulmonary perforation, inadvertent insertion in the trachea or bronchi, nasopharyngeal discomfort (4, 5). Complications correlated to percutaneous feeding tubes include: bleeding, perforation, peristomal wound infection, peritonitis, inadvertent removal, tube fracture and leakage, balloon ruptures, skin irritation as well as granulation tissue (4, 5, 24, 25).

1.1.5.2 Gastrointestinal complications

Reported gastrointestinal complications include: nausea, vomiting, regurgitation, aspiration, osmolality, altered gastric emptying, abdominal bloating, constipation, diarrhea, GERD (1, 4, 5, 26, 27).

Especially long-term tube feeding is associated with a high complication rate. Late complications are reported in 44% of all children receiving long-term enteral nutrition (28). Furthermore, tube feeding does not lead to improved nutritional state or improved weight situation in all cases (27, 29), even not in children (30). A recent study could show that even >40% of a sample of tube fed children was underweight (31). Furthermore, an impact on appetite can not be denied (32).

These various negative side-effects often lead to the development of an eating disorder (33, 34).

1.1.5.3 Tube dependence

Tube dependence, is described as followed:

„A tube dependent child remains tube fed although his/her medical condition and developmental potential would allow the transition to oral nutrition. Children with TD show characteristic symptoms such as food refusal and opposition to any oral feeding attempts. They often suffer from additional episodes of vomiting, nausea, gagging and retching and in some cases develop severe failure to thrive“ (34).

So children suffering from TD remain being tube fed although there are no medical reasons left to continue the ENS. TD can be seen as a negative, unintended side effect of enteral nutrition (35).

There are different stages of TD:

- Stage 1:
the child is 100% fed by tube and does not accept any caloric food orally.
- Stage 2:
the child is 80-100% fed by tube but shows interest in food, but eats only <20% of its required caloric intake orally.
- Stage 3:
The child has 1 or 2 meals orally a day. About 60% of the daily intake are fed by tube.
- Stage 4:
The child eats about 50% orally and receives the other 50% by tube, often only during nighttime.
- Stage 5:
The child is mainly on oral feeds, but receives 10-30% supplemental feedings by tube (34).

1.1.6 Home enteral nutrition (HEN)/Home enteral tube feeding (HETF)

Whereas short-term enteral nutrition is mostly administered during inpatient stays in hospitals, some children are discharged to home care with a feeding tube. Parents have to be trained to administer tube feedings and care about the tube feeding equipment.

Although there are only few scientific reports on the use of HEN, patients receiving HEN might have increased during the last years (36-38).

A close monitoring of the enteral nutrition by local professionals is essential. Monitoring should include regular review of nutritional status by a nutritional support team such as intake, weight, height, biochemical and haematological indices, general clinical state, well-being, gastrointestinal function, tube integrity, tube-related complications (5).

A recent systematic review and meta-analysis analysed the effectiveness of care delivery models for HEN patients, most of them using multiple simultaneous strategies, such as family and staff education, continuous auditing and feedback methods. Nevertheless, no significant reductions in complications, hospital admissions or infections could be found, although the hospital costs decreased significantly (39). Another investigation could also show significant decrease of hospital treatment costs by involving a specialized HETF program, in this study, it could even be proved that number of admissions and duration of hospital stays as well as tube feeding related complications could be reduced significantly (40).

Furthermore, a study proved that HETF techniques of caregivers of children receiving long-term HEN (>5 years) due to metabolic disorders decline over time: accurate feed ingredient measurement decreased as well as correct flushing of tubes, checking tube position, correct hand washing (41).

Sometimes problems occur when discharging the patients from inpatient treatment by not providing ideal information for local teams – GPs in Northern Ireland claimed that they had only little involvement in patient selection for the procedure as well as poor or no discharge information, they felt inadequately trained and there has been poor communication between primary and secondary care.

Even anger and frustration occurred due to lack of resources and the perception to be “used as a dumping ground” (42). In Brazil, an electronic protocol for home enteral nutrition therapy has been successfully developed to collect data on treatment of patients receiving HEN (43).

In the near future, such tools might be of great help to ensure best possible monitoring of patients and to be up-to-date regarding their nutritional state, side-effects as well as exit strategies for the tube.

Highly specialized teams, effective discharge plans, adequate long-term plans for tube maintenance and exit strategies, clear responsibilities for monitoring HEN and a valuable correspondence between the tube-placing institution and the maintenance team seem to be crucial (35, 37, 38).

1.1.7 Psychological aspects of tube feeding

Some studies focus specifically on the psychological aspects of tube feeding, especially on being a relative or caregiver to a patient being in need of long-term enteral nutrition support.

One study investigated stress levels of parents whose children were enterally fed using the Parenting Stress Index (PSI). It could be shown that >40% of the parents showed high stress levels, stress scores were significantly higher than in the norm group (44).

Patients who receive HETF themselves, often show a reduced quality of life. Regarding the satisfaction with gastrostomy feeding of parents of children receiving HEN, it could be shown that lengths of HEN and age at tube placement are the main factors associated with parental satisfaction (45).

In children with severe neurological impairment, the insertion of a tube did not increase quality of life of their parents although some positive aspects could be shown on feeding and administration of medications (46).

A metaanalysis on the impact of tube feeding on maternal emotional state figured out that „feeding a child orally is not only an important aspect of mothering but also a key element for the development of a motherhood identity.

Nonoral feeding often results in psychological stress and a struggle to negotiate the motherhood identity successfully“ (47).

Furthermore, close relatives of persons receiving HETF mentioned, that their life changed a lot and is restricted by the HETF (48). It could be shown that it is very important to consider also the parental perception regarding decision of a PEG tube placement and the following provision of care (49).

A review study could show that „mixed messages and pressure from health professionals and relatives made decision making about tube feeding more difficult for parents“ (50).

Adequate information, (psychological) support from health care systems for close relatives, especially parents of tube dependent children, is indispensable (47, 48).

1.1.8 Tube weaning

If a feeding tube is not necessary any more from a medical and nutritional point of view, tube weaning is possible and should be the ultimate goal (6). Unfortunately, many children suffer from tube dependence after long-term tube feeding. In these cases, specific tube weaning programs are necessary to ensure the transition from tube feeding to oral nutrition.

1.1.8.1 Tube weaning programs/strategies

Various tube-weaning programs have been developed and published. They differ regarding treatment options, duration, therapeutic techniques.

Hunger provocation is one of the frequently used techniques.

Ishizaki et al. used either reduction/discontinuation of tube feeding, encouragement for self-feeding or the training of perioral muscles. 28 of 35 children (80%) could be weaned successfully through these treatment options (31).

Kindermann et al. treated 10 children with a multidisciplinary hunger provocation program. Children were treated in a hospital for 9-33 days, weight loss was between 3.7% - 15.6%. Three and six months after the weaning phase, 80% of the

children were eating adequate amounts and gained weight. Two children received partial tube feeding (51).

Behavioural treatment is also used in some cases. After completing an intensive inpatient behavioural tube weaning program, 51% of 77 children could be weaned off their feeding tubes completely, another 12% completed the weaning on an outpatient basis during the following year (52). Benoit et al. compared behaviour therapy with nutritional therapy regarding outcomes of tube weaning. Children who participated in the nutritional intervention group received structured schedules to stimulate hunger/satiety. The patients in the behavioural intervention group received the same schedules as well as behavioural therapy. 4.5 months after treatment start, 47% of 32 children in the behaviour therapy group could be weaned successfully, whereas no child receiving only nutritional intervention was successful in the transition from tube to exclusive oral feeding (53).

Another study dealing with outcomes of a multicomponent behavioural program for gastrostomy dependent children, could show, that 9 children who were admitted together with their mothers for a mean of 11.4 days were treated through the family-focused behavioural feeding program. At discharge, 4 out of 9 children (44%) were completely weaned off their tubes. About 3 months after the treatment, the success rate increased to 67%.

Mc Grath et al. used medication for the sake of tube weaning. They treated children in a 14-week outpatient program. After ensuring that all children had the motor skills necessary for eating as well as necessary mealtime behaviours. Then they started to give the children amitriptyline and/or gabapentin as well as continuous drip gastrojejunal tube feedings. After 8 weeks, an appetite stimulant was prescribed additionally and tube feedings were reduced. The appetite stimulant was discontinued after 6 weeks, the other medication was stopped after 6 months. Eight out of nine children who underwent the treatment were weaned successfully (took 100% of their intake orally, maintained their weight), one remained at 50% tube feeding (54).

Another great discussion point is the duration of tube weaning programs. Short-term weaning programs have been quite successful.

A rapid home-based feeding program helped children in establishing oral feeding in 89.7% of all cases. One to three years after treatment, no deceleration of growth could be found (55).

Wright states that a rapid tube weaning program is not ideal for all children (56).

Although some programs on tube weaning are established, many patients do not get adequate treatment, some even have to travel abroad (56). Many medical professionals are not yet trained in the techniques of tube weaning.

1.1.8.2 The Graz model of tube weaning

The „Graz model of tube weaning“ was established at the University Children’s Hospital Graz during the last 25 years.

The multidisciplinary program treats children suffering from tube dependency in an in- or outpatient setting. The team consists of paediatricians, nurses, different therapists (physiotherapists, speech- and language therapists, occupational therapists), clinical psychologists and dieticians. It is offered on an inpatient as well as on an outpatient basis. Generally, the program lasts for 3 weeks, shorter and longer treatment is possible in individual cases.

The program follows 2 basic principles:

1. Physical approach: let the child become hungry
2. Psychological/developmental approach: increase autonomy (57)

Principle 1 means that the nutrition by tube is reduced “as drastically as possible and as gradually as necessary” (35).

One of the main interventions of the Graz model is the so-called “Play Picnic”, a session based on elements of psychodynamic play therapy. The patients, their caregivers as well as the medical personnel take part in the 60 minutes lasting session. Different kinds of food are served on little toy plates, the children are allowed to do anything they like with the distributed nutrition (touching, licking, smearing, throwing, playing...).

The adults must eat themselves while letting the children explore the world of food. It is not allowed to interfere, wipe, clean the children or offer food actively (58).



Figure 4: Play Picnic. Source: Archives of the University Children's Hospital Graz with friendly permission of the children's parents.

Force feeding or the use of any medication are not part of the program (58).

The accepted weight loss during the process differs from child to child based on starting weight and growth situation, medical condition, age, but should not exceed 10% of the body weight.

Success rates show complete weaning in 80-92% (34, 58, 59) of all cases.

1.1.8.2.1 The Netcoaching program

The Netcoaching program has been developed in 2009, as the onsite program has not been accessible for some families for various reasons such as

- 1) Travel and treatment costs
- 2) Immune deficiency of the child (e.g. after transplantation or in case oncological diseases)
- 3) Inability to travel (60) .

An online platform has been specifically developed for guiding the process of tube weaning using the principles of the Graz model.

If a family is interested in joining the Netcoaching program, they need to register on the platform www.notube.com.

The treatment is offered in German, English and French. Technical preconditions are a regular access to a web-enabled PC, a stable internet connection as well as a device able to recording short video sequences. Furthermore, a daily access to digital scales is obligatory.

