

CER Question Grid Template

Project Number: 3414288 *Improving Oral Health for Vulnerable Populations: Individuals with Intellectual and Developmental Disabilities*

Preamble. Dental caries, an intractable chronic disease for adults with intellectual and developmental disabilities (IDD), adversely affects their systemic health and quality of life. Individuals with IDD face special challenges maintaining good oral health because of commonly associated physical, cognitive and sensory impairments, and chronic medical and behavioral conditions. Adults with IDD who are unable to perform preventive at-home oral care themselves rely upon family members or paid caregivers for assistance. Dental caries persists in members of this group even when they utilize dental services on a regular basis. There are no studies documenting effective prevention of dental caries in adults with IDD.

Early childhood caries (ECC) was also considered an intractable disease until recently. Newly emerging data, however, from clinical trials show that chronic disease management interventions are effective in reducing the onset and progression of caries in pre-school children (Edelstein and Ng, 2015; Ng, Ramos-Gomez, Lieberman, Lee, Scoville, Hannon and Maramaldi, 2014; Garcia, Borrelli, Dhar, Douglass, Gomez, Hieftje, Horowitz, Li, Ng, Twetman and Tinanoff, 2015; Ng and Fida, 2016). Chronic disease management (CDM) interventions for caries utilize comprehensive patient-specific management plans based on Individual Risk Assessments (IRA) to address the underlying disease processes that result in caries (Edelstein and Ng, 2015). In the case of ECC, important risk factors – brushing regularly with fluoride toothpaste, flossing, consuming sugar-sweetened food and beverages, visiting the dentist regularly - are heavily influenced by the caregiver and patient, and study participants work with study personnel to set and meet self-management goals. Chronic disease management plans typically include clinical interventions, such as professional fluoride applications, as well as efforts to modify patient and caregiver behavior through motivational interviewing and other supportive techniques.

During Tier II, our Partnership discussed the complex web of factors that influence dental disease in the general population, reviewed scientific evidence about the effectiveness of oral health interventions, and considered the implications of this information for adults with IDD. **The Partnership believes that the chronic disease management (CDM) interventions found effective in preventing and managing dental caries in pre-school children hold promise for adults with IDD and we have formulated our comparative effectiveness research (CER) questions accordingly.** Certain parallels between the populations bode well for applying lessons learned from CDM of childhood caries to adults with IDD. Those at risk for caries in both groups depend heavily on family members and paid caregivers for assistance. Federal and state policies support organized systems of supports for both young children and adults with IDD and their families. Also, regulations governing services for adults with IDD mandate annual Individual Service Plans (ISP), not unlike Individual Risk Assessments, to guide service delivery.

There is also a strong evidence-base about the effectiveness of interventions to prevent childhood caries to guide selection of strategies for inclusion in an eventual proposal for a chronic disease management intervention to prevent caries in adults with IDD. Both professionally applied and self-applied topical fluoride has been found to be effective at preventing childhood caries (Fluoride Recommendations Work Group, 2001; Marinho, Higgins, Logan and Sheiham, 2003 (Issue 1); Gibson, Jurasic, Wehler, Orner, Nunn, Nunez, O'Toole, Arola, Bestgen and Jones, 2014; Jurasic, Gibson, Wehler, Nunn, Orner, Nunez, O'Toole and Jones, 2014; Marinho, Higgins, Logan, Sheiham, 2003 (Issue 4); Marinho, Higgins, Sheiham, Logan 2004 (Issue 1); Weyant et al for the American Dental Association Council on Scientific Affairs Expert Panel on Topical Fluoride Caries Preventive Agents, 2013; Walsh, Worthington, Glenny, Appelbe, Marinho and Shi, 2010. Supervised use of self-applied fluoride increases benefits (Marinho, Higgins, Logan, Sheiham, 2003). While the effectiveness of fluoride has been much less studied in adults, results of a recent clinical trial show that both professionally and self-applied fluoride reduced caries incidence in adults with complex medical conditions (Gibson et al., 2014; Jurasic, Gibson et al 2013; Griffin, Regnier, Griffin and Huntley, 2007).

In addition, there is some evidence, albeit weak, about the effectiveness of behavioral interventions (school-based and family-based), although studies suggest that motivational interviewing is the most effective behavioral strategy to date in terms of childhood caries prevention (Albino and Tiwari, 2016).

