PREFERENCES FOR DISCLOSING ADVERSE CHILDHOOD EXPERIENCES FOR CHILDREN AND ADULTS WITH CYSTIC FIBROSIS

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October 12, 2020

Abstract

Introduction: The 2017-2018 National Survey of Children's Health estimates that 30 million (42%) US children have experienced at least one adverse childhood experience (ACE), including abuse, neglect, and household dysfunction. ACEs negatively impact long-term health, and there has been no study of ACEs in cystic fibrosis (CF). We assessed willingness to disclose ACEs experienced by children with CF by surveying their parents and adults with CF. Methods: We anonymously surveyed parents of children with CF and adults with CF at the Northwestern University/ Lurie Children's CF Center to determine their willingness to disclose ACEs. Results: The survey was completed by 46/157 (29%) parents and 36/105 (34%) adults with CF. Few parents (22%) and adults (17%) were willing to discuss most or all specific ACEs, more were willing to disclose the number of ACEs experienced in a category (57% parents, 47% adults), and the majority were willing to participate in anonymous research about ACEs (76% parents, 67% adults). Most parents (63%) and adults (50%) would prefer to have ACEs screened separately from their CF appointment, and most parents (63%) and adults (56%) wanted to learn more about ACEs from a member of their care team. Conclusions: Participants preferred to disclose the number of categorical ACEs rather than specific ACEs and most were open to participating in anonymous ACEs research. More research is needed before widespread adoption of ACE screening in CF.

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Financial Support or Conflicts of Interest:

None

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Abbreviated title: Preferences for Screening ACEs in CF

Keywords: ACE, Toxic Stress, Abuse, Neglect

Abbreviations:

ACE = Adverse Childhood Experience

CF = Cystic Fibrosis

CYW = Center for Youth Wellness

NSCH = National Survey of Children's Health

Introduction:

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Methods:

We anonymously surveyed parents of children with CF and adults with CF at the Northwestern University/ Lurie Children's CF Center to determine their willingness to disclose ACEs.

Results:

The survey was completed by 46/157 (29%) parents and 36/105 (34%) adults with CF. Few parents (22%) and adults (17%) were willing to discuss most or all specific ACEs, more were willing to disclose the number of ACEs experienced in a category (57% parents, 47% adults), and the majority were willing to participate in anonymous research about ACEs (76% parents, 67% adults). Most parents (63%) and adults (50%) would prefer to have ACEs screened separately from their CF appointment, and most parents (63%) and adults (56%) wanted to learn more about ACEs from a member of their care team.

Conclusions:

Participants preferred to disclose the number of categorical ACEs rather than specific ACEs and most were open to participating in anonymous ACEs research. More research is needed before widespread adoption of ACE screening in CF.

Introduction:

Adverse childhood experiences (ACEs) are a collection of maltreatments that occur before the age of 18, encompassing the domains of abuse, neglect, and household dysfunction. ACEs were first studied in 1998,

when the landmark Kaiser-CDC study demonstrated a dose-dependent negative impact of ACEs on adult health conditions including ischemic heart disease, cancer, substance abuse, and depression.[1] Since then, those findings have been confirmed[2] and have been linked to earlier onset of chronic disease.[3]

The biology linking ACEs to risk of disease is postulated to be related to increased levels of stress hormones, referred to as toxic stress. Childhood toxic stress is "severe, prolonged, or repetitive adversity with a lack of the necessary nurturance or support of a caregiver to prevent an abnormal stress response."[4] Accumulation of toxic stress can lead to a persistent inflammatory response, epigenetic modification, and telomere shortening.[5–7]The cumulative nature of ACEs was estimated in 2019 to have an annual cost to North America of more than $748 billion US dollars in disability adjusted life years.[8] In addition, researchers have demonstrated the manifestations of ACEs in children and adolescents, including learning and behavior issues, substance use and abuse, obesity, depression, anger, and suicidality.[9,10]

The 2017-2018 National Survey of Children's Health (NSCH) estimates that 30 million (42%) US children have experienced at least one ACE, and 62.3% of children with more complex health needs have at least 1 ACE.[11] Furthermore, the NSCH shows that ACEs are increased in certain populations, including children from low socioeconomic backgrounds and minority race and ethnicity.[11] There is a paucity of research into ACEs in children with chronic illnesses, though elevated ACE scores are associated with increased prevalence of asthma, attention deficit-hyperactivity disorder, and autism.[12-14]

To date, there have been no studies evaluating ACEs in people with cystic fibrosis (CF). However, the CF Foundation and the European CF Society currently recommend screening for and, when present, treating depression and anxiety.[15] There are significant health outcome disparities in CF: affected people who are racial and ethnic minorities and/or of lower socioeconomic status have an increased risk of mortality from CF before the age of 18,[16] and Hispanic CF patients have a higher mortality, even after adjusting for clinical severity.[17] ACEs are more frequent in minority and low socioeconomic populations, and thus ACEs may contribute to disparities in health in CF. The comprehensive multidisciplinary care at CF centers could facilitate screening and appropriate intervention, as demonstrated by implementation of the mental health screening guidelines.

