

## Research Article

### Feeding Aversion in Children with Neurodisability and its Assessment Using CEBQ: A Tertiary Feeding Clinic Experience

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#### Abstract

**Background:** Children with history of prematurity and developmental delay have feeding problems not solely attributed to oropharyngeal structural and motor impairments. Aversion to food and feeding can occur in normal development, but can be severe in various neurodevelopmental syndromes. There is lack of a validated tool to classify and measure the severity of the problem.

**Objective:** The aim of this study was to define the spectrum of aversive feeding behaviours seen in children with neurodisability and assess the usefulness of Child Eating Behaviour Questionnaire (CEBQ) to systemically delineate eating behaviours amongst these children.

**Methods:** This study was done in two phases. In phase one 38 case notes of patients attending the multidisciplinary feeding clinic were analyzed retrospectively for a period of six months. The details of description of their feeding problems, use of antireflux medications and gastrostomy were recorded. In phase two we prospectively used the CEBQ assessment tool on 16 patients showing aversion to food and feeding. Mean sub-domain scores of CEBQ were compared to population norms.

**Results:** In phase one 50% (19/38) patients had feeding difficulties unrelated to oropharyngeal structural or motor problems. The behavioural feeding problems were described as aversion to food and feeding, selective eating, drinking excess fluids, poor oral intake,

dislike for solids, disinterest in feeding, slow feeding and aversion for fluids. In phase two 16 patients with aversion to food or feeding and other such behaviours were assessed using CEBQ. Significant group differences in mean CEBQ sub domain scores compared to population norms were noted for food responsiveness (mean CEBQ score 1.75 vs. population mean 2.20), desire to drink (1.92 vs. 2.60), emotional overeating (0.78 vs. 1.90), satiety response (2.5 vs. 3) and enjoyment of food (2.8 vs. 3.5). Subdomain scores were reflective of non-rewarding experience of feeding in this group.

**Conclusion:** This study identifies that feeding difficulties not related to oropharyngeal structural or motor impairment can be complex to assess and manage needing feeding gastrostomy. CEBQ has affirmed non-rewarding experience of feeding in this group and with its limitations can be helpful assessment of feeding aversion.

#### Introduction

#### Background

Feeding disorders are common in children with neurodevelopmental conditions [1,2] and the estimated prevalence of feeding problems varies between studies from as low as 33 to as high as 80% [3,4]. Such a wide variation in the estimated prevalence is part due to the heterogeneous aetiologies of children in the studies. These children not only vary severity and type of feeding problem itself but also have motor, sensory, cognitive and behaviour co-morbidities which impact on feeding. Premature children or ones with developmental delay have a feeding difficulty not explained by physical or motor impairments [5]. Lack of objective oromotor dysfunction on clinical examination or normal investigations such as video fluoroscopy can be confusing to clinicians and parents for understanding their feeding behavior and unable to explain why these children not transition to a developmentally appropriate feeding level. These children have difficulty in adapting to food textures, utensil in mouth, disinterested in food or not just hungry. These children can be hard to manage for therapists and clinician and may be empirically treated for gastroesophageal reflux, require nasogastric tube or have feeding gastrostomy. To date, no standardized measures have been developed to evaluate these aversive feeding behaviour difficulties in children with neurodisability. There is no consensus about the description of these behaviours and terms like chronic food refusal [6-9], oral sensory dysfunction [10], infantile anorexia nervosa [11], food neophobia [12], conditioned food or taste aversion [13], can be used to describe the problem or aetiology. With this background the authors performed this two phased pilot study. For simplicity of description we have used the term feeding aversion to describe behaviours of not showing pleasure for food or feeding.

#### Aim

The aim of this study was to define the spectrum of aversive feeding behaviours seen in children with neurodisability and assess the usefulness of Child Eating Behaviour Questionnaire (CEBQ) to systemically delineate eating behaviours amongst these children.

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## Method

This study was conducted in multidisciplinary tertiary gastroenterology clinic. The team consisted of gastroenterologist, neurologist, speech and language therapist and dietician. In phase one of the study case notes for a period of six months were retrospectively reviewed. Feeding behaviours of all children seen in clinic were reviewed in the context of diagnosis and clinical examination. Patients were divided into two groups: with and without aversive feeding behaviours. Feeding behaviors of the former group were described. Use of anti reflux treatment and gastrostomy was compared between the two groups.

Phase two of this study was done prospectively. Children attending the clinic who were showing aversive feeding behaviors were assessed by using CEBQ completed by the parents. Patients with oromotor or structural abnormality were excluded from this assessment. CEBQ is a validated tool for assessing eight eating styles in children [14]. It is a parent rated questionnaire about rewarding (like food responsiveness, enjoyment of food, emotional overeating and desire to drink) and aversive (like slowness in eating, satiety response, fussiness and emotional under eating) feeding styles. It has 35 items with a score of 1-5. The results of all domains of the questionnaire were analyzed using Microsoft excel and scores compared to the normal population. This study was approved by the audit department for baseline service evaluation. Formal ethical approval was not obtained.

