

Caregiver Perspectives on School Participation Among Students With Craniofacial Microsomia

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Importance: Knowledge of unmet school participation needs for students with craniofacial microsomia (CFM) can inform decisions regarding intervention support.

Objective: To compare students with and without CFM on school participation (i.e., frequency, involvement, desire for participation to change) and caregivers' perceptions of environmental support for participation in occupations.

Design: Cross-sectional design using secondary analyses of a subset of data.

Setting: Multisite cohort study.

Participants: Caregivers of students with CFM ($n = 120$) and of students without CFM ($n = 315$), stratified by history of education- and health-related service use.

Outcomes and Measures: School participation and environmental support, obtained with the Participation and Environment Measure–Children and Youth.

Results: Significant group differences were found in frequency of school participation (effect size [ES] = -0.38 , 95% confidence interval [$-0.64, -0.12$], $p = .005$), level of involvement (ES = -0.14 , $p = .029$), and desired change ($p = .001$), with students with CFM exhibiting greater participation restriction than students without CFM and no history of service use. No statistically significant group differences were found in environmental support for participation in the school setting. Item-level findings showed statistically significant higher desire for participation to change in three of five school occupations (odds ratio = 1.77 – 2.39 , $p = .003$ – $.045$) for students with CFM compared with students without CFM and no history of service use.

Conclusions and Relevance: The results suggest that students with CFM experience restriction in participation at school.

What This Article Adds: Students with CFM may benefit from targeted school-based interventions to optimize their inclusion.

Children's participation in school occupations is a priority outcome of occupational therapy and a key indicator of children's inclusion in an educational context (American Occupational Therapy Association [AOTA], 2016). To ensure equal access to educational programming, professionals working in school settings need to identify (1) children with inclusion needs (e.g., children who have difficulty participating in school occupations) and (2) correlates of exclusion (i.e., pertinent factors that hinder a child's school participation; Mâsse et al., 2012).

Both the child's attendance and level of involvement in school occupations should be assessed when evaluating their current school participation (Imms et al., 2017). Previous studies assessing attendance and involvement (Adair et al., 2018) have detected disparities in school participation and perceived environmental support for school participation between children with and without a range of disabilities (Coster et al., 2013). Prior studies of disparities in school participation have excluded students with craniofacial microsomia (CFM).

People with CFM present with asymmetric underdevelopment of craniofacial structures that can lead to vision, hearing, and speech impairments, as well as performance difficulties related to eating and feeding (Strömland et al., 2007) and sleeping (Caron et al., 2015). CFM is estimated to occur in 1 in 3,500 to 5,600 live births (Poswillo, 1988) and

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can result in surgeries to restore craniofacial form and function, such as for the ear, eye, mandible, facial nerve, and orbit (Birgfeld & Heike, 2012). Students with CFM can receive rehabilitation services, including occupational and speech therapies (Luquetti et al., 2018; Speltz et al., 2017), and can experience reduced quality of life relative to unaffected peers, as indicated by caregiver-reported difficulties in physical, social, and school functioning (Khetani et al., 2013) that may hinder their school participation. These functional difficulties include lower verbal processing skills, including vocabulary, reading, and writing (Speltz et al., 2017), and behavioral–social adjustment (Wallace et al., 2018), as well as greater social concerns (Johns et al., 2018; Khetani et al., 2013; Luquetti et al., 2018) and stigma because of their appearance (Hamilton et al., 2018; Luquetti et al., 2018). However, the full extent to which these functional difficulties hinder school participation and caregivers' perceptions of support in the school environment remains unknown. Knowledge about unmet participation needs can inform decisions about whether school-based occupational therapy services need to be designed for students with CFM (AOTA, 2016).

The aims of this study were to compare students with and without CFM on (1) school participation and (2) caregivers' perceptions of environmental support for school participation. We hypothesized that students with CFM would experience greater participation difficulty in school occupations (i.e., less frequent participation, lower level of involvement, higher desire for participation to change) than students without CFM. We also expected that caregivers of students with CFM would perceive less environmental support for their child's school participation than would caregivers of students without CFM.

