

# A Structured Approach Using the Systematic Review Data Repository (SRDR): Building the Evidence for Oral Health Interventions in the Population With Intellectual and Developmental Disability

Evaluation Review

2017, Vol. 41(2) 111-129

© The Author(s) 2016

Reprints and permission:

[sagepub.com/journalsPermissions.nav](http://sagepub.com/journalsPermissions.nav)

DOI: 10.1177/0193841X16664811

[journals.sagepub.com/home/erx](http://journals.sagepub.com/home/erx)



Alexandra Bonardi<sup>1</sup>, Christine J. Clifford<sup>1</sup>,  
and Nira Hadar<sup>2</sup>

## Abstract

**Background:** This review describes the methods used for a systematic review of oral health intervention literature in a target population (people with intellectual and developmental disability (I/DD)), which spans a broad range of interventions and study types, conducted with specialized

---

<sup>1</sup> University of Massachusetts Medical School, Worcester, MA, USA

<sup>2</sup> School of Public Health, Brown University, Providence, RI, USA

## Corresponding Author:

Christine J. Clifford, University of Massachusetts Medical School, 55 Lake Avenue North, S3-301, Worcester, MA 01655, USA.

Email: [christine.clifford@umassmed.edu](mailto:christine.clifford@umassmed.edu)

software. **Objective:** The aim of this article is to demonstrate the review strategy, using the free, online systematic review data repository (SRDR) tool, for oral health interventions aimed at reducing disparities between people with I/DD and the general population. **Research Design:** Researchers used online title/abstract review (Abstrackr) and data extraction (SRDR) tools to structure the literature review and data extraction. A practicing clinician and an expert methodologist completed the quality review for each study. The data extraction team reported on the experience of using and customizing the SRDR. **Results:** Using the SRDR, the team developed four extraction templates for eight key questions and completed extraction on 125 articles. **Conclusions:** This report discusses the advantages and disadvantages of using an electronic tool, such as the SRDR, in completing a systematic review in an area of growing research. This review provides valuable insight for researchers who are considering the use of the SRDR.

### **Keywords**

methodological development, content area, physical health care and policy, content area, systematic review, oral health, intellectual/developmental disability

Systematic reviews are a valuable tool to summarize the body of evidence on a particular question. For topic areas in which the body of evidence is developing, and research questions require the inclusion of studies with a range of study designs and methodologies, there is limited guidance on approaches to systematically review and examine available evidence. The structured reviews are nevertheless critical to synthesize and advance the understanding of existing research (Petticrew & Roberts, 2006).

This article explores the use of a systematic review methodology in an emerging body of literature focused on oral health interventions for people with intellectual and developmental disability (I/DD). In response to demonstrated oral health disparities in the population with I/DD (Anders & Davis, 2010; U.S. Department of Health and Human Services, 2000), clinical, policy, and public health interventions are being identified to improve oral health and reduce health disparities in this population. There is variability in the degree to which these interventions are being documented in research studies, with few controlled study designs. The body of literature is therefore broad in the type of interventions described as well

as in the types of research design. This challenges standard systematic review approaches to data extraction and synthesis of results using electronic or other data extraction tools. A structured approach must be imposed, however, in order to allow for clear and objective review of findings in available studies. Using the web-based Systematic Review Data Repository (SRDR), the authors describe efforts to use a structured approach to complete a systematic review of a relatively broad range of oral health and behavioral interventions, and study designs, in the existing literature. The experience and findings described are intended to inform future systematic reviews in areas of study with similarly varied intervention types and study methodologies.

Effectiveness research focused on the population with I/DD is in an early and developing stage. There are few published systematic reviews of interventions specific to this population. (Brylewski & Duggan, 1999; Deb et al., 2008; Spanos, Melville, & Hankey, 2013). Among those that exist, findings and recommendations are often presented with caution because of limited numbers of eligible studies, and design or methodological flaws among the studies reviewed that limit generalizability of findings. Brooker et al. (2015) describe factors in research that reduce the likelihood that people with I/DD will be included and identified in research that meets high standards for the level of evidence often seen through the use of randomized control trials (RCTs). These factors include designs that explicitly exclude people with I/DD for the purpose of easing consent requirements for research as well as designs in which the population *may* be included, but there is no means for distinguishing outcomes for this subpopulation. Existing literature must therefore be examined thoroughly and appropriately for its potential, in isolation or combined with, other studies to build the evidence base for effective interventions.