## Results

### Phase one

50% (19/38) patients had aversive feeding behaviours unrelated to oropharyngeal structural or motor problems. 17 of these patients had pure feeding behaviour problems and only two (patient 11 and 18 in group 1) had associated oromotor dysfunction. The feeding behavior problems were described as aversive behaviour to food and feeding, selective eating, drinking excess fluids, poor oral intake, dislike for solids, disinterest in feeding, slow feeding and aversion for fluids. Table 1 describes the diagnosis and co-morbidities of all children divided in group 1 with aversive behavior and group 2 with oromotor dysfunction.

6/19 (31%) patients in each group were on antireflux treatment, chisquare: p value of 1.000. 9/19 (47%) in group 1 had feeding gastrostomy as compared to 15/19 (79%) in group 2, chisquare: p value of 0.0436. 1/19 (5%) patient in group 1 had nasogastric tube as opposed to 3/19 (16%) in group 2, chisquare: p value of 0.29.

Group 1: Children with feeding aversion			
Patient	Diagnosis	Comorbidity	Feeding difficulties unexplained by oromotor difficulties
1	Russel silver syndrome	Poor growth	Not interested in voluntary feeding
2	Russell silver variant	-	Not interested in feeding
3	Russell Silver Syndrome	-	Aversive feeding behaviour
4	Congenital CMV infection	Bilateral hearing loss	Aversion to feeding
5	Expreterm	Chronic lung disease, atrial septal defect	Slow to feed

6	Expreterm	-	Not interested in feeding
7	Expreterm	-	Aversion to feeding
8	Expreterm	-	Selective eating
9	Expreterm, maternal parvovirus infection	-	Drinks excess of fluids
10	Expreterm	Epilepsy, visual impairment	Selective eating
11	Developmental delay	Epilepsy, swallowing difficulty	Aversion for fluids
12	Developmental delay	-	Reluctant to try solids
13	Developmental delay, post extra-dural haematoma evacuation	Constipation	Aversion to feeding
14	Repaired encephalocele	Developmental delay, VP shunt	Not like solids
15	Cerebral palsy	Epilepsy	Reduced oral intake
16	Cerebral palsy	Epilepsy	Aversion to feeding
17	Ataxic cerebral palsy	-	Aversion to feeding
18	Goldenhar syndrome	Oromotor dysfunction	Takes very little by mouth
19	Costello syndrome	Developmental delay	Aversion to feeding
Group 2: Children with oromotor difficulties			
Patient	Diagnosis	Co-morbidities	Oro-motor difficulties
1	Cerebral palsy	Epilepsy	Struggle to swallow solids
2	Cerebral palsy	None	Yes
3	Chromosome 18q deletion	Eczema	Yes
4	Developmental delay	Epilepsy	Aspiration with thin fluids
5	Cerebral palsy	Epilepsy	Yes
6	Pierre Robin sequence with repaired cleft palate	None	Yes
7	Klippel Feil syndrome	Hydrocephalus	Aspiration with liquids
8	Cerebral palsy	None	Yes
9	Smith Lemli Optiz syndrome	None	Yes
10	Cerebral palsy	None	Yes
11	Cerebral palsy	Epilepsy	Yes
12	Micrognathia	None	Yes
13	Developmental delay	Epilepsy	Yes
14	Ex-preterm	Hydrocephalus and epilepsy	Yes
15	Criduchat syndrome	None	Yes
16	Hypoxic ischemic encephalopathy	None	Yes
17	Cerebral palsy	None	Yes
18	Cerebral palsy	Movement disorder	Yes
19	Cerebral palsy	Epilepsy	Yes

**Table 1:** Children with feeding difficulties: diagnosis, co-morbidity and feeding behaviours.

### Phase two

In this patients showing feeding aversion were assessed using CEBQ prospectively. We recruited 16 patients with prematurity, global developmental delay, hypoxic ischaemic encephalopathy, Russel silver syndrome, Aicardi Goutieres syndrome, epilepsy and

hydrocephalus, epilepsy and white matter changes, Down’s syndrome, lissencephaly and epilepsy, cerebral palsy and Leighs disease. Table 2 summarizes the demographic features and feeding behavior difficulties. The mean age of the study population was six years, 4 females and 12 males.

