Feeding Dysfunction in Children with Single Ventricle Following Staged Palliation

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Objective To determine the prevalence of feeding dysfunction in children with single ventricle defects and identify associated risk factors.

Study design Patients aged 2-6 years with single ventricle physiology presenting for routine cardiology follow-up at the Children's Hospital of Wisconsin were prospectively identified. Parents of the patients completed 2 validated instruments for assessment of feeding dysfunction. Chart review was performed to retrospectively obtain demographic and diagnostic data.

Results Instruments were completed for 56 patients; median age was 39 months. Overall, 28 (50%) patients had some form of feeding dysfunction. Compared with a normal reference population, patients with single ventricle had statistically significant differences in dysfunctional food manipulation ($P < .001$), mealtime aggression ($P = .002$), choking/gagging/vomiting ($P < .001$), resistance to eating ($P < .001$), and parental aversion to mealtime ($P < .001$). Weight and height for age z-scores were significantly lower in subjects with feeding dysfunction (-0.84 vs -0.33; $P < .05$ and -1.46 vs -0.56; $P = .001$, respectively). Multivariable analysis identified current gastrostomy tube use ($P = .02$) and a single parent household ($P = .01$) as risk factors for feeding dysfunction.

Conclusion Feeding dysfunction is common in children with single ventricle defects, occurring in 50% of our cohort. Feeding dysfunction is associated with worse growth measures. Current gastrostomy tube use and a single parent household were identified as independent risk factors for feeding dysfunction. *(J Pediatr 2014;164:243-6)*.

Patients with single ventricle require staged palliation to create a passive flow circuit to the lungs. As survival has improved, it has become evident that the effects of single ventricle physiology and the palliations required are seen in many organ systems. There are known neurologic consequences that manifest as lower IQ and a higher prevalence of attention deficits.[1,2] There is also a high risk for restrictive lung disease in older patients.[3] Problems with nutrition and the gastrointestinal system, however, are nearly universal and occur throughout the palliative stages.[4-6] These include an increased risk of slow growth and feeding disorders.[4,7] Of all cardiac defects, patients with single ventricle lesions most frequently manifest feeding problems both at time of initial discharge and at 2-year follow-up.[4,8]

Studies have shown that up to 89% of patients with single ventricle hearts failed to meet Centers for Disease Control and Prevention standards for adequate growth and 50% were considered severely underweight at admission for Stage 2 palliation (S2P).[9,10] Compared with transposition of the great arteries, a heart defect that similarly requires neonatal heart surgery but results in a biventricular heart, those with hypoplastic left heart syndrome had a longer duration to achieve goal feeding levels after surgery and demonstrated slower weight gain at every interval measured up to 1 year of age despite both groups starting with comparable anthropometrics.[11]

Suboptimal growth continues in older patients with single ventricle despite surgical palliation and targeted interventions. The cause of these long-term growth problems remains unknown. The abnormal physiology has frequently been implicated. Caregiver nutrition concerns may negatively affect caregiver–child interactions around feeding and might exacerbate this feeding dysfunction. We sought to compare the prevalence of feeding dysfunction in patients with single ventricle aged 2-6 years with the known prevalence in the general population and identify risk factors for feeding dysfunction in these patients.

AYCE	About Your Child's Eating
G	Gastrostomy
HAZ	Height for age z-score
MBQ	Mealtime Behavior Questionnaire
S1P	Stage 1 palliation
S2P	Stage 2 palliation
WAZ	Weight for age z-score

Methods

After approval from the Children's Hospital of Wisconsin institutional review board, patients with single ventricle were recruited sequentially from the cardiology clinic, the catheterization lab, at the time of standard presurgical catheterization, or at the time of hospital admission for elective surgery at the Children's Hospital of Wisconsin over a 6-month period from April 2012 through September 2012. Patients were approached if they were between 2 and 6 years of age at study initiation and had completed S2P prior to 2 years of age. This age range was selected because it was the range used in the validation study of the Mealtime Behavior Questionnaire (MBQ). Patients were excluded if they had a congenital gastroesophageal malformation that required surgical repair or were enrolled in the Single Ventricle Reconstruction Trial. Additionally, as the instruments have only been validated in English, parents unable to complete them as such were excluded. A caregiver completed the MBQ, the About Your Child's Eating (AYCE), and a brief demographic questionnaire at the time of consent. Diagnostic and anthropometric data were collected retrospectively by chart review on all consented patients. Study data were collected and managed using REDCap electronic data capture tools at the Medical College of Wisconsin.[12]

The MBQ is a 33 question Likert scale instrument with 4 subcategories of feeding dysfunction. These subcategories are distraction/avoidance, food manipulation, mealtime aggression, and choking/gagging/vomiting. The AYCE is a 25 question Likert scale instrument with 3 subcategories. These subcategories are child resistance to eating, non-positive mealtime environment, and parental aversion to mealtime. Cut-off values for each sub-category have been determined by validation studies. Any positive subcategory was defined as being positive for feeding dysfunction.