We acknowledge the need to further explore the nature of potential risk factors for dental caries – specific to adults with IDD - and the feasibility of implementing strategies to address them in the context of their day-to-day lives. Efforts to better understand the oral health role of caregivers and the nature of potential supports to help them are central to these discussions. Although there is some evidence about the effectiveness of caregiver training programs on the oral health of individuals with IDD, it is both limited and weak (Binkley, Johnson, Abadi, Thompson, Shamblen, Young and Zaksek, 2014; Faulks and Hennequin, 2000; Fickert and Ross, 2012; Glassman & Miller, 2006). We also need to better understand the nature of behavioral challenges – resulting from fear and anxiety about dental visits as well as neuro-behavioral conditions - to regular oral home care and receipt of dental services, and strategies to overcome them.

It is possible that our efforts during Tier III will lead us to conclude that it's overly ambitious or unrealistic to implement a chronic disease management intervention to prevent caries in adults with IDD at this time. If so, we will turn our attention to developing a research proposal that investigates just one strategy of the several strategies, which we propose to study in Tier III - for example, application of different forms of topical fluoride - and measure their relative effectiveness in preventing caries in adults with IDD. CER Question 2 reflects this possibility. Of the caries prevention strategies we've considered to date, we are most interested, at present, in fluoride applications and efforts to train caregivers in oral home care practices.

A list of references is provided after the PICOT Grid.

**CER Question
1:**

Which of the following variations of a chronic disease management intervention for dental caries in adults with intellectual and developmental disabilities (IDD) is most effective at reducing the onset and recurrence of dental caries:

- **Intervention Group A.** Following completion of an Individual Risk Assessment, implementation of a patient-specific management plan including tailored preventive dental services and supports for both the adult with IDD AND a caregiver.
- **Intervention Group B.** Following completion of an Individual Risk Assessment, implementation of a patient-specific management plan including tailored preventive dental services and supports for the adult with IDD only.
- **Control Group.** Following completion of an Individual Risk Assessment, subjects follow their usual and customary oral health practices, including preventive dental visits and oral home care practices.

Population

Adults with IDD have significant limitations in both intellectual functioning and adaptive behavior, with onset of limitations before 18 years of age. Intellectual disability (ID) is a type of developmental disability (DD), a broader category representing various chronic conditions associated with physical impairments, mental impairment or both. To the best of our knowledge, no information exists from nationally representative samples about the prevalence of dental caries among individuals with IDD. Estimates from convenience- and population-based samples, however, suggest that people with IDD have a higher prevalence of caries and poorer oral hygiene than the general population. An epidemiologic study of the oral health of 4,732 adults with IDD 20 years or older in Massachusetts – based on clinical examinations recorded in electronic dental records – reported the prevalence of caries experience was 87.8% and of untreated caries was 32.2% (Morgan, et al., 2012). According to the U.S. Centers for Disease Control and Prevention, the prevalence of untreated caries for the general population 20 years or older is 22.7%. **Thus, study subjects demonstrated a higher disease burden, despite visiting the dentist 4.6 times, on average, for preventive care during the study period.**

Of the estimated 4.9 million Americans with IDDs, the majority (72 percent) live with family members who help them with oral health. The rest live in supervised settings (13 percent) or in their own homes independent of families (16 percent). People with IDDs who do not live with family members generally rely on direct support staff – employed by organizations that support people with IDDs – to assist them with oral health care. A convenience sample of 808 caregivers of adults with DD in Massachusetts who completed personal interview surveys reported that 85% of dentate adults received assistance with tooth cleaning; and caregivers reported low adherence to brushing (79 percent) and flossing (22 percent) recommendations (Minihan et al., 2014).

	<p>Adults with IDD represent a heterogenous population and we have yet to determine the specific sub-populations that would be most appropriate for inclusion in a research study. Considerations involve variables with potential impact on caries, including dentition, age, level of functioning, ability to cooperate with dental care, presence/absence of specific medical and/or behavioral conditions, use of certain medications and dietary supplements, and type of residence and associated involvement of caregivers. Further considerations include having a regular source of dental care, including perhaps receiving dental care services from the Tufts Dental Facilities Serving Persons with Special Needs, a statewide network of dental clinics in Massachusetts designed specifically to provide comprehensive oral health care to this population, and enrollment in MassHealth/Medicaid. In situations where an adult with IDD has been assigned a legal guardian, a final and important consideration involves informed consent. The logistics of obtaining informed consent from legal guardians is daunting and presents challenges to enrolling research subjects.</p>
Intervention	<p>Because there are no studies documenting effective prevention of dental caries in adults with IDD, we have decided to apply 'lessons learned' from successful efforts to prevent early childhood caries to adults with IDD. We are considering a chronic disease management intervention which will utilize patient-specific management plans, based on Individual Caries Risk Assessments, to address the underlying disease process that results in caries. Caries Risk Assessments will include clinical oral examinations and office-based preventive treatments, such as topical fluoride applications, as well as assessments of modifiable risk factors. Clinical exams and preventive treatments will be provided by dentists; other still-to-be-identified staff will assess modifiable behavioral risk factors. Our focus is on caries risk factors that are modifiable, for example, oral home care practices or dietary sugar consumption, vs. risk factors that are not. The latter includes congenital malformations of the mouth or teeth. Other potentially modifiable caries risk factors include caregivers' oral home care knowledge and skills, and challenging behaviors. The components that will guide patient-specific management plans and goal-setting activities will need to be determined following extensive research and consultation. Components discussed to date include: strategies to train adults with IDD in oral home care skills; supports to help adults with IDD improve their oral home care practices and avoid sugar-sweetened foods and beverages, perhaps using electronic devices; training and supports to help caregivers improve oral home care practices and dietary choices; and behavioral management supports specific to dental visits. How and where individualized supports to adults with IDD would best be provided also needs to be determined. Curricula to help adults with IDD improve their oral home care practices and dietary choices exist although there is no information about their efficacy. Curricula to train paid caregivers in oral hygiene skills also exist; evaluations have focused on implementation and not on dental outcomes.</p>