All children have to participate in a (tele-)medical assessment, including

- a) The completion of a specifically developed medical questionnaire, including biographical data, information about underlying medical diagnoses, growth, tube details, birth details, information about the child's and the parental behavior with food, information on the current nutrition and feeding regime (see Appendix).
- b) The forwarding of all medical documents of the child dealing with the topic of tube feeding or the underlying medical condition.
- c) Videos of feeding/eating/dealing with food scenes.
- d) Provision of the contact details of a local medical doctor who knows the child well and is informed about the requested participation in the Netcoaching program.

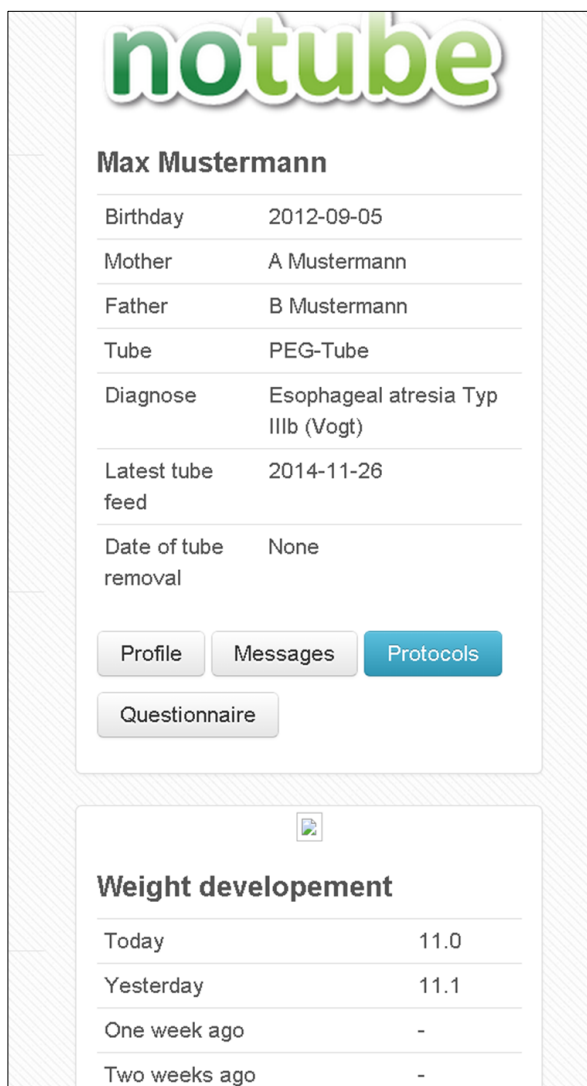
After all these documents have been submitted, two medical doctors receive them and decide independently if the child is suitable for the program.

If needed, a clinical psychologist and/or feeding therapist are involved in the decision.

The parents/caregivers receive the information about their child's suitability via e-mail or by phone. After that, the program can start right away.

The first step is to log in to the protected „Ticket“, specifically created for their child.

Every child who starts the treatment gets an own „online medical chart“ including Name, a photo of the child, contact details, medical details, tube details, weight charts, intake protocols.



The screenshot shows the 'notube' interface for a user named Max Mustermann. The top section displays the user's name and a table of personal and medical details. Below this are navigation buttons for Profile, Messages, Protocols, and Questionnaire. The bottom section is titled 'Weight development' and shows a table with weight data for Today, Yesterday, One week ago, and Two weeks ago.

Max Mustermann	
Birthday	2012-09-05
Mother	A Mustermann
Father	B Mustermann
Tube	PEG-Tube
Diagnose	Esophageal atresia Typ IIIb (Vogt)
Latest tube feed	2014-11-26
Date of tube removal	None

Profile Messages Protocols Questionnaire

Weight development	
Today	11.0
Yesterday	11.1
One week ago	-
Two weeks ago	-

Figure 5: Example for an „online medical chart“, derived from <https://login.notube.com/user/login/>

Feeding Data						
	Tube Feed [mls]	Tube Water [mls]	Oral Feed Offer	Oral Food Taken	Oral Liquied Offer	Oral Liquid Taken
-01:00						
-02:00						
-03:00						
-04:00	100ml Nutrini					
-05:00	100ml Nutrini					
-06:00	100ml Nutrini					
-07:00			50ml puree	10ml puree		
-08:00					50ml milk	5ml milk
-09:00						
-10:00						
-11:00						

Figure 6: Extract of an intake protocol, derived from <https://login.netube.com/user/login/>

Every tube weaning by Netcoaching is guided by at least one senior medical doctor (paediatrician) and an specifically trained and experienced clinical psychologist. If needed, a physiotherapist and/or a music therapist can be involved. The whole communication is performed in written form in either English, German or French. Every statement of one of the parties (professionals and parents/caregivers) is included in a „news feed“ with time and date mark and can be read again anytime during the process.

As soon as the treatment starts, the medical professionals introduce themselves and ask final questions about the current state and feeding regime. All parents receive a document on their rights and duties of being involved in the online process as well as a detailed description of the medical professional’s rights and duties.

Duties and rights of the parents/caregivers are: daily submission of a detailed intake protocol including height, weight, intake (tube and oral), as well as „date of last tube feed“ and „date of tube removal“ after the child got rid of all tube feeds. Furthermore, they must send a short message on each day of treatment on their qualitative observations: mood of the child, general state, change in behavior,

child's interaction with food (touching, playing, smelling...) as well as their personal issues, emotions and fears. The parents/caregivers are asked regularly to upload videos of specific scenes of their child's interaction with food. The parents/caregivers are able to access the platform 24/7, they are invited to share all their questions, fears and concerns regularly. In case of any medical emergencies (illnesses, injuries...) the parents are responsible to take the child to a medical doctor or a hospital onsite and follow the instructions of this institution. The online team will suggest immediate medical checkup and the weaning program is interrupted until the child has fully recovered. If a family doesn't update the ticket within one week, it will be temporarily closed. All families receive a document dealing with the philosophy, principles and accomplishment of the „Play picnic“ and are asked to perform it with their children as well as other family members, siblings or friends on a regular basis. Any attempts of force-feeding are strictly prohibited.

Duties of the medical online professionals: It is guaranteed that all tickets will be answered within 24 hours, mostly, it happens more often (2-3 times a day). As many patients live in different time zones, a synchronic communication is not always possible. Each child gets a detailed individual plan of tube feed reduction, based on the intake protocol the suggested feeding plan for the next day is adjusted daily. All questions, concerns and fears of the parents must be addressed and treated respectfully and answered immediately. If needed or wished, medical professionals who know the child well and want to accompany the process, are invited to join the communication in the ticket system. Parents/caregivers get constant information on the following themes: feeding devices, recommended aliments for their children during the weaning process, interaction during eating/feeding scenes.

The primary objective is to initiate and increase oral feeding/eating of the child up to a sufficient amount while simultaneously reducing the tube feeds and finally stop them completely. The accepted weight loss follows the same rules as in the in- and outpatient treatment option.

The program ends 35 days after the very last tube feed of the child, if the child eats sufficient amounts orally and is able to keep its weight stable. Every family

receives a coaching report including recommendations for the future (e.g. tube removal, expected weight gain...). This document is sent to the parents/caregivers as well as the medical professionals onsite.

A sign-up for an aftercare treatment (bookable on a monthly basis) is possible at all times.

1.1.8.3 Comparison of the two treatment options

The following table gives an overview on the similarities and differences of the Graz model treatment options:

	Online	Onsite
Basis	“Graz model” of tube weaning	“Graz model” of tube weaning
Cornerstones	Telemedical assessment of suitability, medical supervised reduction of tube feeds, serviced 24/7, weight and intake monitoring by intake protocols, video analysis, guided by at least one paediatrician and one clinical psychologist	Medical supervised reduction of tube feeds, medical rounds 2x/day, speech therapy 3x/week, occupational therapy 3x/week, nursing team, psychology for parents 2x/week, physiotherapy 2x/week, play picnic 5-7x/week
Duration	Time investment/patient: approximate average of 20 hours/child/wean with a large range between 6-150 hours! Continued until 35 days after the last tube feed and stable weight and good general condition is ensured	Time investment/patient: Rigid time schedule of 51-55 hours/child/wean with hardly any individual range. Standardized 3-weeks-program, shorter or longer duration possible in exceptional cases

Costs	3400.- Euro	Outpatient around 9000.- Euro, inpatient about 19000 Euro, depending on country of referral based on Austrian Health regulations
Surrounding	Home, internet-based, one medical doctor onsite who agrees to participation	University Children's Clinic, Graz, Austria (on an in- or outpatient basis)
Insurance coverage	Covered by individual decisions of insurance	Covered for Austrians, covered individually by S2 form for EU patients, no coverage for non-EU patient

Table 1: Comparison of the two treatment options

1.2 Telemedicine

1.2.1 Telemedicine in pediatrics

The use of telemedicine and telepsychology has increased over the last couple of years in many areas of pediatrics (62).

Spooner et al. (63) list the following fields of use for telemedicine in pediatrics:

- Radiology
- Mental health
- Dermatology
- Cardiology
- Emergency and transport services
- Hospital care and family education
- Pathology
- Child abuse
- Patient education and chronic disease
- School Health
- Home health
- Other services such as dentistry, ophthalmology.

1.2.2 Use of telemedicine for the treatment of eating disorders

Some telemedical/telepsychological programs have been introduced for the treatment of eating and feeding disorders.

Wagner et al. (64) evaluated the outcomes of a new technology assisted guided self-help program in patients adolescents and adults with Bulimia nervosa. Patients were randomly allocated to a cognitive behavioural therapy-based self-help program, either delivered by Internet as by bibliotherapy. It could be shown that bingeing as well as compensatory behaviour decreased almost equally in both groups over time.

Telemedical treatment of obesity (65) of children living in rural areas could show that the use of telemedicine could be one mode of intervention.

Silverman (66) deals with the topic of feeding disorders and telehealth interventions. It is stated, that there are quite few experts who are specialized in the treatment of feeding disorders and that telehealth interventions may help families to access therapies not available in their communities. Telehealth services were compared with onsite services and it could be shown that telehealth store and forward services may be useful especially for medical record review, feeding record analysis whereas telehealth real-time communication might be very useful for clinical interviews, behavioural observation of the child while being fed, behaviour management, parent training as well as consultation with specialists from other clinics.

Furthermore, telehealth is considered as an „effective low cost alternative to traditional therapy“. The following advantages of telehealth services are listed:

- Therapeutic efforts are delivered directly to a patient's home
- Decreased travel time
- Reductions in time being away from work.

Factors that support the use of telehealth services in treatments for feeding problems are: nutrition and behavioural interventions do not require physical examinations, feeding therapy needs several consultations – travelling can be reduced, feeding therapy can be conducted in the families home (Silverman) .

Silverman states that „telehealth technologies will play a significant role in the future“ regarding the treatment of feeding problems.

2. Material and methods

2.1 Participants

All patients who participated in one of the two treatment options (onsite vs. online) in five consecutive years (January 1st, 2009 – December 31st, 2013) were included in the study (N=473). All patients had participate in a medical assessment prior to starting the treatment, either onsite (outpatient visit at the University Children's Clinic Graz) or online. The medical assessment included the completion of a specifically developed medical questionnaire (see Appendix), the forwarding of all medical documents of the child dealing with the topic of tube feeding or the underlying medical condition, in case of telemedical assessment: videos of feeding/eating/dealing with food scenes analysed by at least two professionals, in case of onsite assessment: watching of a feeding/eating/dealing with food scene by at least two professionals. After submission of all documents it was decided whether the child is suitable to participate in the program.