Method

In this study, we used secondary analyses of a subset of data that were collected in the third phase of a longitudinal cohort study of children diagnosed with CFM and children without craniofacial anomalies. Children were initially enrolled as infants between 1996 and 2002 as part of the first phase of this cohort study, which focused on demographics and risk factors for CFM (Werler et al., 2004). The second and third study phases focused on their neurodevelopmental and psychosocial status and enrolled children when they were, on average, between ages 7 and 13 yr (Collett et al., 2011; Duffton et al., 2011; Speltz et al., 2017; Wallace et al., 2018). We obtained ethics approval from the institutional review boards of Boston University and Seattle Children's Hospital for data collection and from the University of Illinois at Chicago for this study.

Participants

We used data from caregivers of children with CFM who were initially recruited from craniofacial specialty clinics and caregivers of children without CFM (the control participants) who were recruited through pediatricians of children with CFM or other pediatric practices (Werler et al., 2004).

Students With Craniofacial Microsomia

At the time of initial recruitment, children with CFM were included if they were younger than age 36 mo and had been diagnosed with CFM by a craniofacial physician per established criteria for hemifacial microsomia, facial asymmetry, unilateral microtia, oculo-auriculo-vertebral syndrome, or Goldenhar syndrome. Children were excluded if they had been diagnosed with chromosomal anomalies, Mendelian-inherited disorders, or in utero isotretinoin exposure or if they were adopted. A total of 279 children with CFM and their families enrolled in the first study phase (Werler et al., 2004); 198 enrolled in the second study phase (Collett et al., 2011; Duffton et al., 2011). For the third study phase, 18 families could not be contacted, 30 declined to participate, and 8 were not approached or were ineligible; thus, 142 children with CFM and their families were included in this study (Wallace et al., 2018).

Students Without Craniofacial Microsomia

Children without CFM were included if they had no known birth defect, were not adopted, and were within 2 mo of the age of the children with CFM at the time of recruitment. A total of 884 children and families participated in the first study phase (Werler et al., 2004); 568 children and families participated in the second phase (Collett et al., 2011; Dufton et al., 2011). For the third study phase, the families of 2 control children per child with CFM were contacted. Five children were not contacted for nonmatching reasons (e.g., ineligibility, child death) or case diagnosis, 169 families could not be approached or reached, and 79 declined to participate; thus, 315 children without CFM and their families were included in this study (Wallace et al., 2018).

Measures

Child and Family Characteristics

Caregivers reported on demographics (e.g., marital status, annual income, formal education), updated student medical history information, and the child's history of education- and health-related service use (e.g., occupational therapy services).

School Participation and Environmental Support

The Participation and Environment Measure—Children and Youth (PEM–CY; Coster et al., 2010) is a caregiver-report measure of a student's participation in home, school, and community occupations, as well as of perceived environmental support for participation in each setting. Data from the school section of the PEM–CY were used in this study.

The PEM–CY school section consists of five types of school occupations that occur inside and outside the classroom and comprise a school day (e.g., getting together with peers outside of class). For each type of school occupation, caregivers rated (1) their child's participation frequency (ranging from 0 = *never* to 7 = *daily*), (2) their child's level of involvement (ranging from 1 = *not very involved* to 5 = *very involved*), and (3) whether they desired a change in their child's participation (yes or no). Then caregivers rated 17 features and resources in the school environment (e.g., physical layout, school policies) according to their perceived impact on the student's school participation (ranging from 1 = *usually makes it harder/usually no* to 3 = *no impact/usually helps/usually yes*).

The PEM–CY school scales have acceptable to good internal consistency and test–retest reliability for large-sample research (Coster et al., 2011, 2013) and as confirmed with the data for this study ($\alpha = .56-.78$ for school participation items and $\alpha = .80$ for environmental items). The PEM–CY also distinguishes between students with and without disabilities (effect size [ES] = 0.51–1.44; Coster et al., 2011, 2013).

Four PEM–CY scores were calculated. Mean participation frequency and mean level of involvement summary scores were computed. For desire for change in school participation, a count score was calculated by summing all “yes, change desired” responses across school participation items. For environmental support in school participation, a summary score was derived by calculating the sum of ratings across environmental items, dividing by the maximum possible score, and multiplying by 100.