In order to build the evidence base for effective interventions, researchers must establish and demonstrate consistent approaches to synthesize studies in the population across a range of settings and interventions. The aim of the current study was to pilot, demonstrate, and describe a valid approach to a systematic review as a foundation for future reviews in this population. We explored and assessed the applicability of existing tools developed to synthesize and advance the methodology of systematic review research in the population with I/DD.

The SRDR 1.0 (n.d.) tool is a free, web-based, data extraction and management tool for systematic reviews and meta-analyses, funded by the Agency for Healthcare Research and Quality (AHRQ), and maintained at Brown University Evidence Based Practice Center (<http://srdr.ahrq.gov/>).

The tool provides researchers with a platform from which to design and complete a systematic review extraction. The customized extraction tool and its extracted data are maintained in a database that is available to researchers online and can be made public at the completion of the systematic review. This feature allows for transparency in definitions of populations, interventions, and outcomes and holds the potential for systematic reviews to build upon the original research as the evidence accumulates (Robinson et al., 2014). The SRDR has been used in multiple traditional systematic reviews in which the studies reviewed conform primarily to RCT designs. As a free and publicly available tool, the study team chose to test the utility of the SRDR for use in the current systematic review, with a range of interventions and study designs, many of which did not conform to the clinical trial design and reporting structure that is typically included in systematic reviews.

At the outset of the study, two research questions were identified to examine interventions that aim to increase access to care as well as interventions to support and improve good oral health behavior as a means to improve oral health status: (1) What effective interventions/strategies exist to improve access to oral health care for the I/DD population and (2) what effect do interventions that support good oral health behaviors have on improved oral health care for the I/DD population?

While the population is well defined (people with I/DD), the two research questions required the study team to include for review a range of intervention types, with a range of anticipated outcomes. This article examines the approaches and feasibility of using the SRDR as an electronic data collection tool in a systematic review that goes beyond narrowly defined intervention-outcome studies. Results include the final approaches adopted by the research team as well as lessons learned from the research team in data extraction for a varied set of articles into SRDR.

## Method

The systematic review described adheres to the core principles of systematic reviews: The scope of the review was defined, in advance, with identified research questions; a comprehensive search was completed and documented; explicit inclusion and exclusion criteria were applied; there were explicit methods of extracting and synthesizing study findings; and established standards critical to review study quality were applied (Higgins & Green, 2011).

**Table 1.** Concept/Search Terms.

| Key Words                                  |   |                              |
|--|---|------------------------------|
| Population                                 | Access to Care                            | Health Behaviors             |
| Oral health, dental/dentistry,<br>AND I/DD | Access, treatment, dental<br>services     | Dentist, fear                |
| Mental retardation                         | Health education                          | Oral hygiene                 |
| Physical disability                        | Outcomes (long-term and<br>short-term)    | Tooth brushing               |
| Learning disability                        | Providers, dentists,<br>hygienists        | Flossing                     |
| Cerebral palsy                             | Payments/costs                            | Fluoride                     |
| Down syndrome                              | Models (service delivery)                 | Adaptive tools/<br>equipment |
| Neurodevelopmental disorder                | Mobile clinics<br>Special needs dentistry |                              |

Note. I/DD = intellectual and developmental disability.

A detailed search strategy was developed with the assistance of a research librarian in consultation with the study team and advisory panel and was conducted in two stages. Initially, studies were identified by searching electronic databases, reviewing the resources from professional and state dental organizations, scanning reference lists of articles, and consultation of experts in the field. The search was limited to articles published between 1990 and June 2013 (inclusive). Dates were chosen (1) to focus on care post deinstitutionalization and (2) to focus on current practices, techniques, and technologies for improving access, and behaviors for individuals with I/DD. Databases searched included MEDLINE (PubMed, 1966- PRESENT), CINAHL, EMBASE, Scopus, Google Scholar, Cochrane, ERIC, PsycInfo (OVID), and Global Health (OVID). Other sources for studies were PubMed Central, BioMed Central, and TRIP. Key articles' references/bibliographies were hand searched for additional relevant publications. In addition, gray literature resources, Scirus, New York Academy of Medicine, and OAISTER (OCLC) were searched. All relevant dental associations' websites were searched for white papers and reports. The search was revised appropriately for each database to take into account differences in controlled vocabulary and search requirements, such as truncation of search words. Proposed search terms were reviewed to include the population, the intervention, and the outcomes. A search string was developed from a concept table and modified appropriately for each database (Table 1).