Random allocation to the treatment groups was not possible due to ethical reasons (costs, travel distance, parental freedom of choice).

2.1.1 Inclusion criteria

Positive assessment, presence of a feeding tube (nasogastric, G-tube or button, Jejunal tube, JET-PEG) and tube dependency of stage 1 or 2 at treatment start have been defined as inclusion criteria for the study.

2.1.2 Exclusion criteria

General exclusion criteria for the participation were defined as followed: inability to swallow, severe dysphagia, aspiration issues, malformations of the gastrointestinal tract making oral nutrition impossible from a physiological point of view.

- *Exclusion criteria for the Netcoaching treatment:*
 - An illness (e.g. severe hyperinsulinism, inborn error of metabolism) demanding permanent medical onsite monitoring during food reduction
 - Severe social, psychological or psychiatric problems of the family resulting in non-compliance.

- *Exclusion criteria for the onsite treatment:*
 - Children with reduced immunological competence (e.g. after transplantation)
 - Fear of nosocomial infections
 - Refusal of inpatient or outpatient treatment by patients or parents (due to traumatic hospital experiences).

2.1.3 Annual comparison

Table 2 and Figure 5 give an overview about the number of patients in the different treatment options during the five consecutive years.

One can see that in 2009 and 2010, when Netcoaching started, more patients participated in the onsite treatment. In 2011 and 2012, the number of patients treated onsite and in the Netcoaching program was quite equal, whereas in 2013, significantly more patients participated in the Netcoaching than in the onsite program.

Year	Program			Total
		Netcoaching (n=245)	Onsite (n=228)	
2009	n	29 (11.8%)	49 (21.5%)	78 (16.5%)
	std. residual	-1,8	1.9	
2010	n	40 (16.3%)	63 (27.6%)	103 (21.8%)
	std. residual	-1.8	1.9	
2011	n	51 (20.8%)	53 (23.2%)	104 (22.0%)
	std. residual	-0.4	0.4	
2012	n	47 (19.2%)	35 (15.4%)	82 (17.3%)
	std. residual	0.7	-0.7	
2013	n	78 (31.8%)	28 (12.3%)	106 (22.4%)
	std. residual	3.1	-3.2	
Total	n	245 (100.0%)	228 (100.0%)	473 (100.0%)
p < 0.05, df=4, X ² = 35.08				

Table 2: Statistical analysis of „Annual comparison of number of patients” in the two treatment groups

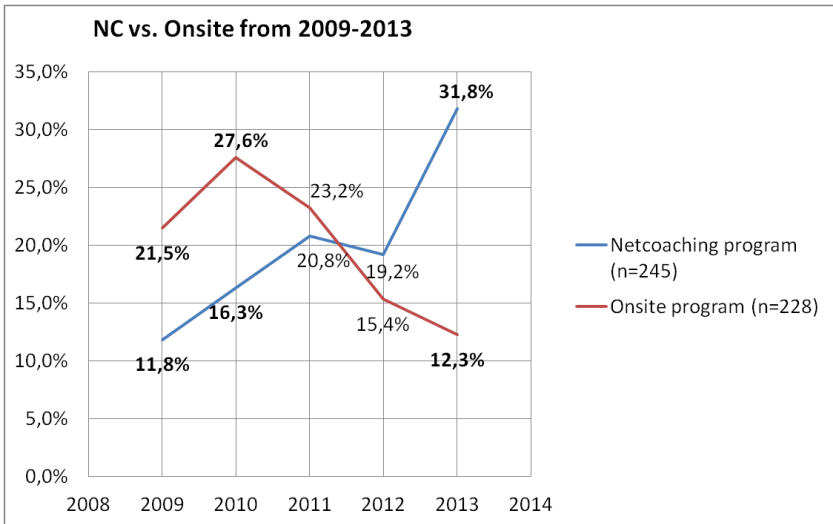


Figure 7: Development of number of patients in the treatment groups

2.1.4 Gender

Generally, a few more girls (N=258; 54.5%) than boys (N=215; 45.5%) participated in the tube weaning programs, but no significant difference could be found. About the same number of male and female patients participated in both programs.

Gender	Program		Total
	Netcoaching (n=245)	Onsite (n=228)	
Female	138 (56.3%)	120 (52.6%)	258 (54.5%)
Male	107 (43.7%)	108 (47.4%)	215 (45.5%)
Total	245 (100.0%)	228 (100.0%)	473 (100.0%)
<i>p</i> > 0.05, df=1, X ² = 0.65			

Table 3: Statistical analysis of „Gender distribution“ in both programs

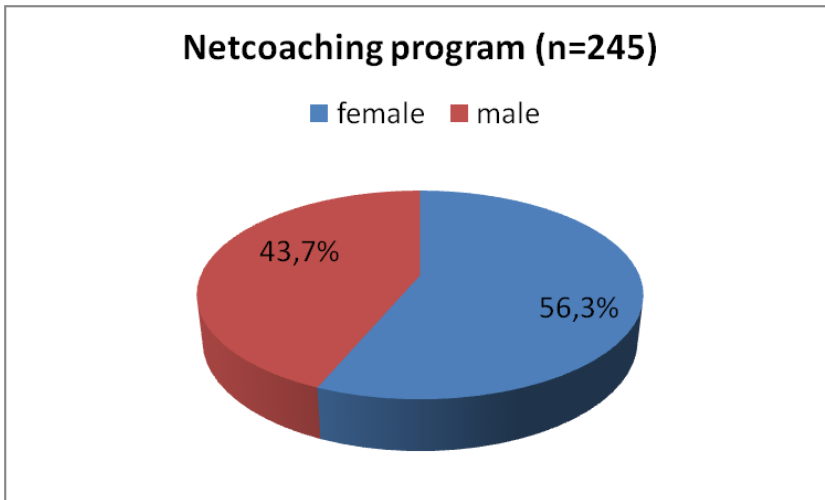


Figure 8: Gender distribution in the Netcoaching program

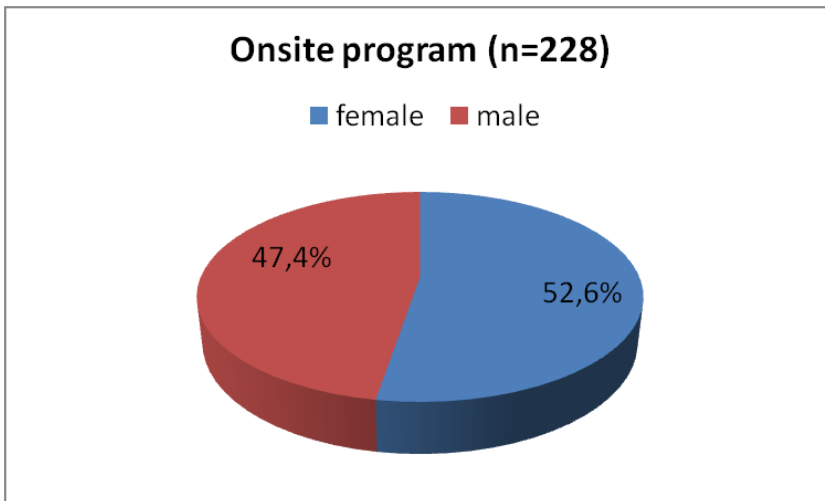


Figure 9: Gender distribution in the onsite program

2.1.5 Age

The patient's age ranged from 0.21 years to 19.65 years in the onsite group and from 0.27-23.65 years in the Netcoaching group.

The median age of the Netcoaching group was almost 1 year lower than in the onsite treatment (1.28 vs 2.19 years, $p < 0.05$).

Program	Age in years				
	Mean	Median	SD	Min.	Max.
Netcoaching (n=245)	1.78	1.28	2.09	0.27	23.65
Onsite (n=228)	2.79	2.19	2.42	0.21	19.65
$p < 0.05$ (p= 0.0), Mann-Whitney-U= 18455.5					

Table 4: Statistical analysis of „age distribution“ in both treatment groups

Children in the Netcoaching program were significantly younger than in the onsite program.

2.1.6 Countries of referral

Overall, patients from 34 different countries participated in the programs:

- United Arab Emirates
- Belgium
- Brazil
- Canada
- Switzerland
- Denmark
- Ireland
- Croatia
- Hungary
- Israel
- India
- Italy
- Japan
- Liechtenstein
- Mexiko
- Namibia
- Netherlands
- Norway
- New Zealand
- Poland
- Romania
- Russia

- Sweden
- Singapur
- Slovenia
- Turkey
- Ukraine
- South Africa
- Germany
- Austria
- United Kingdom
- France
- Australia
- United States of America.

Most patients who participated in the onsite program were referred from Austria (N=80), followed by Germany (N= 41) and United Kingdom (N=20) as well as Australia (N=18), whereas in the Netcoaching group, most patients lived in the United States of America (N=65), followed by Australia (N=42), Germany (N=37) and United Kingdom (N=20).

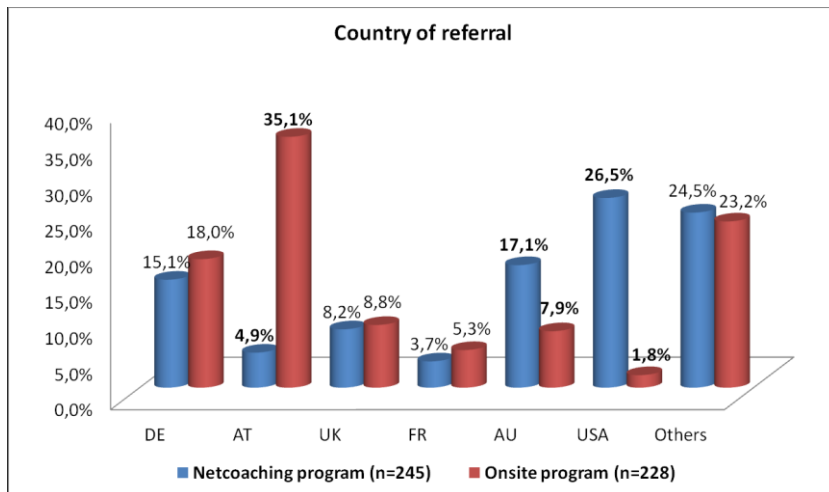


Figure 10: Countries of referral

2.1.8 Types of tubes

214 patients (45.2%) were fed by a nasogastric tube whereas 253 patients (53,5%) had a PEG tube. Only six children (2.5%) had a Jejunal tube.

Table 5 shows, that significantly more children in the Netcoaching program had a nasogastric tube, whereas more children in the onsite program were fed by a PEG tube.

Furthermore, it is shown that only children participating in the Netcoaching treatment had a Jejunal tube.