Procedure

Caregivers were administered demographic and service history items and the PEM–CY while their children were being administered a 4- to 5-hr battery of standardized neurocognitive assessments by trained psychometrists in a community setting (e.g., library; Speltz et al., 2017; Wallace et al., 2018). Caregivers each received a \$35 gift card.

Data Analysis

Data were analyzed using SAS software (Version 9.4; SAS Institute, Cary, NC). Participants with missing values on all PEM–CY school items were excluded ($n = 50$). Data were first examined for the total sample (via descriptive statistics, histograms, regression diagnostics) to ensure they met the underlying assumptions of the selected statistical tests.

As a result of known associations between service use and children's participation (Khetani et al., 2018), students without CFM were further grouped on the basis of their history of service use (i.e., whether they currently received or had received occupational therapy, physical therapy, speech-language therapy, visual therapy, hearing services, mental health services, special education services, or other services). Eleven children without CFM were excluded from further analyses as a result of missing data on service use. Insufficient sample size hindered the grouping of children with CFM on the basis of their history of service use. However, sensitivity analyses for the main comparisons of students with and without CFM who had a history of service use ($n = 94$) revealed no significant differences in the results obtained, but in the item-level analyses caregivers of students with CFM reported a significantly higher percentage of change desired in four out of five activities when compared with caregivers of students without a history of service use. The characteristics of the three groups (students with CFM; students without CFM and a history of service use; students without CFM and no history of service use) were then summarized using descriptive statistics.

Four sets of main analyses were performed to examine group differences in school participation frequency, level of involvement, desire for change (Aim 1), and perceived environmental support for school participation (Aim 2). To test for differences in school participation frequency and environmental support for school participation, multiple linear regression analyses were used, adjusting for covariates identified by previous research on participation, in this study cohort, or both: student gender, age, and race/ethnicity; maternal age at child's birth; and caregiver education, marital status, and annual income.

For categorical covariates, the reference groups were those with the largest sample size in the subgroup of students with CFM (married, White non-Hispanic, annual income \geq \$65,000, at least a high school or general education diploma). Cohen's d was used to estimate the magnitude of group differences (with 0.2 = small, 0.5 = medium, and 0.8 = large; Cohen, 1988). To examine group differences in level of involvement, Wilcoxon–Mann–Whitney tests were used because of skewed data distribution and an insufficient regression model fit. Effect sizes were calculated using r , based on the z score (.3 = medium, .5 = large; Rosenthal, 1991). Group differences in the number of occupations for which change is desired were examined using Poisson regression.

Because desire for change denotes participation needs from the caregiver's perspective (i.e., greater desire for change indicates greater caregiver dissatisfaction), it is of clinical relevance. Thus, item-level comparisons for desire for change were pursued using logistic regression analysis and odds ratio (OR) for effect sizes. Otherwise, descriptive statistics were applied to item-level data to report on average participation frequency, level of involvement, and caregiver perceptions of environmental support for students with and without CFM.

Because this is the first study of caregiver perspectives on school participation of students with and without CFM, we did not adjust for multiple comparisons to avoid missing potentially important associations that merit further study (i.e., Type II errors). At the same time, rather than viewing p values as dichotomous tests of significance, we focused on the magnitude and precision (i.e., 95% confidence intervals [CIs]) of observed associations (Greenland et al., 2016).

Results

Participants were 120 caregivers of students with CFM, 140 caregivers of students without CFM and a history of service use, and 136 caregivers of students without CFM and no history of service use. Students were between ages 11 and 17 yr ($M_s = 13.15$ – 13.52 , $SD_s = 1.25$ – 1.52) and White, non-Hispanic (range = 82.35%–82.86%; Table 1). Most students with CFM and students without CFM who had a history of service use accessed rehabilitation services (i.e., occupational therapy, physical therapy, speech therapy; 68.07% and 50.74%, respectively). Most caregivers were married (range = 75.94%–85.59%) and earned more than the annual median U.S. income of \$65,000