Following guidelines laid out in AHRQ's Community Guide: The Guide to Community Preventive Services (n.d.; <http://www.thecommunityguide.org/about/methods.html>), the study team began the search for, and development of, an extraction tool. The team considered adapting a variety of extraction tools, or developing its own tool using Access, Excel, or Word tables. However, the tools proved cumbersome for the type and amount of data the team planned to collect. Ultimately, the team chose the SRDR and its companion tool the Abstrackr 1.0. The team determined that the structure of the SRDR, the ability to modify the extraction questions, the online aspect, and the ability to work with systematic review experts would prove beneficial to our review process.

Abstracts were uploaded to Abstrackr (Wallace, Small, Brodley, Lau, & Trikalinos, 2012), a free, web-based citation-screening tool. The tool allows multiple reviewers at various locations to participate with ease and allows for a quick and easy response to include/exclude, tag (label), or identify the need for more information. Abstracts were uploaded electronically, from a RefWorks database, and reviewers were randomly assigned a group of abstracts to review both the title and abstract for inclusion criteria. Reviewers were trained on the tool as well as the inclusion/exclusion criteria prior to the start of the review; after 2 weeks, a check-in was conducted. Each abstract was reviewed twice and assigned an inclusion/exclusion tag. When there was disagreement, the study team reviewed the title and abstract and made the final decision. Criteria were applied to identify the I/DD population and intervention studies only, and inclusion/exclusion tags were assigned, thus reducing the number of full-text reviews. At this stage, any study design was included, but commentaries and editorials were excluded (Table 2).

The number of articles remaining in the body of literature after title and abstract review was 602 (Figure 1). Full-text screening was conducted with these articles to examine whether the intervention types described in the research literature were responsive to the key questions. Identified interventions covered a broad range and were categorized by the research team to allow for a focused review by topic area, essentially developing a secondary (sub) series of targeted systematic reviews. The categories identified were (1) home and community-based prevention strategies such as tooth brushing, (2) office-based prevention strategies such as the use of fluoride, (3) sedation and anesthesia use, (4) specific behavioral interventions, (5) education for treatment and prevention directed at providers and caregivers, (6) access to care such as insurance interventions, (7) interventions to address drooling in the population with neurological impairment

**Table 2.** Inclusion/Exclusion Criteria.

| Include                                   | Exclude   |
|---|---|
| Population with I/DD clearly identified   | Background article (state of problem, editorial)  |
| Oral health intervention English language | Not relevant (did not address key questions)  |
| 1990–June 2013                            | No intervention described<br>Not population of interest, population could not be determined, or population not reported/identified in results |
|   | Prior to 1990   |
|   | No abstract available   |
|   | Not peer-reviewed journal <sup>a</sup>  |

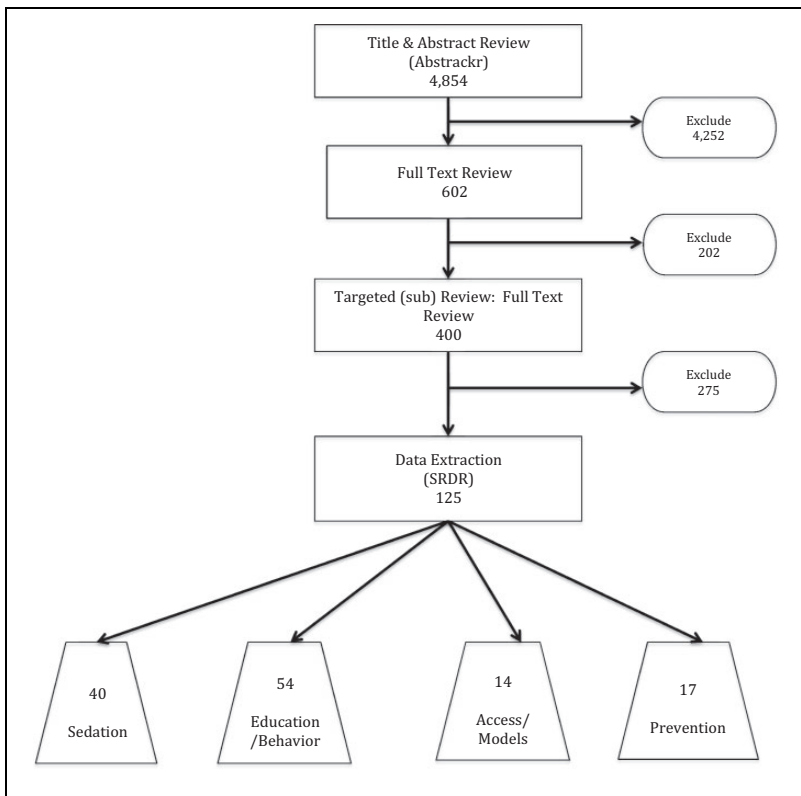
Note. I/DD = intellectual and developmental disability.