Kind of tube	Program		Total
	Netcoaching (n=245)	Onsite (n=228)	
NGT	126 (51.4%)	88 (38.6%)	214 (45.2%)
PEG	113 (46.1%)	140 (61.4%)	253 (53.5%)
J	6 (2.5%)	0 (0.0%)	6 (1.3%)
Total	245 (100.0%)	228 (100.0%)	473 (100.0%)

$p < 0.05$, Fisher's Exact Test=15.09

Table 5: Distribution of types of tubes in the two treatment options

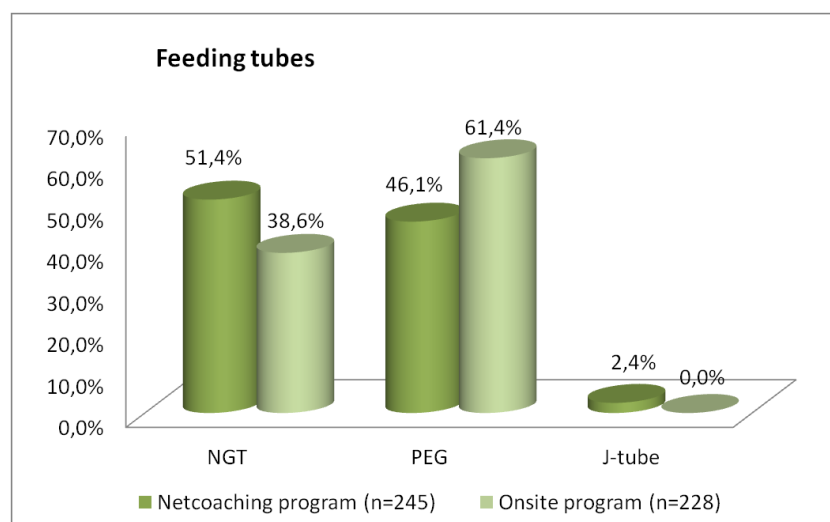


Figure 11: Distribution of types of tubes in the two treatment options

2.1.9 Reaction to food

During the medical assessment, all parents were asked about their child's reaction towards food. The parents were able to choose between three options:

- positive
- indifferent
- negative.

It could be shown that about a third of all patients in both programs considered their child's reaction to food as positive, a third as indifferent, a third thought that their child reacted negative to the exposure to food. No significant differences between the treatment options could be found regarding this variable (Table 6).

Reaction to food	Program		Total
	Netcoaching (n=245)	Onsite (n=228)	
Positive	73 (29.8%)	78 (34.2%)	151 (31.9%)
Indifferent	86 (35.1%)	74 (32.5%)	160 (33.8%)
Negative	86 (35.1%)	76 (33.3%)	162 (34.3%)
Total	245 (100.0%)	228 (100.0%)	473 (100.0%)
$p > 0.05$, $df=2$, $X^2= 1.07$			

Table 6: Statistical analysis of „Reaction to food“ in both treatment groups

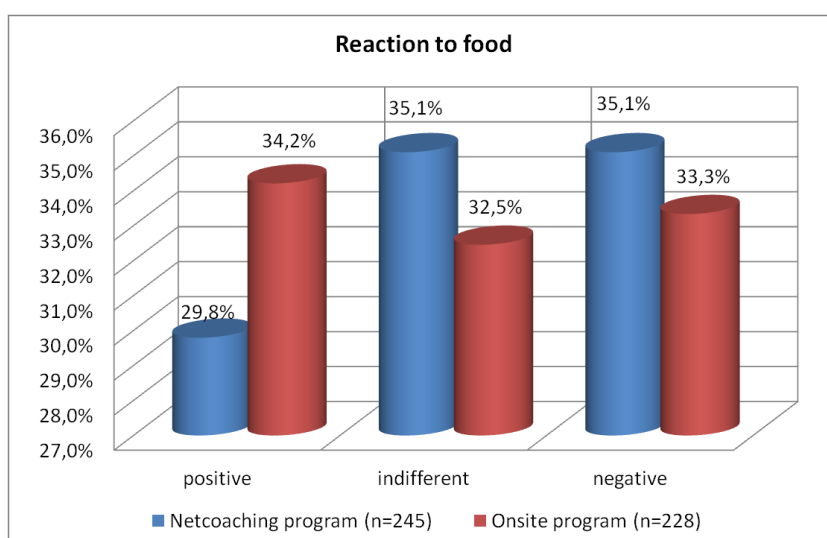


Figure 12: Reaction to food in both treatment groups

2.1.10 Duration of tube feeding

It has been evaluated, how long the child has been tube fed prior to participating in the program. It was calculated how many months the child has been tube fed on the date of admission to one of the treatment options.

Program	Duration of tube feeding in months				
	Mean	Median	SD	Min.	Max.
Netcoaching (n=245)	16.85	12	15.27	1	97
Onsite (n=228)	25.61	20	21.39	1	128
$p < 0.05$, Mann-Whitney-U= 19688.0					

Table 7: Statistical analysis of „duration of tube feeding in months“ in both treatment options

Using Mann-Whitney- U-Test, it is shown, that there are significant differences about the duration of tube feeding between the treatment groups. Children who participated in the Netcoaching program, showed significantly shorter intervalls of tube feeding (MD: 12 months vs 20 months, mean: 16.85 months vs 25.61 months).

As patients in the Netcoaching group were significantly younger, the absolute duration of tube feeding could not be considered as completely expressive.

Therefor, the percentage of lifetime a child has been tube fed was calculated for each single child. It could be shown that the children have been fed between 2% (Netcoaching group) – 3%(onsite group) and 100% of their lifetime. Most patients have been tube fed since their birth. Median was equal in both treatment groups– using Mann-Whitney-U, no significant differences could be shown between the groups.

Program	%Lifetime of tube feeding				
	Mean	Median	SD	Min.	Max.
Netcoaching (n=245)	83.11%	100%	23.31	2%	100%
Onsite (n=228)	82.92%	100%	25.87	3%	100%
$p > 0.05$, Mann-Whitney-U= 27366.5					

Table 8: Statistical analysis of „percentage lifetime tube feeding“ in both treatment groups

2.1.11 Underlying medical diagnoses

The underlying medical diagnosis of the children were widely spread, most children had multiple medical problems. Out of all diagnosis, a pediatrician decided which was the „main diagnosis“ of the child. Based on this information, the diagnosis were allcoated to specific groups of diagnoses. Classification of Trabi et al. (58) has been used and modified.

The following ten diagnostic groups have been used. Each patient could be included in one of the ten groups, the main diagnoses are listed below:

1. Prematurity/birth complications

- a. Immaturity (24-36 completed weeks of gestation) (N=96)
- b. Intrauterine growth retardation (N=1)
- c. PDA Ligation (N=1)
- d. Peripartal asphyxia (N=3)

2. Malformation of the heart/cardiological disease

- a. Ventricular septal defect (N=17)
- b. AVSD (N=1)
- c. Atrial septal defect (N=3)
- d. Hypoplastic left-heart syndrome (N=6)
- e. Pulmonary (valve) stenosis (N=3)
- f. Dextrocardia (N=1)
- g. Coarctation of the aorta (N=4)
- h. Atresia of the aorta (N=1)
- i. Tetralogy of Fallot (N=6)
- j. Dilatative cardiomyopathy (N=3)
- k. ALCAPA (N=1)
- l. Truncus arteriosus communis (N=1)
- m. Myocarditis (N=2)
- n. Heart insufficiency (N=2)
- o. Hypoplastic right-heart syndrome (N=1)
- p. Heart failure (N=1)
- q. Double inlet ventricle (N=2)
- r. Atresia A.pulmonalis (N=3)

- s. TAPVR (N=2)
- t. Single ventricle (N=1)

3. *Metabolic disease*

- a. OTC deficiency (N=1)
- b. Hyperinsulinism (N=2)
- c. Cytochrom-C-Oxidase deficiency (N=1)
- d. Neonatal hepatitis (N=1)
- e. Infantile botulism (N=1)
- f. Pancreatitis (N=1)
- g. Disorder of lymphatic vessels (N=1)
- h. Blackfan-Diamond-Anemia (N=1)
- i. Neonatal Hyperammonemia (N=1)
- j. Unspecified mitochondrial disease (N=1)

4. *Malformation or disease of the gastrointestinal tract*

- a. Gastroesophageal reflux disease (N=32)
- b. Short bowel syndrome (N=1)
- c. Omphalocele (N=3)
- d. Esophageal atresia (N=17)
- e. Hirschsprung disease (N=1)
- f. Dysmotility of esophagus (N=1)
- g. Hiatal hernia (N=1)
- h. Dysphagia (N=4)
- i. Lip and/or cleft palate (N=6)
- j. Atrophia microvillus (N=1)
- k. Anal atresia (N=1)
- l. Recurrent vomiting (N=1)
- m. Pyloric stenosis (N=1)
- n. Transesophageal fistula (N=1)

5. *Genetic syndroms/chromosomal aberrations*

- a. 22q11 deletion syndrome (N=6)
- b. CHARGE syndrome (N=13)
- c. Aberration chromosome 5 (N=1)
- d. Noonan syndrome (N=2)
- e. Down syndrome (N=14)
- f. Costello syndrome (N=6)
- g. Pierre Robin syndrome (N=5)
- h. Fetal alcohol syndrome (N=4)
- i. Escobar syndrome (N=1)
- j. 1q21.1 syndrome (N=1)
- k. 8p deletion syndrome (N=1)
- l. Malformation syndrome –unknown origin (N=5)
- m. Netherton syndrome (N=1)
- n. Wolf-Hirschhorn syndrome (N=3)
- o. Di George syndrome (N=2)
- p. Prader-Willi syndrome (N=1)
- q. Monosomy 1q25.3-q31.1 (N=1)
- r. Cystic fibrosis (N=3)
- s. Marshall-Smith syndrome (N=1)
- t. 1q36 deletion syndrome (N=1)
- u. Oculo-facio-cardio-dental syndrome (N=1)
- v. Cornelia de Lange syndrome (N=4)
- w. Kabuki syndrome (N=1)
- x. Goldenhar syndrome (N=3)
- y. Branchio-oculo-facial syndrome (N=1)
- z. Collins-Simons syndrome (N=1)
- aa. 3q28 deletion syndrome (N=1)
- bb. VACTERL association (N=2)
- cc. Congenital dwarfism (N=1)
- dd. Deletion chromosome 7 (N=1)
- ee. Cardio-facial-cutaneous syndrome (N=1)
- ff. Stickler syndrome (N=1)
- gg. Turner syndrome (N=2)

- hh. Athrogyrosis multiplex congenita (N=2)
- ii. 4q21.23q21.21 syndrome (N=1)
- jj. Allagille syndrome (N=1)
- kk. Chromosomal anomaly t15,7 (N=1)
- ll. Seckel syndrome (N=1)
- mm. Moebius syndrome (N=1)
- nn. Smith-Lemli-Opitz syndrome (N=3)
- oo. Pfeiffer syndrome (type II) (N=1)
- pp. Oto-palato-digital syndrome (N=1)
- qq. Klinefelter syndrome (N=1)
- rr. Silver Russel syndrome (N=1)
- ss. Trisomy 17 (N=1)
- tt. imagE syndrome (N=1)

6. *Psychiatric/psychosomatic disease, NOFTT*

- a. Infantile anorexia (N=3)
- b. Autism (N=7)
- c. Non-organic failure to thrive (N=16+1)
- d. Complex regional pain syndrom (N=2)
- e. Disorder of state regulation (N=1)
- f. Global developmental delay (N=2)
- g. Attachment disorder (N=3)
- h. Obsessive compulsive disorder (N=1)