Due to the sensitive nature of ACE screening, the purpose of this study was to educate our CF population about ACEs and survey patient preferences for future ACEs screening.

Methods:

Study Design and Population:

An anonymous survey was distributed by email to two populations at our CF Center: (1) Parents/legal guardians of children with CF and (2) Adults with CF. This study was deemed exempt by the Lurie Children's Institutional Review Board because it was an anonymous survey of adults.

Anonymous Research Electronic Data Capture (REDCap) Survey:

A REDCap survey was emailed to all participants from distribution lists maintained by the adult and pediatric CF programs with address list management to preserve anonymity of recipients. The survey included an initial paragraph explaining ACEs, demographic information, exposure to the age-specific screening tools, questions about preferences for screening, and where to find more information or get assistance should the information cause concern for a participant. A downloadable informational document that offered more detail about health consequences of ACEs was attached. The survey, information sheets, and introductory email were offered in both Spanish and English and distributed in the language of preference recorded at the CF Center, a standard of practice for all information sent to parents and patients.

Demographic information collected included age range, race/ethnicity, education, employment status, and length of time treated at the CF Center. After viewing age-specific ACE screening tools, a 5-point Likert scale was used for participants to rate willingness to discuss specific ACEs, disclose the number of applicable ACEs in a category, and to participate in anonymous ACE research related to health outcomes. Participants

were asked to select a preference for when they would like to be screened for future ACEs and how they would prefer to communicate and receive more information about ACEs. For every question, participants were able to select "prefer not to answer" or enter free text. The last item of the survey invited participants to share any thoughts, comments, or suggestions. Both parent and adult participants were instructed to contact the CF Center if they had concerns for their child or themselves.

Pediatric CF Parent/Guardian Survey:

The survey was sent to parents/guardians of patients seen in the pediatric CF program with instruction that the survey was to be filled out by a guardian, even if their child with CF was older than 18 years. In the case of multiple children with CF, each parent/guardian was asked to fill out the survey with their oldest child with CF in mind. The ACE questionnaire displayed was from the Center for Youth Wellness (CYW) (Supplemental Table 2).[18] Preferences about future participation in ACE screening and research were asked after both CYW Section 1 and 2.

Adult CF Patient Survey:

The adult patients were shown the original ACE screening tool only (Supplemental Table 1).[1]

Statistical Analysis:

Pearson's Chi-squared tests were used for comparisons of categorical variables and Mann-Whitney U tests for Likert scale data. Significance was set at $p<0.05$.

Results:

Pediatric CF Parent/Guardian Survey:

Among 170 patient families in the pediatric program in the summer of 2019, 157 (92%) had parent/legal guardian email addresses available. The survey was completed by 46/157 (29.3%) families. All respondents were parents of CF patients. Three surveys were excluded from analysis because of incomplete fields for the ACE screen preferences section. The largest proportion of the parent cohort were between the ages 35-44 (23, 50.0%), non-Hispanic white (34, 73.9%), had a college degree or greater (39, 84.8%), were employed (34, 73.9%), had an oldest child with CF under the age of 12 (35, 76.1%), and had been at the Pediatric CF Program for more than 5 years (25, 54.3%) (Table 1). There was no difference in the distribution of race/ethnicity for Lurie Children's Pediatric CF Program and the survey respondents (non-Hispanic white 70.9%, p=0.075).

As displayed in Tables 2A and 2B, few parents were willing to discuss all or most specific ACEs for sections 1 and 2 (10, 21.7% and 18, 39.2% respectively). More parents were willing to disclose the number of events that would apply to their child in a specific category – section 1 (26, 56.5%) and section 2 (37, 80.4%). The majority of parents were willing to participate in anonymous health outcomes research related to their child's ACEs score – section 1 (35, 76.1%) and section 2 (37, 80.4%). There was no difference in parental preference to participate in specific ACE screens or anonymous ACE research between Section 1 and Section 2 (p=0.38, p=0.73, respectively). Parents were more willing to participate in a categorical ACE screen of Section 2 versus Section 1 (p=0.024).