<sup>a</sup>In some cases, gray literature was included if met inclusion criteria.

and I/DD, and (8) “others” such as implants and orthodonture. Additional screening conducted by the research team for relevance to this systematic review resulted in approximately 400 articles for further review. A second search was performed for each targeted subject area in order to confirm no literature was missed, that is, *population terms AND dental terms AND sedation*.

Within the repository, project-based extraction tools are developed using standard systematic review principles. Following the structure of the SRDR “tabs,” data extraction questions were developed allowing for summarization of the literature in an easily extracted, manipulated, and reportable format. Two members of the study team received in-person training on the SRDR and initiated the development of the extraction tools. Starting with a broad-based framework, articles were reviewed for data points of interest for extraction. These data points included such items as study type, interventions, outcomes, and relationship to the study questions. The SRDR is organized by “tab,” and each tab is designed to draw out specific data for the review and to focus the extractor on collecting pertinent information for the analysis. The framework was applied to the SRDR tabs. The tabs are as follows:

- *Key question:* allows for the alignment of the article to the appropriate key question;
- *Publications:* includes the citation and abstract for the article under review;



**Figure 1.** Modified prisma chart.

- *Design*: includes questions on study design or other design details such as how an intervention is measured or to whom it is targeted;
- *Arms*: identifies the intervention(s) such as a drug, device or other intervention;
- *Arm details*: extracts detail of the intervention, dosage of a drug, or specific type of intervention such as modeling;
- *Baseline*: contains details of the study population such as sample size, gender and age;
- *Outcomes*: identifies the outcomes and whether they are categorical or continuous;
- *Outcomes details*: contains the data on how outcomes were measured;



- *Results*: includes actual results of the statistical analysis of the intervention and can include such items as standard deviation and other statistical information needed for a meta-analysis;
- *Adverse events*: identifies any adverse events in a study such as an adverse drug reaction;
- *Quality*: allows for a series of questions designed to measure the quality of the study design, intervention, and analysis; and
- *Finalize*: an administrative tab allows the extractors and project lead to follow the progress of the extraction and add notes.

Using the information collected in our framework, the study team navigated the SRDR tabs and internal standardized “question builder,” to develop an extraction tool for each intervention type. The first topic area was “sedation” and the study team developed specific key questions, refining the focus of the overarching key questions, which targeted access to oral health services, as well as oral health behaviors, to sedation.

- *Sedation key question 1*: What is the relationship between sedation methods and oral health care outcomes for individuals with I/DD?
- *Sedation key question 2*: What indicators were used to determine the need for, and what type of, sedation to use when providing oral care to an individual with I/DD?

The study team piloted two articles and adjusted the extraction form based on feedback regarding usability and ability to extract information. Once the customized extraction tool was finalized, the sedation articles were randomly distributed to extractors. In order to validate the results, an additional member of the research team provided a review of the data in the Results tab of each article.