Scores for individual patient was obtained across the 8 subdomains of satiety response, slowness of eating, emotional undereating, fussiness, food responsiveness, enjoyment of food, desire to drink and emotional overeating. First 4 subdomains are related to undereating tendency; whereas the rest are for overeating tendency. Mean score of our patient in each subdomain was calculated and compared with the population mean from Wardle et al., 2001[6] (Table 3). Our study patients significantly underscored compared to population mean in categories of satiety response (p: 0.02), food responsiveness (p: 0.017), enjoyment of food (p: 0.005), desire to drink (0.019) and emotional over eating (p: 001) (Table 3).

A mean of 6 items per child were not answered particularly in emotional overeating, emotional undereating and slow eating subdomains. Figures 1-4 shows individual scores of each subdomain for each patient, and are compared for the rewarding and aversive styles. Figure 5 shows the stars plot representing rewarding and aversive styles for each patient.

## Discussion

Our clinical experience at a tertiary hospital feeding clinic for children with neurodevelopmental conditions suggests that children who are labelled as having “feeding aversion” can show a spectrum of behaviours like some had sensory aversion to food and feeding whereas others showed lack of food seeking. In our study we have described the spectrum of these behaviours. Irrespective of the behavior phenotype management was with multidisciplinary approach with support of dietician, speech and language therapist and gastroenterologist. The burden of care is high amongst these patients as 47% patients needed gastrostomy for supporting calorie and fluid intake.

The intrinsic component to the feeding behaviour is not always recognized in these children characterized by undereating. There may be fruitless attempts at identifying a structural abnormality, ascription of the problem to normal structural variants (tongue-tie), or to poor parenting. There is little research to describe in detail the patterns of deficient eating seen in these children with neurodevelopmental disorders. This is in marked contrast to the well-studied overeating behavioural phenotype characteristic of Prader-Willi syndrome, which is often taken as a model of intrinsic appetite control and obesity in normal population [15]. In addition there is little controlled research to evaluate interventions, which can partly be due to the lack of consistency amongst different workers, from different professional disciplines, in use of terminology and outcome measures. Speech and language therapist observation is essential in all these children but it is not readily quantified or can be used in large scale studies and remains subjective.

Different assessment tools are developed like behaviour assessment procedure [9], semi structured interviews of parents and video tape of lunch times [16], Child Eating Behaviour Inventory (CEBI) [17], Dutch Eating Behaviour Questionnaire [18] and CEBQ [14]. There are more research based tests for classifying children further

although not used for clinical purposes such as 6-n-propylthiouracil (PROP) testing of supertasters, fungiform papillae counting [19,20]. As none of the tools capture the entire spectrum of feeding problem “multidimensional approach” to assessment taken by Archer and Szatman 1990 [21] in a child with “food aversion” need to be considered. This includes parental report, excluding gastro-oesophageal reflux, video fluoroscopy, and speech and language therapy assessment. We used CEBQ which captures feeding styles seen in our practice essentially a tool, which with other measures, would contribute for multidimensional assessment of this cohort of children with feeding aversion.

Patient	Age in years	Sex (M: Male, F: Female)	Diagnosis	Description of feeding behaviours
A	2	M	Not recorded	Food refusal of solids, intermittently, once a month. Will still be drinking milk during these time.
B	4	M	Down’s syndrome with feeding gastrostomy. Cleft lip and palate repaired.	Feeding aversion, gradually improving
C	4	M	Lissencephaly, epilepsy, visual impairment	Aversive behavior for food and feeding.
D	Not recorded	M	Cerebral palsy	Aversive behavior to food, gradually improving.
E	8	M	Expreterm, chronic lung disease	No signs of hunger, no oral intake
F	10	M	Leigh’s syndrome	Takes long time to feed and reduced fluid intake
G	8	M	Wolf-Hirschhorn syndrome	Feeding aversion may put a banana and enjoys chocolate mousse.
H	3	F	Expreterm	Aversion to solid foods, can manage some pureed foods.
I	2	F	Cardiac rhabdomyoma and global developmental delay	Fussy eater, likes to chew on textured objects like cardboard.
J	3	F	Hypoxic ischaemic encephalopathy	Only tolerates smooth textures like pureed foods.
K	4	M	Russel Silver syndrome	Aversion to feeds only has juices and soft textured foods.
L	2	M	Aicardi Goutieres syndrome	Refusal of oral feeds
M	8	M	Unconfirmed genetic syndrome	Feeding aversion, now resolving.
N	19	M	Cerebral palsy, epilepsy, ventriculo-peritoneal shunt and global developmental delay.	Refuses to eat, keeps food in mouth for long periods before swallowing.
O	6	M	Global developmental delay, tracheostomy for trachea-bronchomalacia.	Aversion to food and feeding.
P	3	F	Global developmental delay and visual impairment.	Refusal of all solid meals.