7. Neurological conditions

- a. Traumatic brain injury (N=3)
- b. Hypoplasia of the corpus callosum (N=1)
- c. Hypoxic-ischemic encephalopathy (N=5)
- d. Cerebral palsy (N=11)
- e. Epilepsy (N=5)
- f. Agenesis of corpus callosum (N=5)
- g. Unspecified neurological disease (N=1)
- h. Spastic quadriplegia (N=1)
- i. Hydrocephalus (N=1)

- j. Intracerebral hemorrhage (N=1)
- k. Demyelination of the brain (N=1)
- l. Vein of galen malformation (N=1)
- m. Macrocephaly (N=2)
- n. Myopathy (N=1)
- o. Stiff baby syndrome (N=1)
- p. Cerebral edema (N=1)
- q. Brain aneurysm (N=1)
- r. Spina bifida aperta (N=1)

8. *Malformation or disease of respiratory tract*

- a. Respiratory insufficiency (N=1)
- b. Congenital diaphragmatic hernia (N=9)
- c. Paresis of N.phrenicus (N=1)
- d. Laryngomalacia (N=4)
- e. Surfactant-C-deficiency (N=3)
- f. Malformation of the pharynx (N=1)
- g. Interstitial lung disease (N=1)
- h. Tracheomalacia (N=1)
- i. Lung dysplasia (N=3)
- j. Obstructive lung disease (N=1)
- k. Floppy larynx (N=1)
- l. Paresis of vocal chord (N=1)
- m. Breath-holding apnea (N=1)

9. *Oncology and hematology*

- a. Wilm's tumor (N=1)
- b. Leucemia (N=2)
- c. AT/RT brain tumor (N=1)
- d. Rhabdomyosarcoma (N=1)
- e. Lymphangioma (N=1)

10. Renal diseases

- a. Megavesica (N=1)
- b. Renal insufficiency (N=7)
- c. Nephrotic syndrome (N=1)

101 patients were included in group 1, the majority's (N=96) main diagnosis was Immaturity (birth between 24 and completed 36 weeks of gestation).

61 children suffered from congenital malformations of the heart or a cardiological disease – 17 of them were diagnosed with a ventricular septal defect, six with Tetralogy of Fallot and also six children with Hypoplastic left-heart syndrom. Most of the children have undergone open-heart surgeries prior to the tube weaning trial.

In group 3 (metabolic diseases), the underlying diagnoses were widely spread (10 different diagnoses for 11 children), two children suffered from Hyperinsulinism. Overall, 71 children were diagnosed with malformations or diseases of gastrointestinal tract. The largest group were patients suffering from GERD (N=32), followed by those with esophageal atresia (N=17) and cleft and/or lip palate (N=6). The largest group is group 5 (genetic syndroms/chromosomal aberrations). 109 patients with 46 different diagnoses were included in this group. Most common diagnoses were Down Syndrom (N=14), CHARGE syndrome (N=13) as well as Costello syndrome (N=6) and 22q11 deletion syndrome (N=6). 35 patients were included in subgroup 6, the majority of them (N=16) diagnosed with non-organic failure to thrive, followed by Autism (N=7).

Neurological conditions were considered as main diagnoses in 43 children, most of them (N=11) suffered from cerebral palsy, 7 children were diagnosed with agenesis or corpus callosum, 5 suffered from hypoxic-ischemic encephalopathy. 28 children were included in subgroup 7, nine of them were born with a diaphragmatic hernia. The smallest group were children with oncologic/hematologic diseases. Five patients with five different diagnosis (Wilm's tumor, Rhabdomyosarcoma, brain tumor, Lymphangioma, Rhabdomyosarcoma) were included.

Nine children suffered from renal diseases, most of them (N=7) were diagnosed with renal insufficiency, some of them underwent NTX before participating in the tube weaning program.

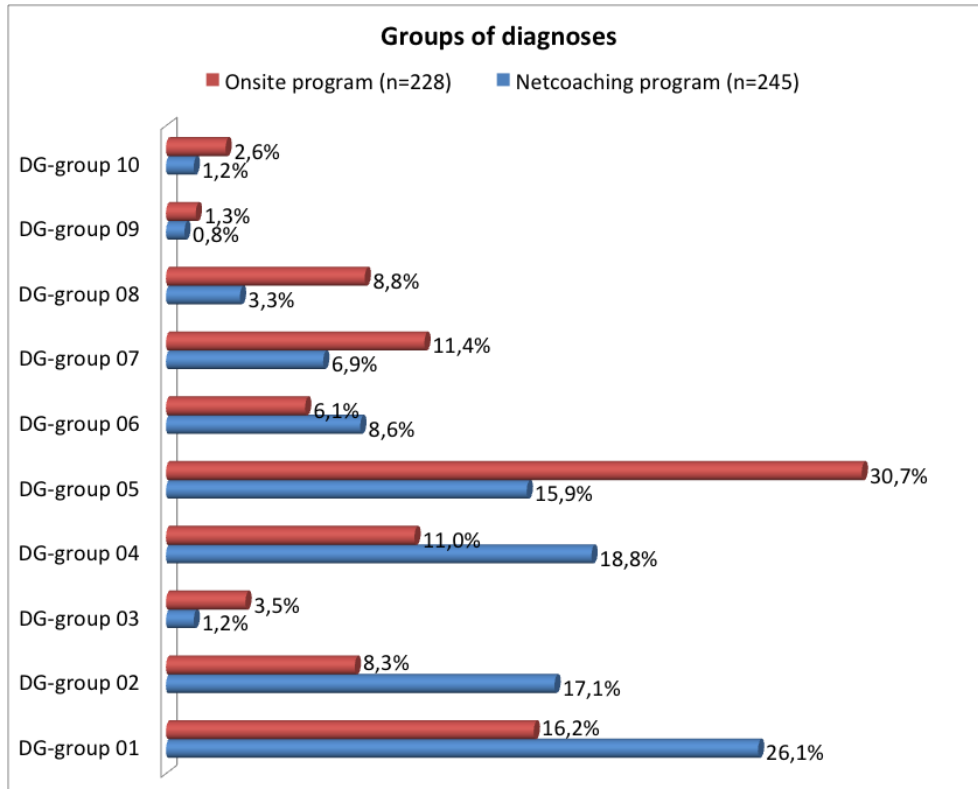


Figure 13: Groups of diagnoses for both treatment options

Table 9 shows that in eight out of ten treatment groups, the number of patients did not differ significantly between the two treatment options.

Only in subgroup 2 (malformation of the heart/cardiological disease), significantly more patients participated in the Netcoaching program whereas in subgroup 5 (genetic syndroms/chromosomal aberrations) more patients chose the onsite option.

DG-groups	Program			Total
	Netcoaching (n=245)		Onsite (n=228)	
prematurity/birth complications	n std. residual	64 (26.1%) 1.6	37 (16.2%) -1.7	101 (21.4%)
Malformation of the heart/cardiological disease	n std. residual	42 (17.1%) 1.9	19 (8.3%) -1.9	61 (12.9%)
Metabolic disease	n std. residual	3 (1.2%) -1.1	8 (3.5%) 1.2	11 (2.3%)
Malformation/ disease of the GIT	n std. residual	46 (18.8%) 1.5	25 (11.0%) -1.6	71 (15,0%)
Genetic syndroms/ chromosomal aberrations	n std. residual	39 (15.9%) -2.3	70 (30.7%) 2.4	109 (23.0%)
psychiatric/psychoso matic disease, non- organic FTT	n std. residual	21 (8.6%) 0.7	14 (6.1%) -0.7	35 (7.4%)
neurological conditions	n std. residual	17 (6.9%) -1.1	26 (11.4%) 1.2	43 (9.1%)
malformation/ disease of the respiratory tract	n std. residual	8 (3.3%) -1.7	20 (8.8%) 1.8	28 (5.9%)
oncology and hematology	n std. residual	2 (0.9%) -0.4	3 (1.4%) 0.4	5 (1.1%)
renal diseases	n std. residual	3 (1.2%) -0.8	6 (2.6%) 0.8	9 (1.9%)
Total	n	245 (100.0%)	228 (100.0%)	473 (100.0%)
$p < 0.01$, $df=9$, $X^2= 42.26$				

Table 9: Statistical analysis of „groups of diagnoses“ in both treatment groups

3. Results

The following chapters provide the results of the study, starting with the primary objective but also dealing with other factors (weight development, duration, client satisfaction as well as long-term outcomes of the program).

3.1 Primary objective: Success of treatment

Success of treatment was defined as the primary objective. The outcome variables were defined as followed:

1. Completely weaned, only oral intake
2. Partially weaned
3. Weaning trial without success
4. Patient's interruption of treatment on parental decision

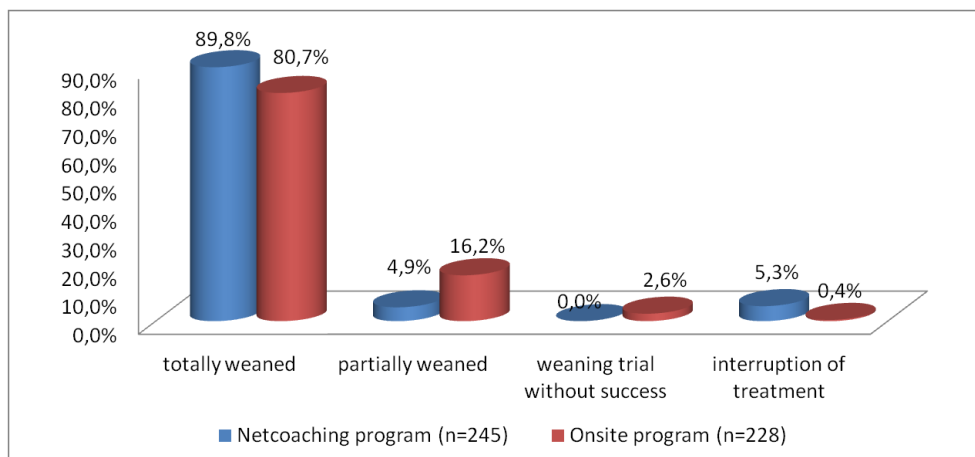


Figure 14: Outcomes in both treatment options

Figure 14 shows the outcomes in both treatment options. 89.8% of all patients in the Netcoaching group and 80.7% of all children have been weaned off their feeding tubes at the end of the treatment. 49 children (12 in the Netcoaching group vs 37 in the onsite group) got partially weaned – they were able to eat substantial amounts orally but needed additional tube feeds to sustain their nutritional needs. No weaning trial in the Netcoaching group whereas 6 weaning trials in the onsite group were considered as unsuccessful (meaning that the child stays on 100% enteral nutrition after completing the treatment).

Reasons for an unsuccessful weaning trial were:

- a progressive disease
- unusual massive weight loss during the weaning trial (N=2)
- severe oral aversion
- dysphagia with unsafe swallow (ruled out during the treatment)
- achalasia (ruled out during the treatment).

13 parents in the Netcoaching treatment decided to interrupt the program whereas only one parent in the onsite group decided to leave the treatment early.