Subsequent topic areas were approached similarly, beginning with the development of intervention-topic-specific key questions and followed by the development of a customized extraction tool that addressed the features, outcome types, and result types of each area. The structure and design of the extraction questions allowed the team to take advantage of similar intervention approaches and, in some cases, combine extraction tools. For example, education and behavior interventions might be delivered in a similar fashion, such as modeling a behavior, but applied to different populations such as providers versus caregivers. Being aware of the similarities allowed the researchers to draft one extraction form that minimizes questions yet identified differences available in the details of the articles. This process

**Table 3.** Key Questions.

|                        |  |
|------------------------|--|
| Sedation               | <p>Key question 1: What is the relationship between sedation methods and oral health-care outcomes for individuals with I/DD?</p> <p>Key question 2: What indicators were used to determine the need for, and what type of, sedation to use when providing oral care to an individual with I/DD?</p>   |
| Education/<br>behavior | <p>Key question 1: Do professional educational experiences for dentists, hygienists, and other providers impact access to or improvement in oral health care for individuals with I/DD?</p> <p>Key question 2: Do educational programs targeted to caregivers (direct care staff, families, and others) impact oral health in individuals with I/DD?</p> <p>Key question 3: Do educational programs, with behavioral intervention components, targeted in individuals with I/DD impact or improve their oral health?</p> |
| Access/models          | <p>Key question 1: What models are effective in improving access to oral health care of individuals with I/DD?</p> <p>Key question 2: What models are effective in improving oral health-care outcomes for individuals with I/DD?</p>  |
| Prevention             | <p>Key question 1: What office-based preventive treatments (such as fluoride) improve oral health outcomes in individuals with I/DD?</p>   |

Note. I/DD = intellectual and developmental disability.

was followed for the four remaining topic areas; unique key questions were developed and specific extraction forms were created (Table 3).

The study team removed interventions that addressed drooling, implants, and orthodonture. Interventions focused on drooling, though relatively common in this literature, did not address our original key questions on disparities in access or behavior. Additionally, there is debate among physicians and dental providers as to the value of reducing drooling on oral health; it can be detrimental to oral health as adequate intraoral saliva is required for good oral health, reduced saliva may increase the risk of caries (Ferraz Dos Santos, Dabbagh, Daniel, & Schwartz, 2015; M. Holder, personal communication, January 22, 2013). Additional articles on implants and orthodonture did not directly address the key questions and therefore were also removed from the review.

The remaining topic areas led to a set of targeted SRDR extraction questions and four unique extraction forms. While some SRDR questions were similar across extraction forms (i.e., study design, sample size, etc.), others were unique to the topic area. For example, delivery method of

sedation versus delivery method of a tooth brushing intervention. Once all four extraction forms were completed, remaining articles were randomly assigned to extractors. The team of extractors met weekly to review articles, share tips, and discuss progress of the extraction.

### *Quality Review*

The research team partnered with clinical experts (i.e., dentists and dental hygienists), as well as a biostatistician, to gain a multifaceted assessment of the quality of each article. Contemporaneous to the data extraction by members of the research team, clinical and statistical experts performed a quality review for each article. Using questions adapted from The Community Guide's resources (Briss et al., 2000), the clinical experts and biostatistician answered the same questions but from their respective perspectives. Reviewers were asked to review and rate the study population, intervention description, sampling strategy, measurement of the intervention, measurement of the outcomes, data analysis, and interpretation of results. Clinicians completed a Word form which was then data entered into the SRDR. The biostatistician reviewer received training in use of the SRDR and entered the quality review data directly into the SRDR.

Finally, a significant advantage of using SRDR or another electronic tool customized for systematic review processes is the ease of data retrieval and analysis. With a completed set of extracted data, the SRDR can be downloaded into a variety of reporting formats including Excel and comma separated values for analysis by intervention type, outcomes, and quality of the studies. In fact, any data field can be used as a unit for sorting, synthesis, or analysis as appropriate.

### *Collection of Extractor Feedback*

The study team was interested in testing the SRDR as a tool to facilitate, or provide a model for, systematic review processes in a body of literature with limited RCTs, and with multiple reviewers and data extractors, including researchers from the community who were not experts in systematic review. Data extraction was completed by the study team in order to maintain consistency, while other experts were included to provide their input in the critical area of assessment of quality for the body of literature. The use of the SRDR, as a structured data extraction tool, introduced a number of constraints that proved challenging for extraction from such a range of interventions. After the extractions were complete, we asked for feedback

from the study team using a semi-structured online survey tool to explore questions about whether the SRDR was a feasible and helpful tool for future systematic reviews to build the evidence base in this population. Extractor feedback is summarized in the Results section.