Table 1. Characteristics of Students With and Without CFM and Their Caregivers

Characteristic	Students With CFM (<i>n</i> = 120), <i>n</i> (%)	Students Without CFM, <i>n</i> (%)	
		With History of Service Use (<i>n</i> = 140)	Without History of Service Use (<i>n</i> = 136)
Male	69 (57.50)	74 (52.86)	61 (44.85)
Student age, yr, <i>M</i> (SD)	13.39 (1.25)	13.52 (1.52)	13.15 (1.33)
Grade in school, <i>M</i> (SD)	7.25 (1.38)	7.50 (1.74)	7.16 (1.44)
History of ≥1 type of service used ^a	94 (78.99)	140 (100.00)	0 (0.00)
Occupational therapy	31 (26.05)	14 (10.14)	0 (0.00)
Physical therapy	43 (36.13)	41 (29.50)	0 (0.00)
Speech-language therapy	69 (57.98)	44 (32.12)	0 (0.00)
Vision therapy	9 (7.56)	24 (17.14)	0 (0.00)
Hearing services	39 (32.77)	3 (2.16)	0 (0.00)
Mental health services	17 (14.29)	37 (26.43)	0 (0.00)
Special education services	37 (31.09)	44 (32.12)	0 (0.00)
Other	28 (23.73)	65 (46.76)	0 (0.00)
Student race/ethnicity			
White, non-Hispanic	99 (82.50)	116 (82.86)	112 (82.35)
White, Hispanic	15 (12.50)	9 (6.43)	12 (8.82)
Other	6 (5.00)	15 (10.71)	12 (8.82)
Marital status ^b			
Married	101 (85.59)	101 (75.94)	114 (84.44)
Single	5 (4.24)	10 (7.52)	4 (2.96)
Divorced, separated, widowed	12 (10.17)	22 (16.54)	17 (12.59)
Annual income ^c			
<\$65,000	43 (37.39)	38 (29.23)	36 (27.27)
≥\$65,000	72 (62.61)	92 (70.77)	96 (72.73)
Caregiver education ^d			
At least high school/GED	45 (38.46)	28 (20.90)	42 (31.11)
Associate's degree	17 (14.53)	25 (18.66)	19 (14.07)
Bachelor's degree	41 (35.04)	44 (32.84)	50 (37.04)
Graduate degree	14 (11.97)	37 (27.61)	24 (17.78)
Caregiver age at child's birth, <i>M</i> (SD)	29.93 (6.00)	30.49 (5.47)	29.89 (4.89)

Note. CFM = craniofacial microsomia; GED = general education diploma.

^aMissing data; *n* = 199 for students with CFM. ^bMissing data; *n* = 118 for students with CFM; *n* = 133 for students without CFM/with history of service use; and *n* = 135 for students without CFM and without history of service use. ^cMissing data; *n* = 115 for students with CFM; *n* = 130 for students without CFM/with history of service use; and *n* = 132 for students without CFM and without history of service use. ^dMissing data; *n* = 117 for students with CFM; *n* = 134 for students without CFM/with history of service use; and *n* = 135 for students without CFM and without history of service use.

(range = 62.61%–72.73%; Fontenot et al., 2018). Participants who did not enroll in this study were more likely to be Spanish speaking and non-White or Hispanic (Speltz et al., 2017).

School Participation Frequency

Students with CFM participated less frequently in school occupations than students without CFM and without a history of service use (ES = $-.38$, 95% CI [$-.64$, $-.12$], $p = .005$) even after adjusting for selected covariates (Table 2). No statistically significant differences were found between students with CFM and students without CFM with a history of service use (ES = $-.19$, 95% CI [$-.46$, $.07$], $p = .16$). Descriptive item-level group differences in school participation frequency were largest for “special roles at school” (e.g., lunch room supervisor, student mentor; *M* difference = 1.00), whereby students with CFM participated less frequently than students without CFM who had no history of service use (Supplemental Figure 1, available online at <http://otjournal.net>; navigate to this article, and click on “Supplemental”).