Outcomes	Program			Total
	Netcoaching (n=245)		Onsite (n=228)	
completely weaned	n	220 (89,8%)	184 (80.7%)	404 (85.4%)
	std. residual	0.7	-0.8	
partially weaned	n	12 (4.9%)	37 (16.2%)	49 (10.4%)
	std. residual	-2.7	2.8	
weaning trial without success	n	0	6 (2.6%)	6 (1.3%)
	std. residual	-1,8	1.8	
interruption of treatment	n	13 (5.3%)	1 (0.4%)	14 (3.0%)
	std. residual	2.1	-2.2	
Total	n	245 (100.0%)	228 (100.0%)	473 (100.0%)
$p < 0.05$, $df=3$, $X^2= 31.68$				

Table 10: Statistical analysis of „outcome“ in both treatment groups

Statistical analysis showed that outcome group 1 and 3 do not differ between the treatment groups – nearly the same percentage of patients could be completely weaned in both treatment options, unsuccessful weaning trials did not take place (Netcoaching group) or were very seldom (onsite group – 2.6% of all patients).

Significantly more patients left the onsite treatment partially weaned (16.2% vs 4.9%), whereas significantly more patients interrupted the treatment in the Netcoaching group (5.3% vs 0.4%).

3.2 Further variables

3.2.1 Does the underlying medical diagnosis influence the outcome?

It was analysed, whether the specific outcome groups showed an influence on the outcome (weaned – not weaned) of the two treatment options.

It could be shown that 91% of diagnostic group 1, 90% of group 2, 91% of group 3, 89% of group 4, 75% of group 5, 89% of group 6, 86% of group 7, 60% of group 9 and 100% of group 10 could be weaned completely.

Only one significant result could be found, children diagnosed with genetic syndroms/chromosomal aberrations could unfortunately but understandably be weaned significantly less often than children of all other subgroups.

DG-groups	Outcomes			Total
		Totally weaned (n=404)	Not (completely) weaned (n=69)	
complicated prematurity/ birth complications	N std. residual	92 (22.8%) 0.6	9 (13.%) -1.5	101 (21%)
congenital malformation of the heart	N std. residual	55 (13.6%) 0.4	6 (8.7%) -1.0	61 (12.9%)
congenital metabolic disease	N std. residual	10 (2.5%) 0.2	1 (1.4%) -0.5	11 (2.3%)
malformation/ disease of the GIT	N std. residual	63 (15.6%) 0.3	8 (11.6%) -0.7	71 (15,0%)

genetic syndroms/ chromosomal abnormalities	N std. residual	82 (20.3%) -1.2	27 (39.1%) 2.8	109 (23.0%)
psychiatric disease of child or parents/ non-organic FTT	N std. residual	31 (7.7%) 0.2	4 (5.8%) -0.5	35 (7.4%)
neurological conditions	N std. residual	35 (8.7%) -0.3	8 (11.6%) 0.7	43 (9.1%)
malformation/ disease of the respiratory tract	N std. residual	24 (5.9%) 0.0	4 (5.8%) 0.0	28 (5.9%)
oncology and hematology	N std. residual	3 (0.7%) -0.6	2 (2.9%) 1.5	5 (1.1%)
renal problems	N std. residual	9 (2.2%) 0.5	0 (0.0%) -1.1	9 (1.9%)
Total	N	404 (100.0%)	69 (100.0%)	473 (100.0%)
$p < 0.05$, $df=9$, $X^2= 15.65$				

Table 11: Statistical analysis of underlying medical diagnosis and „outcome“

3.2.2 Weight development during treatment

As the Graz model of tube weaning goes along with swift but supervised reduction of tube feeds, it is very important to keep an eye on the weight loss during the treatment.

Program	Weight development during treatment in kg				
	Mean	Median	SD	MaxLoss	MaxGain
Netcoaching (n=245)	-0.30	-0.24	0.61	-2.5	+1
Onsite (n=228)	-0.53	-0.49	0.62	-2.7	+2.6
$p < 0.05$, Mann-Whitney-U= 19688.0					

Table 12: Statistical analysis of „weight development“ in both treatment options

Table 12 shows that children lost a maximum of 2.5 kg in the Netcoaching group and a maximum of 2.7 kg in the onsite group. The maximum weight gain during the treatment was 1kg in the Netcoaching group and 2.6kg in the onsite group. Median weight loss showed a significantly higher loss in the onsite group (0.49 kg vs 0.24 kg). Considering the fact that children in the Netcoaching group were significantly younger and therefore also smaller and lighter, the loss in the onsite group was clearly greater and for reasons of comparability the percentages of weight loss were evaluated.

Program	Weightloss in relation to start weight in %				
	Mean	Median	SD	MaxLoss	MaxGain
Netcoaching (n=245)	-2.13	-2.76	6.51	-20.91	28.26
Onsite (n=228)	-4.43	-4.74	4.94	-15.88	20.00
$p < 0.05$, Mann-Whitney-U= 20146.0					

Table 13: Statistical analysis of „Weight loss in %“ in both treatment groups

Statistical analysis showed that even if calculating the percentage of weight gain/loss during the treatment, the patients in the onsite group lost significantly

more weight (MD: 4.43% of body weight vs -2.13% of body weight in the Netcoaching group).

3.2.3 Duration of treatment

Duration of treatment was evaluated for both treatment options. The onsite program is generally set as a 3 weeks treatment with the option of extension in some cases, longer periods of treatment are only possible for children who live nearby the hospital as patients from abroad are not able to stay for months.

In contrast to the time restricted and highly structured traditional inpatient setting, the Netcoaching option does not have a predefined duration, it lasts until 35 days after the very last tube feed if the child is in a stable weight condition with sufficient oral intake. In some rare cases, the children were discharged earlier on parental decision as they did so well. Maximum treatment time is 12 months, in some cases (e.g. severe illness(es) during the treatment, unexpected surgeries etc), this time slot can even be extended.

Program	Duration of treatment in days				
	Mean	Median	SD	Min.	Max.
Netcoaching (n=245)	85.05	62	71.26	12	488
Onsite (n=228)	21.99	18	20.18	3	175
$p < 0.05$, Mann-Whitney-U= 3157.5					

Table 14: Statistical analysis of „duration of treatment“ in both treatment groups

Due to this fact, statistical analysis showed significant differences regarding the duration of treatment in both treatment groups.

The median of the onsite treatment was 18 days which is the time for the standardized three weeks program (Monday week one – Friday week three). Minimum treatment duration has been only three days, maximum duration was 175 days (this duration was only achieved in a few outpatients who lived near Graz and had the chance to come to the outpatient clinic regularly with larger time intervalls between the visits than most other patients who came from far away/abroad).

In the Netcoaching group, the median treatment duration was 62 days, almost three months. Maximum were 488 days (>1 year) in a very complicated case, minimum were 12 days.

3.2.4 Client satisfaction

To assess the parent's satisfaction with the tube weaning program, a standardized questionnaire was used after completion of treatment.

3.2.4.1 The Client Satisfaction Questionnaire (CSQ-8)

The Client Satisfaction Questionnaire (67) has been specifically developed to assess client's satisfaction with health services. Items are drawn from a large pool, originally covering nine conceptual domains of client satisfaction, resulting in a 30-Item-Questionnaire. Shorter versions have been developed, the most widespread use is the version consisting of eight items (CSQ-8) which has also been used in this study. Psychometric results of CSQ-8 show very good internal reliability (Cronbach's Alpha 0.83-0.93), construct validity ranges from .6-.8 (68). The CSQ-8 is available in different languages, in this study, the English as well as the German version have been used. CSQ-scores are collapsed into three levels: low (8-20), medium (21-26) and high (27-32) (67).

3.2.4.2 Outcomes of the CSQ-8

Several parents of patients who completed either in the onsite or Netcoaching treatment option between January and November 2012, were asked to fill out the Client Satisfaction Questionnaire (CSQ-8) on an anonymous basis (no identification of the person who filled out the questionnaire was possible). The onsite group used the paper-pencil-version of the Questionnaire, whereas the Netcoaching group got an online form. All in all, 54 persons filled out an CSQ-8. 31 parents of children participating in the onsite program as well as 23 parents of patients participating in the Netcoaching program filled out the questionnaire completely, their data could be used for statistical analyses.

Program	Client Satisfaction Questionnaire				
	Mean	Median	SD	Min.	Max.
Netcoaching (n=23)	30.61	32	2.52	22	32
Onsite (n=31)	29.16	30	3.22	21	32
p>0.05, Mann-Whitney-U-Test=261.00					

Table 15: Statistical analysis of the „Client Satisfaction Questionnaire“

It could be shown that there were no significant differences regarding the client's satisfaction between the two treatment groups (MD Netcoaching group: 32, MD onsite group: 30). As CSQ scores can range between 8 and 32, the mean score is declared for score 20, therefore, both groups can be considered as being remarkably satisfied with the tube weaning program.

3.2.5 Long-term outcomes

Long-term outcomes of both treatment options have also been evaluated.

All patients who completed one of the two tube weaning programs between January 1st, 2009 and Dec 31st, 2013 were contacted, either via e-mail or personally/data (in the rare cases when the child was still patient of the University Children's Hospital Graz). Parents of patients who interrupted the treatment themselves have not been contacted.

For all patients who started the treatment before January 1st, 2013, the e-mail has been sent in January 2014. For all patients who started the tube weaning in 2013, the e-mail has been sent in October 2014.

Parents were asked to fill out the following details (either in German, English or French):

- Name of the child
- How is your child fed at the moment?
 - o Exclusively oral
 - o Completely by feeding tube
 - o Mixed tube/oral
- If your child is fed completely oral: has the tube been removed already?
 - o Yes
 - o No
- If yes, when?

- If your child is still fed by tube, which type of tube does your child have?
 - o Nasogastric tube
 - o G-tube/Button
 - o J-tube
 - o JET-PEG
- Current weight of your child (in kg)? WO SIND DIESE ZAHLEN??
- Current height of your child (in cm)?
- Any medical complications since the weaning (e.g. severe illnesses, surgeries...)
- Which kind of food does your child eat orally (e.g. normal child diet, selective diet, formula...)
- If your child is fed via a tube – which kind of food and which amount (in ml) does it get?

For this thesis, the question “How is your child fed at the moment” has been evaluated. The important question about weight development is not part of this thesis but will be analysed in the near future.

458 patients have been included to the study. 256 (55.9%) follow-up questionnaires have been filled out, their data was used for statistical analyses.

Total response rate	Follow-up participants (n=458)	Percent (%)
Answer follow-up	256	55.90%
No answer follow-up	202	44.10%
Total participants	458	100.00%

Table 16: Statistical analysis of response rate to follow-up Questionnaire

Parents of three patients of 458 informed us, that their child has died in the meantime due to the underlying medical problem (progressive mitochondropathy, acute pneumonia in a child with severe lung hypoplasia, genetic syndrome). Of course their data has not been included in further analyses.

The response rate of all 5 consecutive years have been evaluated. The lowest response rate was shown in the patients who participated in the tube weaning programs in 2009 (30.77%). They have been contacted 3-4 years after they completed the tube weaning program.

39 of 100 patients who took part in one of the treatment options in 2010 could be integrated in the long-term evaluation, whereas 61 of 100 of all patients who were treated in 2011 responded to the long-term evaluation.

The response rates of patients of the years 2012 (58/79;73.42%) and 2013 (71/98;72.45%) were the highest ones, they were nearly equally high.