## Results

The authors used a multiphase approach to examine the feasibility of using the SRDR for this systematic review. The initial title and abstract Abstrackr team consisted of 10 reviewers: 2 dentists, 4 dental students, 2 researchers, one librarian, and one medical student.

Following the completion of the title and abstract review, with the resulting literature, the team developed four extraction forms with a total of eight key questions. Each topic area defined what was essentially a unique systematic review, with approximately 100 data fields extracted per article. The review team consisted of 5 data extractors and 16 reviewers of clinical quality (9 dentists, 2 primary care doctors, 1 anesthesiologist, 1 nurse, 2 hygienists, and 1 state government administrator). One student intern conducted a validity check on the results tab and made adjustments in consultation with the Principal Investigator and the project coordinator. In addition, one biostatistician conducted a quality review from the statistical perspective. When piloting the sedation extraction, it became apparent that clinicians did not have the time to complete the SRDR training or to devote the time needed to become familiar with the extraction forms. The clinicians were comfortable in reporting on clinical quality approaches and in identifying what interventions are helpful to their clinical practice; however, they were not entirely comfortable or able to determine the quality of study design or analytic approach. What may be of value in the literature to a practicing dentist is often seen differently with a critical review of methodology and analytic approaches. Therefore, the study team developed a second quality review conducted by a research biostatistician. At the conclusion of the analysis, the completed extraction forms as well as all the extracted data will be made publicly available directly from the SRDR website. Overall, the extraction tools allowed for the continued application of the inclusion/exclusion criteria, with the end result of a total of 125 articles extracted across the four extraction forms (Figure 1). In addition, the completed extraction resulted in the review and analysis of a total of 227 interventions and 348 outcomes across the four extraction topics (Table 4). Twenty-seven percent of the extracted studies across all topics were RCT or

**Table 4.** Results Per Extraction Form.

|                        | Number of Key Questions | Number of Extracted Articles <sup>a</sup> | Number of Extractors <sup>a</sup> | Number of Interventions <sup>b</sup> | Number of Outcomes <sup>b</sup> |
|------------------------|-------------------------|---|-----------------------------------|--------------------------------------|---------------------------------|
| Sedation               | 2                       | 40  | 3                                 | 69                                   | 139                             |
| Education/<br>behavior | 3                       | 54  | 2                                 | 87                                   | 126                             |
| Access/<br>models      | 2                       | 14  | 2                                 | 17                                   | 35                              |
| Prevention             | 1                       | 17  | 1                                 | 54                                   | 48                              |

<sup>a</sup>Does not include extractor conducting validity check. <sup>b</sup>Preliminary numbers.

RCTs with crossover design, the remaining studies were nonconforming designs (Table 5).

To further explore the feasibility of the SRDR tool, we asked extractors to report on the clarity of the training, ease of use, amount of time to learn and feel comfort with extraction, the challenges, the positives, and if they would use the tool again. Feedback indicated that the training for the extractors ranged from 1 hr to 4 hr, depending on whether the extractor completed an online or an in-person training. Respondents reported that it took between 3 and 10 article extractions until they were completely comfortable with the tool, and that often they needed to go back to extractions to adjust and readjust the outcomes and results as they became more familiar with how to set up these sections in SRDR. The team also reported that it took anywhere from 45 minutes to six hours to extract an article. The wide variability was measured in how clear the article was written, how clear the interventions and outcomes were described, and how many results were analyzed and published within the article. The extraction team reported that they recognized the power of the system once they had entered approximately four to six articles. Overall, the extractors felt that once trained, the SRDR was a valuable tool for data extraction.

## Discussion

The biggest challenge for the extractors included entering the results and outcomes of the interventions in a manner that was consistent and aligned with the structure of the SRDR. The system was designed for clinical trials with clear intervention, control, and outcome data. It required careful interpretation of findings to define and then enter outcome measures and results

**Table 5.** Design Type by Topic.

| Topic              | RCT/RCT With<br>Crossover | Nonrandomized<br>Trial | Retrospective<br>Cohort | Prospective<br>Cohort | Cross-<br>Sectional | Survey | Other <sup>a</sup> |
|--------------------|---------------------------|------------------------|-------------------------|-----------------------|---------------------|--------|--------------------|
| Sedation           | 10                        | 2                      | 15                      | 11                    | 1                   | —      | 1                  |
| Education/behavior | 10                        | 6                      | —                       | 9                     | 3                   | 6      | 20                 |
| Access/models      | —                         | —                      | 2                       | 1                     | 4                   | 3      | 4                  |
| Prevention         | 14                        | 2                      | —                       | 1                     | —                   | —      | —                  |

Note. RCT = randomized control trials.