Statistical Analyses

Descriptive data are presented as median with range or mean ± SD. We sought to determine the prevalence of feeding dysfunction in patients with single ventricle and compare the frequency of these subcategories with those in the general population established by validation studies for the assessment tools. This was performed using Fisher exact test. Weight for age z-score (WAZ) and height for age z-score (HAZ) were calculated using the World Health Organization Anthro and AnthroPlus applications. These continuous variables were compared between groups using Student t test. Risk factors for feeding dysfunction were evaluated using univariate analysis with Fisher exact test and multivariable analysis with logistic regression. Patient characteristics, including race/ethnicity, diagnosis, and birth WAZ and HAZ, as well as suspected risk factors for feeding problems, including history of stage 1 palliation (S1P), vocal cord injury, gastrostomy (G) tube status, as well as social factors, including household income and parental involvement were entered into the model with backward selection. OR and 95% CI are presented with 95% CI. Statistical analysis was performed using SAS OnDemand (SAS Institute, Cary, North Carolina).

Results

A total of 56 patients were enrolled and completed the instruments. Baseline characteristics of the cohort can be seen in Table I (available at www.jpeds.com). Median age was 39 (24-74) months. Median WAZ and HAZ at the time of enrollment were −0.6 (−3.7 to 1.5) and −1.1 (−3.8 to 1.4) respectively. Twenty-eight patients had feeding dysfunction for a prevalence of 50%. Thirteen patients were positive for 1 subtype of feeding dysfunction, 3 were positive for 2 subtypes, 8 were positive for 3 subtypes, and 4 were positive for >3 subtypes. Comparison between the group with feeding dysfunction and the group without can be seen in Table II with G tube use being associated with feeding dysfunction in this univariate analysis. Multivariable analysis showed current G tube use ($P = .02$) and a single parent home ($P = .01$) were associated with feeding dysfunction. OR and 95% CI for all factors used in multivariable analysis can be seen in Table III (available at www.jpeds.com). Significant factors not associated with feeding dysfunction were a diagnosis of hypoplastic left heart syndrome ($P = .53$), whether a S1P was required ($P = .43$), birth WAZ and HAZ ($P = .74$ and $P = .23$, respectively), and a history of vocal cord injury ($P = .60$).

Comparison by type of dysfunction was made with a normal population consisting of 355 children for the MBQ and 384 children for the AYCE (Figure 1). There was significantly more food manipulation (29% vs 5%, $P < .001$), mealtime aggression (16% vs 4%, $P = .002$), choking/gagging/vomiting (29% vs 8%, $P < .001$), child resistance to eating (13% vs 1%, $P < .001$), and parental aversion to mealtime (11% vs 1%, $P < .001$) in the single ventricle cohort.

WAZ and HAZ were compared between the patients with and without dysfunction at 3 time points, birth, at S2P, and at study enrollment as seen in Figure 2. At birth and S2P, the group with feeding dysfunction had a lower WAZ although this did not reach statistical significance in either ($P = .07$) but by enrollment the difference was significant ($P < .05$). This was also seen in HAZ, which were similar at birth ($P = $

Table II. Comparison of feeding dysfunction and non-feeding dysfunction groups by univariate analysis

	No feeding dysfunction	Feeding dysfunction	P value
Male, n (%)	17 (61)	17 (61)	1
Diagnosis of HLHS, n (%)	7 (25)	12 (43)	.26
Completed S3P, n (%)	11 (39)	6 (21)	.24
S1P required, n (%)	12 (43)	14 (50)	.79
G tube use, n (%)			.02
Never	18 (55)	15 (45)	
Former	8 (73)	3 (27)	
Current	2 (17)	10 (83)	
Vocal cord injury, n (%)	8 (29)	9 (32)	1
Lower household income, n (%)	15 (54)	16 (59)	.79
Single parent home, n (%)	4 (14)	11 (39)	.03

S3P, Stage 3 palliation.