Year	Follow-up received (n=253)	Total number of contacted patients
2009	24	78
%	30.77%	100.00%
2010	39	100
%	39.00%	100.00%
2011	61	100
%	61.00%	100.00%
2012	58	79
%	73.42%	100.00%
2013	71	98
%	72.45%	100.00%
Total	253	455
%	55.60%	100.00%

Table 17: Statistical analysis of „Response rate over the years of treatment“

Long-term outcomes of 256 patients have been evaluated. It could be shown, that 229 of 256 patients (89.45%) were fed completely orally. 18 patients (7.04%) were partially, 6 children (2.34%) exclusively tube fed (table 18).

Follow-up outcomes	Participants (n=256)	Percent %
Totally weaned	229	89.45%
Partially weaned	18	7.04 %
Completely tube fed	6	2.34%
Deceased	3	1.17%
Total participants	256	100.00%

Table 18: Follow-up outcomes

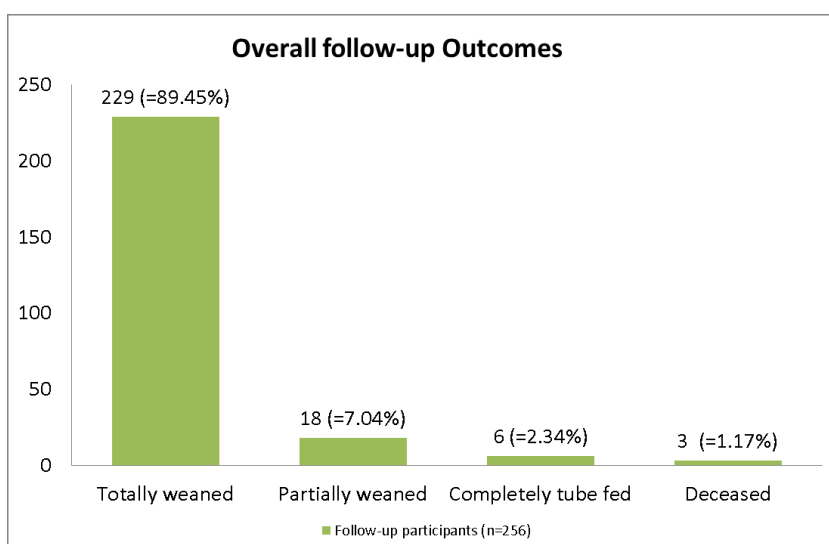


Figure 15: Follow up outcomes

When focusing on the long-term outcomes of the different treatment options, it could be shown that response rate of both treatment groups has been nearly equal. 134 follow-ups (53%) of Netcoaching patients, 119 follow-ups of onsite patients have been integrated in the evaluation.

126/134 children (94%) of the Netcoaching patients and 103/119 (87%) of the onsite patients were fed completely orally.

7/134 (5%) children participating in the Netcoaching program vs 11/119 (9%) taking part in the onsite program were partially tube fed, whereas only 1/134 of the Netcoaching patients (1%) and 5/119 of the onsite (4%) patients were completely tube fed.

Program	Follow-up outcome-groups			Total
	totally weaned	partially weaned	completely tube fed	
Netcoaching % within follow-up groups	126 (55.0%)	7 (38.9%)	1 (16.7%)	134 (53.0%)
Onsite % within follow-up groups	103 (45.0%)	11 (61.1%)	5 (83.3%)	119 (47.0%)
Total % within follow-up groups % within total	229 (100.0%) (90.5%)	18 (100.0%) (7.1%)	6 (100.0%) (2.4%)	253 (100.0%) (100.0%)
$p > 0.05$, Fisher exact test = 6.86				

Table 19: Follow-up outcome in the two treatment options

Furthermore, the development of the outcomes (end of treatment – follow-up) has been analyzed. Outcomes right after the treatment's completion was compared with long-term outcome.

221 of 232 patients who have been completely weaned after the treatment were still 100% orally fed at follow-up. 8 children who were originally completely tube weaned needed partial tube feeds at the time of long-term checkup.

Reasons for partial tube feeding after successful weaning were as followed:

- too little oral intake (N=2)
- progressive dysphagia due to spastic quadriplegia (N=1)
- upcoming surgery (plan to skip tube feeds again after surgery) (N=1)
- respiratory status became more compromised (N=2)
- St.p. stroke (N=1)
- Syringomyelia (N=1).

Reasons for complete tube feeding after successful weaning were as followed:

- development of severe dysphagia with aspiration (N=1)
- got diagnosed with a life-shortening progressive disease – lost swallow reflex (N=1).

9 of 20 patients (45%) who were partially weaned right after participating in the tube weaning program were still partially tube fed at the time of the follow up. Fortunately, 8/20 (40%) children who were discharged partially tube fed, are eating 100% orally at follow up. Three patients who were originally partially weaned, were put back to full tube feeds.

Only one patient whose treatment was not successfully at all participated in the long-term checkup, the patient was still completely tube fed at this time.

Outcome, right after treatment	Follow-up outcome-groups			Total
	totally weaned	partially weaned	completely tube fed	
totally weaned	221	9	2	232
% within follow-up groups	(97%)	(50.0%)	(33.3%)	(91.7%)
% within outcome after treatment	(95.2%)	(3.9%)	(0.9%)	(100.0%)
partially weaned	8	9	3	20
% within follow-up groups	(3 %)	(50.0%)	(50.0%)	(7.9%)
% within outcome after treatment	(40.0%)	(45.0%)	(5.0%)	(100.00%)
weaning trial without success	0	0	1	1
% within follow-up groups	(0.0%)	(0.0%)	(16.7%)	(0.4%)
% within outcome after treatment	(0.0%)	(0.0%)	(100.0%)	(100.0%)

Total	229	18	6	253
% within follow-up groups	(100.0%)	(100.0%)	(100.0%)	(100.0%)
% within outcome after treatment	(90.5%)	(7.1%)	(2.4%)	(100.0%)
% within total	(90.5%)	(7.1%)	(2.4%)	(100.0%)
p<0.05, Fisher exact test = 50.57				

Table 20: Statistical analysis of comparison of „outcome after treatment – follow up“

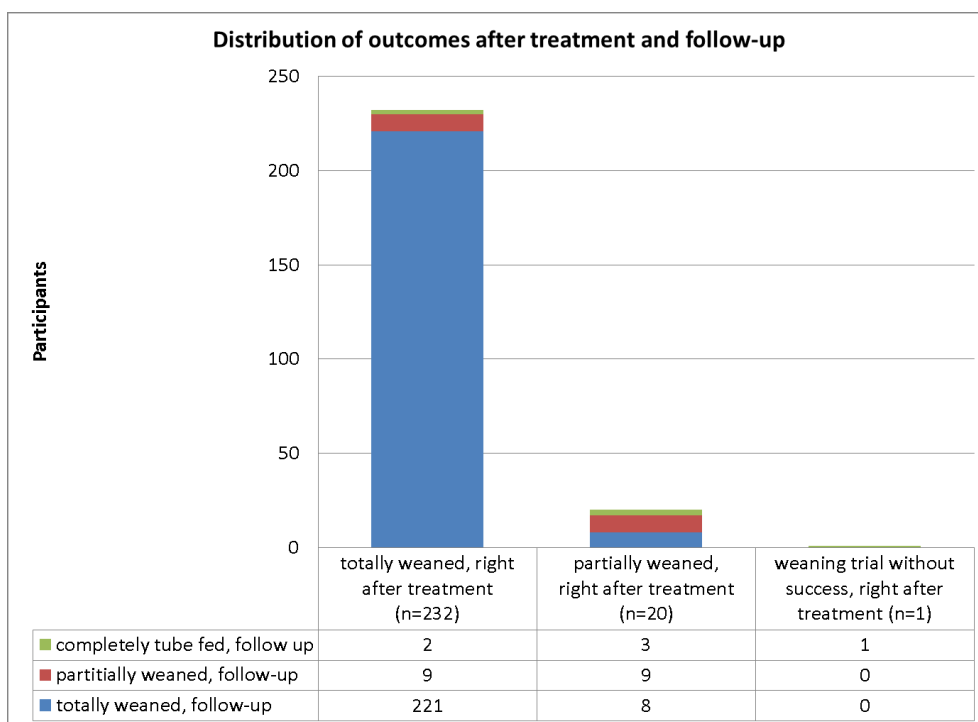


Figure 16: Comparison „Outcome after treatment – follow up“

4. Discussion

This study includes the largest sample of children (N=473) referred for the sake of tube weaning ever published. It compares two treatment options of the „Graz model of tube weaning“. The primary objective was to compare the outcomes of the onsite treatment option with those of the newly invented online program defined as Netcoaching option.

Furthermore, other variables were also evaluated: Biographic data (age, country of referral, gender), biometrical and medical data (diagnoses, weight development, type of tube, duration of tube feeding) as well as psychological (reaction to food, client satisfaction). Furthermore, the long-term success rate was analyzed.

It could be shown, that both treatment options showed a comparable outcome, both of them show that between 80-90% of all patients could be weaned completely and with lasting sustainability off their feeding tubes. The Netcoaching program results are even slightly (not significantly) better.

The significant differences regarding the rate of partially weaned patients may be related to the fact, that the onsite treatment lasts shorter and mostly has a predetermined duration. Furthermore, it takes place in a hospital setting, inpatients are admitted to the parent-child rooms) in the department of General pediatrics. This means that nosocomial infections can't be avoided completely. If a child catches e.g. a gastroenteritis, the weaning trial must be interrupted and the child loses some treatment days. As most of the children referred for the sake of tube weaning are medically fragile, such infections might be dangerous and potentially life-threatening. This was one of the reasons why the Netcoaching program was invented, since children e.g. after transplantation and under immunosuppression mostly cannot be admitted as inpatients into a traditional hospital setting.

To help those children who undergo the onsite treatment but are not fully weaned at the end of treatment, a telemedical aftercare-program was also introduced in 2012. It can be booked on a monthly basis with the goal to help the child make the last steps towards exclusive oral intake in the familiar environment at home.

The fact that significantly more parents interrupt the Netcoaching program might be related to different reasons. First of all, parents might find it easier to skip virtual communication without needing to justify this decision or offer specific arguments as would be the case in any hands-on venue. Furthermore, as the program is cheaper, the level of inhibition and commitment to interrupt the program might be smaller. But overall, only 13/245 children in the Netcoaching group interrupted the treatment, so interruption rate can still be considered as very small.

The fact that there was no „weaning trial without success“ in the Netcoaching group might be related to the fact, that medical data of those children who sign-up for the online treatment is evaluated very carefully by two independent pediatricians to make sure that the child is suitable for tube weaning from a medical point of view. All necessary examinations (e.g. videofluoroscopy) must be performed and evaluated prior to treatment start. Of course this is also the case for all children treated onsite, but in some cases additional examinations can be done at the clinic during the first part of the assessment phase, after which these children were included into the study. In some rare cases, these examinations proved that a weaning trial should not be continued at this point of time.

One could see that the patients participating in the Netcoaching program were significantly younger. This might be correlated to the fact, that there is no waiting list for the telemedical treatment, it can be started immediately after a positive medical assessment, whereas for onsite treatment one needs to wait for weeks up to a few months.

One reason why children with genetic syndroms were significantly less often weaned than children of all other subgroups might be correlated with the fact that these children often suffer from a great variety and severity of neurological and medical problems. Many of them are severely developmentally delayed and are born with multiple malformations, affecting different regions of the body.

Overall, the range and diversity of underlying medical diagnoses is quite large in both treatment groups. One can see that psychiatric/psychosomatic diagnoses are only a very little part of all underlying problems.