<sup>a</sup>Includes descriptive, case-control, qualitative, convenience samples, and so on.

from the types of studies that were included in our review such as qualitative, descriptive, survey, and quasi-experimental designs. In numerous cases, the extractors relied on trial and error, followed by review with other members of the study team to confirm reporting and discuss how to interpret the data. The process allowed a variety of extractors at different skill levels a standardized approach and structure to the extraction.

The intervention type was related to the ease of data extraction from the articles, in part because some interventions were more aligned with a clinical trial type approach. The sedation and prevention extraction forms proved easier to create the articles easier to extract as compared to education/behavior and access/models. Measures in the sedation and prevention extraction forms were primarily quantitative in nature versus the qualitative measures that are more prevalent in the latter study areas.

There was a mixed response on whether reviewers (members of the review team) would recommend the use of the SRDR in a future systematic review in this population, and with such a variety of interventions. It is not an intuitive tool for many users but after being trained on the application, extractors saw the value in using it for more traditional systematic reviews. Extractors did report wanting to see a more flexible approach to entering results that may not derive from RCTs. The ability to work on the SRDR from any location with an Internet connection, allowed for broader access to reviewers, the ability to share the work in a public forum, and receive feedback on the process, all benefits to using the technology. Many of the challenges reported by the extractors were not unique to using the SRDR as these challenges and questions arise in any systematic review process. While the SRDR was chosen in part because of the ability to readily make public the extracted data for future reviews, there have not been any examples of this feature being used to date. The SRDR has not documented a publicly shared review revisited for either duplication or expansion (Dr. Lau, personal communication, December 10, 2014).

The SRDR's web-based access provided a "template" for question design, which allowed for control over the development of extraction questions. The tool provided standardization in training and extraction and ease of organizing and reporting data. Training extractors and reviewers was time-consuming, requiring a 1-day training (attended by study team reviewers), or a 2- to 3-hr training in order to gain mastery (for remote reviewers). The inclusion of clinical reviewers for quality added an important dimension to this review. Their quality reviews were captured in the SRDR, however our "volunteer" reviewers did not have the time to learn the tool, rather their reviews were done "on paper," with members of the

study team doing data entry. In the context of this study, limitations of the SRDR were primarily the time necessary for training and to feel comfortable with use of the online system, particularly in the extraction of results.

The research team has completed both the statistical and clinical quality reviews and is in the process of analyzing the relationship between the results of the two types of reviews. As there is a recognized risk of clinical bias in the review by clinicians, the ability to examine quality reviews by input from statistical expert and clinical expert will allow the research team to examine the impact of this potential bias and determine the value of bringing these two perspectives into a systematic review such as this.

## Conclusion

This work describes the methods used for developing a systematic review of oral health intervention literature in a target population (people with I/DD), spanning a broad range of interventions and study types. Interventions varied significantly, outcomes were variably reported, and the quality of study design and analysis varied, creating a highly complex set of results for synthesis. While the research team found that the SRDR data extraction tool did assist with an organized extraction of data from included articles, the forced structure of the SRDR made data extraction cumbersome. Those studies that employed control trial methodologies were most well suited to extract using SRDR.

The review did identify and, more importantly, synthesize evidence that previously had not been assembled or analyzed. As discussed in the introduction, research protocols may limit the inclusion of people with I/DD in studies, or make it impossible to identify this population, thereby limiting the ability to extend the evidence base for interventions in this population. Use of the SRDR, coupled with clinical and statistical expert review, allowed for the systematic examination of a body of literature that may have otherwise been excluded from traditional systematic review approaches. This approach, combining data extraction with a quality review, is proposed as a means to support the advancement of an emerging evidence base such as interventions specific to the I/DD population. It may also serve as a model for researchers exploring other populations in which the base of evidence has similar limitations.