Table 2. School Participation and Environmental Support Among Students With and Without CFM

Variable	Range	Students With CFM vs. Students Without CFM With History of Service Use		Students With CFM vs. Students Without CFM Without History of Service Use	
		B or exp (b) [95% CI]	ES [95% CI]	B or exp (b) [95% CI]	ES [95% CI]
Participation frequency ^a	0–7	–0.21 [–0.51, 0.08]	–.19 [–.46, .07]	–0.42* [–0.70, –0.13]	–.38 [–.64, –.12]
Desire for change in school participation ^{a,b}	0–5	1.01 [0.83, 1.21]		1.41* [1.15, 1.72]	
Environmental support in school participation ^a	0–100	0.22 [–2.35, 2.79]	.02 [–.24, .29]	–2.05 [–4.56, 0.47]	–.21 [–.47, .05]

	Students With CFM, Mdn (Q1, Q3)	Students Without CFM, Mdn (Q1, Q3)	
		With History of Service Use	Without history of service use
Level of involvement in school occupations ^c	1–5	4.50 [4.00, 5.00]	4.67* [4.33, 5.00]

Note. CFM = craniofacial microsomia; CI = confidence interval; ES = effect size; exp (b) = exponentiation of beta; Mdn = median; Q = quartiles.
^aAdjusted for student gender, age, race/ethnicity; caregiver age, education, marital status; annual income. ^bExp (b) reported. ^cSignificance testing was done between students with CFM and each of the control groups.
 * $p < .05$.

Student Level of Involvement in School Occupations

Students with CFM (median [Mdn] = 4.50) were significantly less involved in school occupations than their peers without CFM who had no history of service use (Mdn = 4.67, $W = 13,695.50$, $z = -2.19$, $ES = -.14$, $p = .029$). Item-level descriptors did not reveal further disparities in students’ level of involvement in specific types of school occupations (Supplemental Figure 2). There were no statistically significant differences when we compared students with CFM and students without CFM with a history of service use (Mdn = 4.50, $W = 14,793.50$, $z = -.24$; $ES = -.02$; $p = .82$).

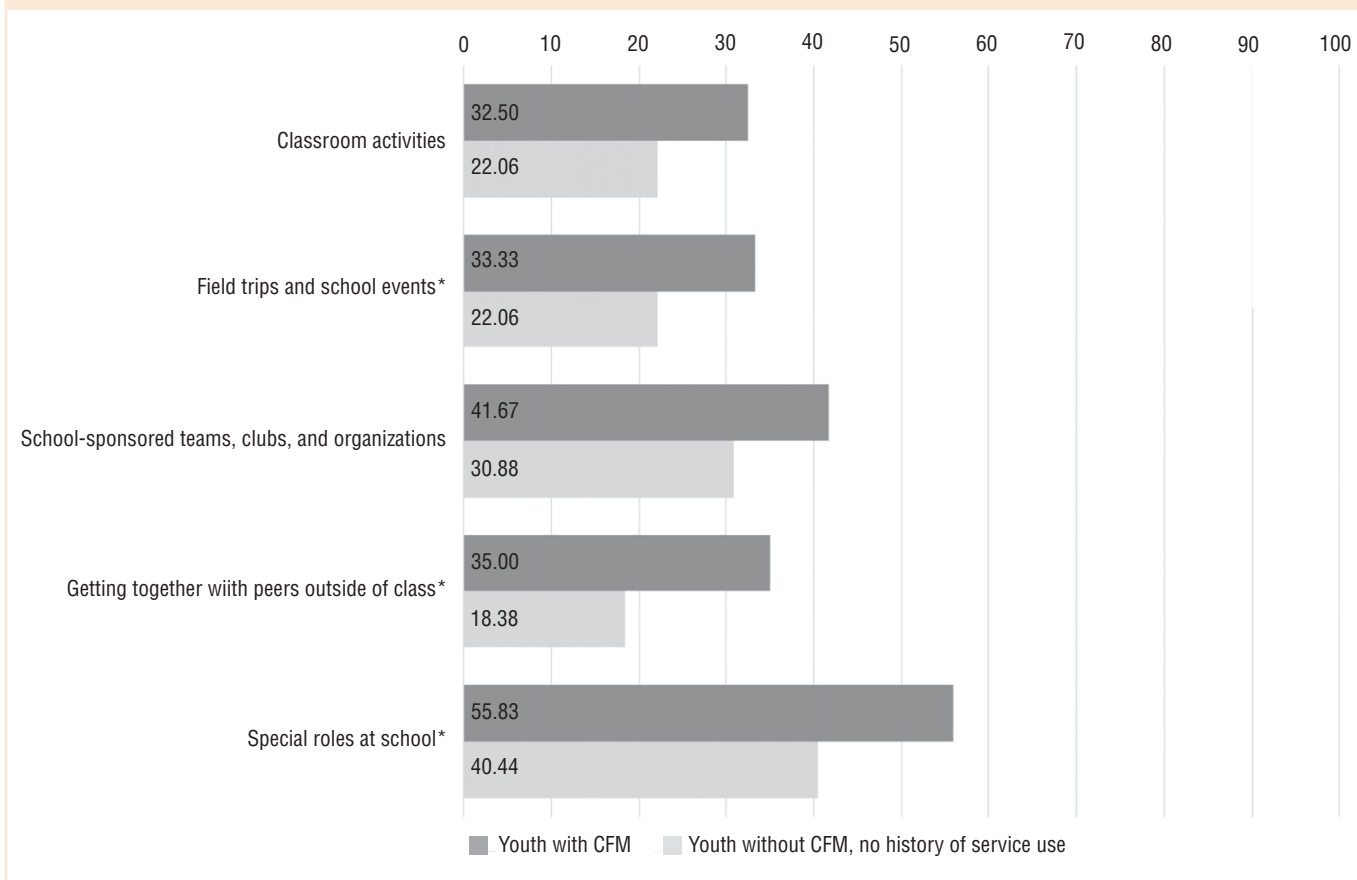
Desire for Change in School Participation