Weight development during a tube weaning program is always a topic which leads to discussion among professionals. Weaning mostly „costs“ some weight. Worries about negative influence on the general development are addressed regularly. However, Beckenbach (69) could show, that tube weaning even enhances development in different areas despite a weight loss during the tube weaning trial. As one can see, the patients in this study lost in average 2-5% of their body weight during the wean. Further analyses of long-term weight development are necessary and planned for the near future.

The duration of treatment is one of the largest differences between the two treatment options. Whereas the onsite treatment follows a predefined period of almost three weeks (Monday week 1 – Friday week 3) and can only be prolonged in exceptional cases, the Netcoaching treatment lasts until 35 days after the very last tube feed of a child (preconditions: stable weight condition, sufficient oral intake), no matter how long the child needs to get there (maximum duration: 12 months, in exceptional cases even longer). So if a child falls ill or undergoes a necessary surgical intervention, the weaning trial can be easily interrupted and continued when the child is completely healthy again.

Regarding children's „reaction to food“ (chapter 2.1.8) it could be shown that there were no significant differences between the two treatment groups, so there is no evidence that children participating in the one or the other group show more signs of food aversion, but it has to be kept in mind that this evaluation has only been based on parental view, it has not been based on professional evaluations. This topic has to be part of further investigations.

Client satisfaction showed high satisfaction rates in both groups. Nevertheless, the number of patients who filled out the CSQ-8 was not convincingly large, so further analyses of larger groups will be necessary.

Focusing on the long-term outcomes of the two programs, it could be shown, that *the majority of all* patients (97%) who were weaned successfully, are still completely orally fed one to four years after starting the weaning program.

This is a finding of high scientific interest, as long-term outcomes of tube weaning programs have barely been evaluated by now. Wilken (55) evaluated 40 children one to three years after they completed a home-based tube weaning program and could show that the growth of the children remained constant from initial evaluation to follow-up. This very interesting finding indicates the need of further analyses of long-term effects on growth and development of different tube weaning programs regarding different factors: weight development, general development, health status, eating behavior, quality of life.

Further studies on these themes are already planned by our multidisciplinary scientific group.

Patient's safety is always a concern when dealing with telemedical methods.

Silverman (66) states, that as well patients as providers of telehealth services expressed concern that treatment will be inhibited by the lack of physical contact between patient and provider. The Netcoaching program demands at least on onsite doctor who knows the patient will to be on board with the intended Netcoaching program they child has been assigned to. The parents agree prior to treatment to visit their doctor onsite in any case of medical emergency, injury, illness or other medical concern. At the end of the treatment, each medical doctor involved receives a final coaching report with specific recommendations.

Furthermore, parents are obliged to report about their child's behavior towards food each day as well as to fill out a protocol with weight and intake information. Frequent video analyses are also part of the program, nevertheless, the physical contact between patient/parents and medical doctor/therapist is missing and the communication is staggered.

This study provides a first comparison of the two treatment options of the „Graz model of tube weaning“. One of the major limitations of the study, is the missing prospective design and randomisation. As it was not possible from an ethical point of view (cost differences, travel effort, technical possibilities and abilities) to allocate patients prospectively to one of the two treatment groups, the choice of program in most cases was determined by the parents, except if there were clear contraindications (listed in chapter 2.1.2) for one of the two treatment options.

Furthermore, the study compared two programs following the same approach. For future work, it would be interesting to compare different models of tube weaning, e.g. behavioral methods.

Another limitation is, that in some subgroups of medical diagnoses, there was only quite a little number of patients included (e.g. only nine children with renal problems, only five children with oncological diseases). Therefore, correlated statistical findings have to be dealt with cautiously.

Overall, this study provides a first evaluation of a large sample of tube dependent infants and children referred for the exclusive sake of tube weaning. They have all been assessed and treated by the „Graz model of tube weaning“, either in the onsite or the online program. Children of both treatment groups had an equal chance to get completely weaned. It could be shown that the acceptance of the newly established Netcoaching program (started in 2009) increased significantly over the years (chapter 2.1.3). Considering medical aspects (eliminated risk of acquiring nosocomial infections), economic aspects (costs for the program, travel effort and expenses, no waiting list) as well as psychological aspects (no risk of resurgence of a hospital-related trauma in medically fragile children, family can stay at home in its familiar environment), the Netcoaching online method for tube weaning is a promising option besides traditional onsite weaning models.

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6. Appendix

6.1 Publications

Marinschek S, Dunitz-Scheer M, Pahsini K, Geher B, Scheer P. Weaning children off enteral nutrition by netcoaching versus onsite treatment: A comparative study. Journal of paediatrics and child health. 2014;50(11):902-7.

AIM

The Graz model of tube weaning has been internationally recognised as a successful and rapid tube weaning program. Beside the onsite treatment option, a telemedical counselling was specifically developed in 2009. This study aims to show outcomes of this newly invented treatment in a large sample of patients.

METHODS

Our retrospective open-label study compared success of onsite versus telemedical Graz-based weaning methods for patients with diverse clinical diagnoses with either nasogastric, gastric or jejunal tubes. Outcome variables were successful transition to oral feeds, partial transition to night tube feeds, and failure or interruption of intervention. Patients and physicians chose the intervention method.

RESULTS

Complete weaning was achieved in 153 of 169 (90.5%) children in the netcoaching group versus 170 of 209 (81.3%) of those opting for onsite treatment (no significant differences, $P > 0.05$). Higher partial weaning rates were observed in the onsite group (15.3% vs. 4.7%, $P < 0.01$, degrees of freedom = 3, $\chi(2) = 22.76$). There were no significant differences regarding the outcomes 'weaning trial without success' (netcoaching: 0% vs. onsite: 2.9%, $P > 0.05$) and 'interruption of programme' (netcoaching: 4.7% vs. onsite: 0.5%, $P > 0.05$) between the two groups.

CONCLUSION

Despite limitations of study design, we have demonstrated similar efficacy of Graz-based less expensive netcoaching versus more expensive onsite intervention in a

large referral population with chronic tube dependency with the majority transitioning to complete oral feeds.

Dunitz-Scheer M, Marinschek S, Beckenbach H, Kratky E, Hauer A, Scheer P. Tube dependency: a reactive eating behavior disorder. ICAN. 2011;3(4):209-15

This article focuses on the issue of tube dependence (TD) in infancy and early childhood. The condition occurs in patients after temporary tube feeding and must be considered as an unintended side effect of modern treatment practices affecting young patients reactively. Whereas some recent literature has described small samples of enterally fed children being exposed to certain weaning programs, the particular phenomenon of unintentional dependence has not been discussed. A tube-dependent child remains tube fed although his/her medical condition and developmental potential would allow the transition to oral nutrition. Children with TD show characteristic symptoms such as food refusal and opposition to any oral feeding attempts. They often suffer from additional episodes of vomiting, nausea, gagging, and retching and in some cases develop severe failure to thrive. Parents of affected children get involved as codependents engaged in constant preparations of the next tube feeds. In this situation, families can become obsessed about wanting their child to learn to eat by himself/herself, ending up in intrusive feeding patterns. Professionals tend to make parents responsible for the behavioral aspects of the condition, but the diagnostic shift of TD into a behavioral category will not help solve the problem. The development of TD can be prevented if typical symptoms are recognized early and effective tube weaning is implemented. Because therapeutic programs exist, the fate of remaining tube dependence should be prevented. This article presents a first overview of a large sample of tube-dependent infants who had been referred specifically for the exclusive sake of tube weaning.

6.2 Conference proceedings

Khan Z, Marinschek S, Pahsini K, Dunitz-Scheer M

Nutritional status based on growth parameters in a large cohort of medically fragile children receiving enteral nutrition support.

2nd International Conference on Nutrition and Growth; Jan 30-Feb 1, 2014; Barcelona. 2014. [Poster]

Marinschek S, Dunitz-Scheer M

STATE OF THE ART SEMINAR: Prävention, Management und Entwöhnung enteraler Ernährung im Kleinkindalter.

5. Wiener Postgraduelle Fortbildung "Essstörungen und assoziierte Krankheitsbilder"; 15.03.2013; AKH Wien. 2013. [Oral Communication]

Marinschek, S; Pahsini, K; Dunitz-Scheer, M; Scheer, PJ.

Evaluation of a large international sample of tube dependent infants receiving long-term enteral feeding.

1st International Conference on Nutrition and Growth; MARCH 1-3, 2012; Paris, FRANCE. 2012. [Poster]

Pahsini, K; Marinschek, S; Dunitz-Scheer, M; Scheer PJ

Non-nutritional variables of long-term enteral feeding in infancy and childhood

Long term tube (enteral) feeding: exploring dilemmas and controversies; OCT 4-5,2012; Glasgow, UK. 2012. [Poster]

Marinschek, S.

Retrospective analysis of a large sample of medically fragile children with tube dependence and other early eating behavior disorders

Kongress Essstörungen; OCT 20-22,2011; Alpbach, AUSTRIA. 2011. [Keynote lecture]

6.3 Medical questionnaire – English version

Biographic data

Name of the child:

Date of birth:

Mother's name:

Father's name:

Phone number:

E-mail-address:

Postal address:

What's your child's native language?

Do you have more children?

Child's biometric and medical data

Current weight:

Current height:

Lowest weight last year (in kg):

Lowest weight in the last three months (in kg):

Highest weight ever (in kg):

Weight at birth (in kg):

Length at birth (in cm):

Head circumference at birth (in cm):

How was your child born?

- spontaneous VD
- caesarian section

In which gestation week?

Actual diagnoses of your child:

In the past diagnosed diseases:

Important earlier interventions like operations, etc.:

Child's behavior with food

How does your child react to food?

- Positive
- Indifferent
- Negative

How does your child react to seeing food?

- Positive
- Indifferent
- Negative

How does your child react to smelling food?

- Positive
- Indifferent
- Negative

How does your child react to tasting food?

- Positive
- Indifferent
- Negative

How does your child react being fed?

- Positive
- Indifferent
- Negative

How does your child react to swallowing food?

- Positive
- Indifferent
- Negative

Does your child dislike touching and playing with food?

- Yes
- No

Is there something your child really likes to eat?

What was the first food your child ate and liked?

Does your child eat sweets or cracker?

- Yes
- No

Does your child drink water?

- Yes
- No

When sugar is added?

- Yes
- No

Are there any complications while or after tube feeding your child?

- Nausea
- Vomiting
- Sweating
- Skin irritation
- No hunger
- Retching/gagging
- Granulation tissue
- Other problems:

Parental behavior:

How do you react when your child refuses food?

- Relaxed
- Not relaxed
- Anxious
- Intrusive

Is someone else in your family suffering from an eating disorder?

- Yes
- No

Do you keep a special diet?

Nutrition:

How is your child fed at the moment?

- NG-Tube
- PEG-Tube
- PEG-Button
- NG tube
- JEG-Tube

Since when does your child have the tube?

My child had more than one tube:

Why did your child get a tube?

What was your child's weight when it received the tube (in kg)?

Did your child ever suffer from infections of the oral cavity?

What kind of food does your child get by tube?

How much food does your child get via the tube per day (in ml)?

How much food does your child eat orally per day (in ml)?

How much water does your child get additionally?

How many calories does your child approximately take in per day?