<p><u>Comparator</u></p>	<p>We are currently considering the following comparisons. Upon completion of an Individual Risk Assessment, including clinical exams and preventive treatments, study subjects will (ideally) be randomly assigned to one of two (or three) study groups:</p> <p>Intervention group 1: Implementation of a patient-specific management plan including tailored services and supports for <u>both the adult with IDD AND a caregiver</u>. Supports for the adult with IDD could include group teaching sessions, 1:1 coaching; online video instruction and use of social media. Caregiver training efforts could include enrollment in an online tutorial, face-to-face or online instruction; or 1:1 coaching.</p> <p>Intervention group 2: Implementation of a patient-specific management plan including tailored services and supports <u>for the adult with IDD only</u>.</p> <p>Control Group – Upon completion of an Individual Risk Assessment, subjects follow their usual and customary oral health practices, including subsequent dental visits and oral home care practices.</p>
<p><u>Outcomes</u></p>	<p>We are currently considering the following outcome measures:</p> <ul style="list-style-type: none"> • Reducing the proportion of adults with IDD with untreated dental caries; • Reducing the proportion of adults with new caries during the study period; • Reducing the proportion of adults with IDD complaining of dental pain during the study period; • Reducing the proportion of adults with IDD referred to the operating room for surgical treatment during the study period.
<p><u>Timing</u></p>	<p>We have yet to determine the optimal length of the intervention. The Early Childhood Collaborative CDM intervention, to which we've referred, reported positive outcomes after a 30-month study period. A follow-up intervention conducted in additional sites over 18-months also reported positive outcomes.</p>
<p><u>Setting</u></p>	<p>We will not be able to determine the setting until we've refined our thinking about the intervention but it is likely to include the Seven Hills Foundation, a primary organizational partner. The Seven Hills approach – to support adults with IDD within the communities where they live, work, and obtain health services – sustains a web of relationships and organizational supports that would enhance recruitment, support the continued enrollment of research subjects, and enhance data collection and quality. It is also likely that the Tufts Dental Facilities (TDF) Serving Persons with Special Needs, a statewide</p>

**PCORI Priority
Area**

network of dental clinics in Massachusetts designed specifically to provide comprehensive oral health care to this population, including a clinic located in a Seven Hills facility, would be involved. Patients in the TDF system receive comprehensive general dental services that include annual dental examinations, dental prophylaxis, restorative dentistry, periodontal care, endodontic care, prosthetic dentistry and tooth extraction. Dental specialty support and a full spectrum of patient care management modalities – including desensitization, sedation and general anesthesia – are available to support the care provided by dental clinicians. The TDF system uses an electronic dental record (axiUm) which would enhance data collection and analysis

Addressing Disparities

**CER Question
2:**

Which topical fluoride application protocol is most effective at reducing dental caries in adults with intellectual and developmental (IDD) disabilities?

Population

Adults with IDD have significant limitations in both intellectual functioning and adaptive behavior, with onset of limitations before age 18 years. Intellectual disability (ID) is a type of developmental disability (DD), a broader category representing various chronic conditions associated with physical impairments, mental impairment or both. To the best of our knowledge, no information exists from nationally representative samples about the prevalence of dental caries among individuals with IDD. Estimates from convenience- and population-based samples, however, suggest that people with IDD have a higher prevalence of caries and poorer oral hygiene than the general population. An epidemiological study of the oral health of 4,732 adults with IDD 20 years or older in Massachusetts – based on clinical examinations recorded in electronic dental records – reported the prevalence of caries experience was 87.8% and of untreated caries was 32.2% (Morgan, et al., 2012). According to the U.S. Centers for Disease Control and Prevention, the prevalence of untreated caries for the general population 20 years or older is 22.7%. Thus, study subjects demonstrated a higher disease burden, despite visiting the dentist 4.6 times, on average, for preventive care during the study period.