From Section 1, a stepwise increase was seen in the number willing to participate in ACE screens using specific, categorical, and anonymous research (specific vs categorical, p=0.0027; categorical vs research, p=0.018; specific vs research, $p<0.00001$). In section 2, more parents expressed willingness to participate in future categorical ACE screens compared to specific ($p<0.00001$). However, there was no difference between categorical versus research screens ($p<0.48$). Most parents (28, 60.9%) wanted to screen for future ACEs through an emailed survey, and most (29, 63.0%) wanted to learn more about ACEs from a member of their CF care team.

Adult CF Patient Survey:

Among adults with CF, 105 of 110 (95%) had a recorded email address. The survey was completed by 36/105 (34.3%) adults with CF in the summer of 2019. No surveys were excluded from analysis. The largest share of the cohort were white (24, 66.7%), had a college degree or greater (23, 63.9%), were employed (17, 47.2%), and had been at the Northwestern Medicine Adult CF Program for more than 5 years (19, 52.8%) (Table 1). At the time of the survey, there was no difference in the distribution of race/ethnicity for the Northwestern Medicine Adult CF Program and the survey respondents (non-Hispanic white, 84.3%, p=0.20).

As displayed in Table 2C, few adults were willing to discuss all or most specific ACEs (6, 16.7%), more were willing to disclose the applicable categorical number of events (17, 47.2%), and most were willing to participate in anonymous health outcomes research (24, 66.7%). Adults were more willing to participate in future categorical ACE screens compared to specific (p=0.0040). There was no difference between the distributions of willingness to disclose ACEs in a category and to participate in ACEs research (p=0.14). Most adults (18, 50.0%) wanted to screen for future ACEs through an emailed survey, and most (20, 55.6%) wanted to learn more about ACEs from a member of their CF care team.

Comparisons:

There were no significant differences in responses for parents of children with CF or adults with CF based on length of time at the CF center, age, race/ethnicity, education, or employment. There were no significant differences between parental and adult willingness to participate in specific, categorical, or in anonymous research about ACEs (Section 1 compared to adults: specific, p=0.17; categorical, p=0.82; research, p=0.94. Section 2 compared to adults: specific, p=0.084; categorical, p=0.087; research, p=1).

Discussion:

Although enthusiasm and support for screening ACEs flourishes in the literature,[19] screening is not routinely performed in primary care or subspecialty practices for children or adults. Many valid concerns for the surveillance of ACEs have been raised, such as the narrow scope of ACE questionnaires, whether to ask about ACEs categorically or specifically, and how to best address a positive ACE finding.[20,21] The answers to these difficult questions require a partnership between ACE experts, physicians, and community members who reflect those who will be screened and offered interventions. Our data from parents of children with CF and adults with CF suggests that adoption of widespread screening for ACEs experienced by people with CF may be premature, that disclosure of categorical rather than individual ACEs exposure may be preferred, and that most are willing to disclose ACEs for research purposes.

Anonymous disclosure for research was most acceptable to our participants: over $\frac{3}{4}$ of parents and 2/3 of adults were willing to participate in ACEs research. There were no differences in willingness to disclose ACEs based on any demographic factor, including the length of time that a child or an adult had been treated at the CF center. While screening for ACEs may benefit people with CF, our results suggest that anonymous research on the prevalence of ACEs exposure in the CF population is an important first step. Results would better inform people with CF, their families, and their health care team about prevalence in conjunction with education about the harms of ACEs and related toxic stress.

Although the American Academy of Pediatrics has acknowledged the significant impact of ACEs and advocated for screening during childhood, there is not consensus concerning the methods – where, when, and which questionnaire.[19] There is an ongoing debate about whether or not ACEs should be screened for categorically, thereby providing the respondent with anonymity to the precise nature of the ACE, or specifically, to better understand necessary treatment and further risk. Our participants preferred to disclose ACEs through a categorical screen, which does not disclose details about adverse experiences.[22] While some research suggests that specific ACE combinations increase susceptibility to specific poor health outcomes, with treatment implications;[23] there is also evidence that identifying ACEs from categorical screens allows beneficial interventions through fostering resiliency and addressing key social needs.[24,25] A positive screen for an ACE, either categorically or specifically, can be addressed through methods including mitigating social needs through social work and referrals, bolstering resilience, addressing mental health, and advocating for mindfulness techniques.[26 –29]