Compared with not having CFM and having no history of service use, having CFM was significantly associated with a 41% increase in the mean number of school occupations for which change was desired (exponentiation of beta [exp [b]] = 1.41, 95% CI [1.15, 1.72], $p = .001$). When compared with not having CFM and having a history of service use, no significant association was found between CFM and the mean number of school occupations for which change was desired (exp [b] = 1.01, 95% CI [0.83, 1.21], $p = .963$). Item-level comparisons revealed that having CFM was significantly associated with about twice the odds of caregiver dissatisfaction for three of five school occupations: (1) field trips and school events (e.g., going to a museum, the school fair; OR = 1.77, 95% CI [1.01, 3.08], $p = .045$); (2) getting together with peers outside of class (e.g., hanging out during lunch, at recess, or other breaks during the school day; OR = 2.39, 95% CI [1.35, 4.24], $p = .003$); and (3) special roles at school (OR = 1.86, 95% CI [1.13, 3.06], $p = .014$). In addition, participation in classroom learning (OR = 1.70, 95% CI [0.98, 2.97], $p = .062$) and school-sponsored teams, clubs, and organizations trended toward statistical significance (OR = 1.60, 95% CI [0.96, 2.67], $p = .074$; see Figure 1).

Perceived Environmental Support for School Participation

Caregivers’ perceptions of school environmental support were similar for caregivers of students with CFM and those of the two groups of students without CFM, when adjusting for select covariates ($ES = -.21$, 95% CI [–.47, .05], $p = 0.11$, and $ES = .02$, 95% CI [–.24, .29], $p = 0.87$, respectively). For students with CFM and students without CFM and with no history of service use, the item-level descriptive findings showed that the largest differences in perceived environmental support pertained to the sensory qualities of the school environment (e.g., noise, crowds, lighting), social (e.g., communication, interaction with others) and cognitive demands (e.g., concentration, attention, problem solving) of school occupations, and peer relations (mean differences ≥ 5.99), for which environmental support was lower for students with CFM (Supplemental Figure 3).

Figure 1. Percentage of caregivers reporting student participation dissatisfaction across school occupations.



Note. CFM = craniofacial microsomia.

* $p < .05$.

Discussion

Participation in school occupations is fundamental for youth skill development and inclusion (AOTA, 2016). A better understanding of school participation among students with facial differences such as CFM is critical to understanding who may benefit from school services including occupational therapy. To our knowledge, this is the first study to illustrate the scope of disparities in school participation among students with and without CFM.