**Table 2:** Phase 2 study population, demographic features and behavior description.

	Mean of study group	Standard deviation	Population mean	Standard deviation	P value
SR	2.55	1.16	3.0	0.7	0.02
SE	2.61	1.25	3.0	0.8	0.07
EU	2.92	1.39	2.7	0.8	0.334
FU	2.66	0.8	3.0	0.8	0.107
FR	1.75	0.85	2.2	0.7	0.017
EF	2.87	1.16	3.5	0.8	0.005
DD	1.92	1.02	2.6	1.1	0.019
EO	0.78	0.74	1.9	1.9	0.001

**Table 3:** Results of CEBQ, mean of study in each subdomain compared with the population mean

;SR: Satiety Response; SE: Slowness of Eating; EU: Emotional Undereating ;FU: Fussiness; FR: Food Responsiveness; EF: Enjoyment of Food DD: Desire to Drink; EO: Emotional Overeating

CEBQ has 35 items and was easy to complete in the clinic setting and only mean of 6 items remained unanswered. Amongst the rewarding eating styles of emotional overeating, desire to drink, food responsiveness and enjoyment of food the scores of study cohort were significantly low. This reinforces our observation that for our study population feeding is not identified as a rewarding experience. CEBQ scores add objectivity to this observation but also identify the subdomains where the interventions can be targeted and the response can be monitored. It was not possible to cluster patients into separate groups based on scores across subdomains. Three patients B, D and M who had already shown clinical improvement did not reflect in their subdomain scores and similarly patient E with no sign of hunger had comparable scores to others. This could be attributed to the complexity of underlying medical diagnosis, previous interventions and the heterogeneity of motor, sensory, cognitive and behaviour co-morbidities which all impact on feeding. A cross sectional view of this study is the limiting factor that did not demonstrate correlation with the clinical phenotype.

Our study has been helpful in showing the spectrum of aversive feeding behaviours and CEBQ can have a potential role in multidimensional assessment of feeding aversion amongst children with neurodisability. Its role in longitudinal follow up and response to strategies is yet to be proven.

## Conclusion

This study identifies that feeding difficulties not related to oropharyngeal structural or motor impairment can be complex to assess and manage needing feeding gastrostomy. CEBQ has affirmed non-rewarding experience of feeding in this group and with its limitations can be helpful assessment of feeding aversion.

## Conflict of Interest

The authors declare no conflict of interest.

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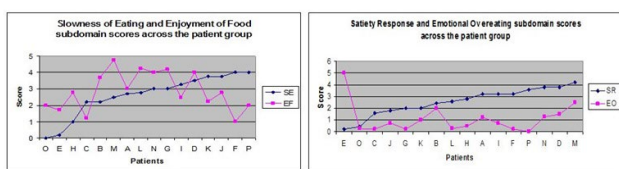


Figure-1

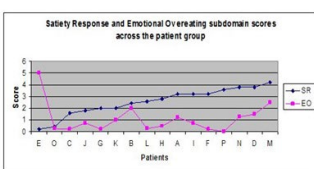


Figure-2

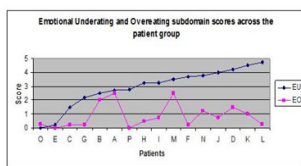


Figure-3

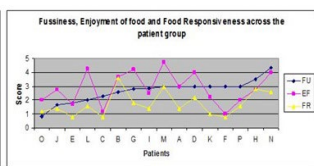
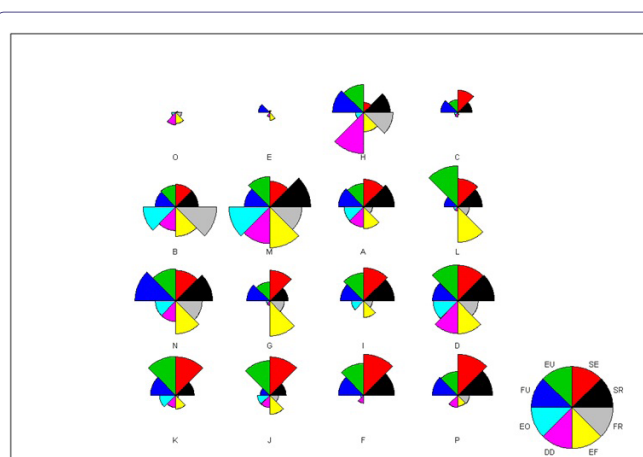


Figure-4

**Figures 1-4:** Scores of each subdomain are compared between the rewarding and aversive behaviours.



**Figure 5:** Stars plot of patient group, upper and lower half of the plot represents undereating and overeating scores respectively

SE: Slowness of Eating, SR: Satiety Response, FR: Food Responsiveness, EF: Enjoyment of Food, DD: Desire to Drink, EO: Emotional Over Eating, .FU: Fussiness, EU: Emotional Undereating



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