Our findings demonstrate the feasibility of screening for ACEs, but more research is needed to further understand whether these results are representative of a large population of families whose children have CF or of adults with CF. For example, the finding that both parents of children with CF and adults with CF preferred to have ACEs screened separately from their appointment and through email may represent a bias given that the current survey was completed by email only. However, the finding may represent lack of knowledge and comfort with routine ACE screening or the wish to reduce difficult discussions during a routine visit; our data are not sufficient to fully understand this preference. Although some studies have demonstrated that parents want to have conversations about their child's ACEs,[30] there may be a more accessible and preferred method of early screening that could evolve over time to in-person discussions. This notion is supported by the parent and adult desire to learn more about ACEs from a member of their care team. Further studies are needed to expand and further explore patient preferences for ACE screening, assess the capacity for CF centers to handle a positive screen, and understand the prevalence of ACEs in the CF community.

There are several limitations of this study. The survey was a small sample size from a single center. The respondents are majority white, educated, and employed, a group that is less likely to have a high number of ACEs. Additionally, the survey was only offered by email to maintain anonymity. Further research with a larger sample size is needed. In spite of these limitations, the devastating nature of toxic stress is too extreme to ignore.[31] Ongoing research suggests the benefit of screening for ACEs and intervening to reduce their negative effects.[32] The results of this survey indicate that parents of children with CF and adults with CF are willing to disclose ACEs for research, prefer categorical ACE screening like that created by CYW to disclosure of specific events, and prefer not to disclose ACES during routine CF care. Furthermore, direct discussion of ACEs during routine clinical care may not be acceptable to parents of children with CF and adults with CF. Additional, separate discussions are preferred. Given the recent rapid growth in telehealth services, this may be feasible for incorporating ACEs screening in research and clinical care settings. In conclusion, anonymous research on ACEs and their effects on health and health care in people with CF is likely feasible but should be structured in a manner that is acceptable to participants.

References:

1. Felitti VJ, Anda RF, Nordenberg D, Williamson DF, Spitz AM, Edwards V, Koss MP, Marks JS. Relationship of Childhood Abuse and Household Dysfunction to Many of the Leading Causes of Death in Adults. Am J Prev Med. 1998;14(4):245–258.

2. Hughes K, Bellis MA, Hardcastle KA, Sethi D, Butchart A, Mikton C, Jones L, Dunne MP. The effect of multiple adverse childhood experiences on health: a systematic review and meta-analysis. Lancet Public Health. 2017;2(8):e356–e366.

3. Sonu S, Post S, Feinglass J. Adverse childhood experiences and the onset of chronic disease in young adulthood. Prev Med. 2019;123(March):163–170.

4. Franke H. Toxic Stress: Effects, Prevention and Treatment. Children. 2014;1(3):390–402.

5. Shonkoff JP, Garner AS, Committee on Psychosocial Aspects of Child and Family Health; Committee on Early Childhood, Adoption, and Dependent Care; Section on Developmental and Behavioral Pediatrics. The lifelong effects of early childhood adversity and toxic stress. Pediatrics. 2012;129(1).

6. Chatterjee N. Stress Runs Deep and Long: Identification of Molecular Biomarkers of Childhood Stress in Adults. BioEssays. 2018;40(9):1800126.

7. Ridout KK, Khan M, Ridout SJ. Adverse childhood experiences run deep: toxic early life stress, telomeres, and mitochondrial dna copy number, the biological markers of cumulative stress. BioEssays. 2018;40(9):1–10. doi:10.1002/bies.201800077

8. Bellis MA, Hughes K, Ford K, Ramos Rodriguez G, Sethi D, Passmore J. Life course health consequences and associated annual costs of adverse childhood experiences across Europe and North America: a systematic

review and meta-analysis. The Lancet Public Health. 2019;4(10):e517–e528.

9. Hussey JM, Chang JJ, Kotch JB. Child maltreatment in the United States: prevalence, risk factors, and adolescent health consequences. Pediatrics. 2006;118(3):933–42.

10. Burke NJ, Hellman JL, Scott BG, Weems CF, Carrion VG. The impact of adverse childhood experiences on an urban pediatric population. Child Abuse Negl. 2011;35(6):408–413.

11. Child and Adolescent Health Measurement Initiative. 2017-2018 National Survey of Children's Health (NSCH) data query. Data Resource Center for Child and Adolescent Health supported by the U.S. Department of Health and Human Services, Health Resources and Services Administration (HRSA), Maternal and Child Health Bureau (MCHB). Retrieved [07/11/2020] from [www.childhealthdata.org].