School Participation Need

The results support our hypothesis that students with CFM participate in school occupations less frequently than peers without CFM. Differences were larger in magnitude and statistically significant when children with CFM were compared with children without CFM who had no history of service use. Caregivers of students with CFM also reported lower involvement in school occupations and greater dissatisfaction with their child’s school participation (i.e., higher number of occupations for which participation change was desired) than did caregivers of students without CFM and no history of service use. Dissatisfaction with participation is primarily evident for three of five occupations. These occupations provide students with important opportunities to build relationships (e.g., field trips and school events, getting together with peers outside of class) and leadership and management skills (e.g., special roles at school, such as lunch room supervisor).

Previous research on community participation among this cohort (Kaelin et al., 2020) and students with mixed disabilities has also revealed differences in caregiver dissatisfaction with similar types of occupations, in particular those with a more pronounced social or leadership component (Coster et al., 2013). Together, these results indicate that students with CFM present with a need for inclusion that, if unaddressed, may hinder their opportunities to build social and leadership skills for a successful postsecondary transition that is focused on higher education, employment, and community integration (Gorter et al., 2011). Students with CFM may therefore benefit from school-based services, including occupational therapy expertise, to address specific school participation challenges (Leigers et al., 2016).

This study did not detect significant differences in school participation between students with and without CFM who had a history of service use. These results may indicate that students with diverse diagnoses share similar needs (Coster et al., 2013). Whether these similarities in school participation difficulty are related to the focus of service use (i.e., the extent to which services are responsive to areas of participation need) remains unclear. Little is known about how to best address transition challenges and postsecondary aspects to support students' school participation. Future research could benefit from capturing data on service use obtained from service record abstraction (e.g., number of participation-focused goals addressing school transition, quarterly estimate of number of benchmarks met for goals) or open-ended items on caregiver strategy use via the PEM-CY to understand the scope of school-based service provision with respect to participation-focused goals in order to identify gaps in service approaches that need to be addressed.

School Environmental Support for Participation

Contrary to our hypothesis, there was no significant difference in caregivers' perceptions of school environmental support between students with and without CFM. However, students with CFM were reported, on average, to have less supportive peer relationships and sensory qualities (e.g., noise, crowds, lighting) in the school environment. Similarly, previous research on school participation showed statistically significant lower perceptions of support for a range of environmental factors, including social and sensory aspects of the school environment, when students with a mixed range of disabilities were compared with students without disabilities (Coster et al., 2013). The current findings may suggest that a select group of environmental aspects are more salient in their impact on school participation for students with CFM (Kaelin et al., 2020; Riklin et al., 2019).

Riklin et al. (2019) highlighted the need to address the social and sociopolitical environment for children and youth with facial differences, which caregivers of youth with physical disabilities have also emphasized in their strategies for promoting their children's participation (Killeen et al., 2018). Killeen et al. (2018) showed a high prevalence of participation-focused strategy use by caregivers that targeted their child's school environment, including advocacy and collaboration with school staff. Because the PEM-CY yields data on caregiver strategy use, additional analyses of these data will help to identify common ways in which specific barriers to school participation are being addressed among children with CFM (Johns et al., 2018). These results could translate to receipt of services by a broader group of children with facial differences.

Limitations


Our results should be interpreted in light of several limitations. We did not control for confounders in one of three main analyses because of skewed data. Because this study used secondary data analysis, we were limited to available data on service receipt and focus (e.g., service type, scope of services) and could not account for service quality. Finally, the results are specific to the caregiver perspective, one of multiple perspectives that are relevant to shared decision making when designing school-based services for transition-age youth. To get a full picture of unmet school participation need among students with CFM, access to the student perspective is needed (Raghavendra, 2013). Youth self-report measures are emerging and could be used to confirm study findings.

Implications for Occupational Therapy Practice

This study contributes new knowledge about students with CFM, who were shown to have unmet school participation needs that may limit their opportunities for socialization and leadership. The study has the following implications for school-based occupational therapy practice:

- Occupational therapists should pay attention to the participation of students with CFM, with and without an Individualized Education Program, in school occupations that are social or involve opportunities for leadership roles.
- Social and sensory (e.g., noise, crowds, lighting) environmental aspects may be important to consider when supporting school participation of students with CFM.

Conclusion

This study extends knowledge of school participation needs and associated factors as experienced by students with CFM. The results indicate that students with CFM have unmet school participation needs that warrant further study to drive the design of tailored interventions. 

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