Of the estimated 4.9 million Americans with IDDs, the majority (72 percent) live with family members who help them with oral health. The rest live in supervised settings (13 percent) or in their own homes— independent of families (16 percent). People with IDDs who do not live with family members generally rely on direct support staff – employed by organizations that support people with IDDs – to assist them with oral health care. A convenience sample of 808 caregivers of adults with DD in Massachusetts who completed personal interview surveys reported that 85% of dentate adults received assistance with tooth cleaning; caregivers reported low adherence to brushing (79 percent) and flossing (22 percent) recommendations among the adults with disabilities in their care (Minihan et al., 2014).

Adults with IDD represent a heterogeneous population and we have yet to determine the specific sub-populations that would be most appropriate for inclusion in a research study. Considerations involve variables with potential impact on caries, including dentition, age, level of functioning, ability to cooperate with dental care, presence/absence of specific medical and/or behavioral conditions, use of certain medications and dietary supplements, and type of residence and associated involvement of caregivers. Further considerations include having a regular source of dental care, including perhaps receiving dental care services from the Tufts Dental Facilities Serving Persons with Special Needs, a statewide network of dental clinics

	<p>in Massachusetts designed specifically to provide comprehensive oral health care to this population, and enrollment in MassHealth/Medicaid. In situations where an adult with IDD has been assigned a legal guardian, a final and important consideration involves informed consent. The logistics of obtaining informed consent from legal guardians is daunting and presents challenges to enrolling research subjects.</p>
<u>Intervention</u>	<p>We are considering an intervention to prevent dental caries in adults with IDD by applying fluoride (professionally or self/caregiver applied). The following data about the effectiveness of fluoride has informed our thinking. No studies have been conducted to determine if fluoride is effective in preventing dental caries in individuals with IDD. There is evidence, however, that fluorides prevent dental caries in the general population (Fluoride Recommendations Work Group, 2001). Toothpaste, rinses, gels and varnishes help prevent caries in adults of all ages (Griffin, Regnier, et al, 2007; Weyant, Tracy, et al. for the American Dental Association Council on Scientific Affairs Expert Panel on Topical Fluoride Caries Preventive Agents). Fluoride (self- and professionally applied) was found to be effective at reducing caries in medically compromised veterans (Gibson, Jurasic, et al. 2014; Jurasic, Gibson, et al., 2014).</p>
<u>Comparator</u>	<p>We are currently considering at least two different potential comparisons: a) comparing different forms of application (professionally applied compared to self/caregiver applied); OR b) comparing different frequencies of professional applications (e.g., four times/year as currently allowed by MassHealth/Medicaid for adults with IDD compared to other frequencies). The latter could refer to the frequency of professional applications allowed by private dental benefit providers in Massachusetts or to that recommended by dentists following the CAMBRA (caries management by risk assessment) protocol. CAMBRA is an evidence-based approach to preventing or treating dental caries at the earliest stages.</p>
<u>Outcomes</u>	<p>We are currently considering the following outcome measures:</p> <ul style="list-style-type: none"> • Reducing the proportion of adults with IDD with untreated dental caries; • Reducing the proportion of adults with new caries during the study period; • Reducing the proportion of adults with IDD complaining of dental pain during the study period; • Reducing the proportion of adults with IDD referred to the operating room for surgical treatment during the study period.
<u>Timing</u>	<p>We have yet to determine the optimal length of the intervention, but review of previous studies suggests that an 18-month to 24-month study period would be appropriate.</p>

Setting

We will not be able to determine the setting until we've refined our thinking about the intervention but it is likely to include the Seven Hills Foundation, a primary organizational partner. The Seven Hills approach – to support adults with IDD within the communities where they live, work, and obtain health services – sustains a web of relationships and organizational supports that would enhance recruitment, support the continued enrollment of research subjects, and enhance data collection and quality. It is also likely that the Tufts Dental Facilities (TDF) Serving Persons with Special Needs, a statewide network of dental clinics in Massachusetts designed specifically to provide comprehensive oral health care to this population, including a clinic located in a Seven Hills facility, would be involved. Patients in the TDF system receive comprehensive general dental services that include annual dental examinations, dental prophylaxis, restorative dentistry, periodontal care, endodontic care, prosthetic dentistry and tooth extraction. Dental specialty support and a full spectrum of patient care management modalities – including desensitization, sedation, and general anesthesia – are available to support the care provided by dental clinicians. The TDF system uses an electronic dental record (axiUm) which would enhance data collection and analysis.

PCORI Priority Area

Addressing Disparities

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