12. Wing R, Gjelsvik A, Nocera M, McQuaid EL. Association between adverse childhood experiences in the home and pediatric asthma. Ann Allergy Asthma Immunol. 2015;114(5):379–384.

13. Brown NM, Brown SN, Briggs RD, German M, Belamarich PF, Oyeku SO. Associations between adverse childhood experiences and ADHD diagnosis and severity. Acad Pediatr. 2017;17(4):349–355.

14. Hoover DW, Kaufman J. Adverse childhood experiences in children with autism spectrum disorder. Curr Opin Psychiatry. 2018;31(2):128–132.

15. Quittner AL, Abbott J, Georgiopoulos AM, Goldbeck L, Smith B, Hempstead SE, Marshall B, Sabadosa KA, Elborn S, Boyle M, et al. International Committee on Mental Health in Cystic Fibrosis: Cystic Fibrosis Foundation and European Cystic Fibrosis Society consensus statements for screening and treating depression and anxiety. Thorax. 2016;71(1):26–34.

16. McColley SA, Schechter MS, Morgan WJ, Pasta DJ, Craib ML, Konstan MW. Risk factors for mortality before age 18 years in cystic fibrosis. Pediatric Pulmonol. 2017;52(7):909–915.

17. Buu MMC, Sanders LM, Mayo JA, Milla CE, Wise PH. Assessing differences in mortality rates and risk factors between hispanic and non-hispanic patients with cystic fibrosis in California. Chest. 2016;149(2):380–389.

18. Bucci M, Gutierrez L, Koita K, Purewal S, Marques SS, Harris NB. Center for Youth Wellness ACE-Questionnaire User Guide. 2015:2012–2015.

19. Siegel BS, Dobbins MI, Earls MF, Garner AS, Pascoe J, Wood DL, High PC, Donoghue E, Fussell JJ, Gleason MM, et al. Early childhood adversity, toxic stress, and the role of the pediatrician: Translating developmental science into lifelong health. Pediatrics. 2012;129(1).

20. Finkelhor D. Screening for adverse childhood experiences (ACEs): Cautions and suggestions. Child Abuse Negl. 2018;85:174–179.

21. Gillespie RJ. Screening for adverse childhood experiences in pediatric primary care: Pitfalls and possibilities. Pediatric Annals. 2019;48(7):e257–e261.

22. Gillespie RJ, Folger AT. Feasibility of assessing parental aces in pediatric primary care: implications for practice-based implementation. J Child Adolesc Trauma. 2017;10(3):249–256.

23. Lanier P, Maguire-Jack K, Lombardi B, Frey J, Rose RA. Adverse childhood experiences and child health outcomes: comparing cumulative risk and latent class approaches. Matern Child Health J. 2018;22(3):288–297.

24. Heard-Garris N, Davis MM, Szilagyi M, Kan K. Childhood adversity and parent perceptions of child resilience. BMC Pediatrics. 2018;18(1):1–10.

25. Gottlieb LM, Hessler D, Long D, Laves E, Burns AR, Amaya A, Sweeney P, Schudel C, Adler NE. Effects of social needs screening and in-person service navigation on child health: A randomized clinical trial. JAMA Pediatrics. 2016;170(11):1–7.

26. Traub F, Boynton-Jarrett R. Modifiable resilience factors to childhood adversity for clinical pediatric practice. Pediatrics. 2017;139(5).

27. Ortiz R, Sibinga E. The role of mindfulness in reducing the adverse effects of childhood stress and trauma. children. 2017;4(3):16.

28. Bellis MA, Hughes K, Ford K, Hardcastle KA, Sharp CA, Wood S, Homolova L, Davies A. Adverse childhood experiences and sources of childhood resilience: A retrospective study of their combined relationships with child health and educational attendance. BMC Public Health. 2018;18(1):1–12.

29. Marie-Mitchell A, Kostolansky R. A systematic review of trials to improve child outcomes associated with adverse childhood experiences. Am J Prev Med. 2019;56(5):756–764.

30. Conn AM, Szilagyi MA, Jee SH, Manly JT, Briggs R, Szilagyi PG. Parental perspectives of screening for adverse childhood experiences in pediatric primary care. Fam Syst Health. 2018;36(1):62–72.

31. Dube SR. Continuing conversations about adverse childhood experiences (ACEs) screening: A public health perspective. Child Abuse Negl. 2018;85(March):180–184.

32. Barnes AJ, Anthony BJ, Karatekin C, Lingras KA, Mercado R, Thompson LA. Identifying adverse childhood experiences in pediatrics to prevent chronic health conditions. Pediatr Res. 2020;87(2):362–370.

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