Additional advantages of the SRDR include the ability to publish the final data extraction once the research team has analyzed it. This is a valuable feature because it creates the potential for future studies by other research groups that build upon this review which may inform both



evidence-based clinical practice and public policy. The public nature of the raw extracted data bolsters efforts to ensure transparency in the completion of the systematic review.

Researchers who are considering a systematic review of a body of literature that includes a range of study designs may consider the use of SRDR or another structured data extraction tool to facilitate the review. They should, however, proceed with caution. While the SRDR does allow for a structured capture of data and quality of the body of literature, it still requires effort on the part of the research team to learn and customize the tool in preparation for extraction. The extra effort is a necessary and valuable step in building the evidence base in a developing field such as oral health interventions in the population with I/DD.

### **Acknowledgements**

The authors would like to acknowledge the members of the Oral Health Systematic Review Advisory Panel for their assistance in guiding the review, the Title & Abstract Reviewers, the Quality Reviewers, Nancy Harger, Research Librarian, for guiding the development of the search string and collecting articles, the Data Extractors, and Bryant Smith at Brown's EPC for his technical assistance with the SRDR.

### **Declaration of Conflicting Interests**

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

### **Funding**

The author(s) disclosed receipt of the following financial support for the research, authorship, and/or publication of this article: Project funded by Centers for Disease Control & Prevention/National Center on Birth Defects & Developmental Disabilities/Division of Human Development & Disability Cooperative Agreement # 5U01DD000914.

### **References**

- Anders, P. L., & Davis, E. L. (2010). Oral health of patients with intellectual disability: A systematic review. *Special Care in Dentistry*, *30*, 110–117. doi: 10.1111/j.1754-4505.2010.00136.x
- Briss, P. A., Zaza, S., Pappanoanou, M., Fielding, J., Wright-DeAguero, L., Truman, B. I., . . . Harris, J. R. (2000). The task force on community preventive services, the community guide, developing an evidence-based guide to community preventive services—Methods. *American Journal of Preventive Medicine*, *18*, 75–91.

- Brooker, K., van Dooren, K., Tseng, C. H., McPherson, L., Lennox, N., & Ware, R. (2015). Out of sight, out of mind? The inclusion and identification of people with intellectual disability in public health research. *Perspectives in Public Health, 135*, 204–211. doi:10.1177/1757913914552583
- Brylewski, J., & Duggan, L. (1999). Antipsychotic medication for challenging behaviour in people with intellectual disability: A systematic review of randomized controlled trials. *Journal of Intellectual Disability Research, 43*, 360–371.
- Community Prevention Services Task Force. (n.d.) Systematic review methods. Retrieved from <http://www.thecommunityguide.org/about/methods.html>
- Deb, S., Chaplin, R., Sohanpal, S., Unwin, G., Soni, R., & Lenotre, L. (2008). The effectiveness of mood stabilizers and antiepileptic medication for the management of behaviour problems in adults with intellectual disability: A systematic review. *Journal of Intellectual Disability Research, 52*, 107–113. doi:10.1111/j.1365-2788.2007.00965.x
- Ferraz Dos Santos, B., Dabbagh, B., Daniel, S. J., & Schwartz, S. (2015). Association of onabotulinum toxin a treatment with salivary pH and dental caries of neurologically impaired children with sialorrhea. *International Journal of Paediatric Dentistry*, March 2, 2015, [epub ahead of print]. doi:10.1111/ipd.12156
- Higgins, J. P. T., & Green, S. (Eds.). (2011). *Cochrane handbook for systematic reviews of interventions* (Version 5.1.0). The Cochrane Collaboration. Retrieved from [www.cochrane-handbook.org](http://www.cochrane-handbook.org)
- Petticrew, M., & Roberts, H. (2006). *Systematic reviews in the social sciences: A practical guide*. Malden, MA: Blackwell.
- Robinson, K. A., Whitlock, E. P., Oneil, M. E., Anderson, J. K., Hartling, L., Dryden, D. M., . . . Chang, S. (2014). Integration of existing systematic reviews into new reviews: Identification of guidance needs. *Systematic Reviews, 3*. doi: 10.1186/2046-4053-3-60
- Spanos, D., Melville, C. A., & Hankey, C. R. (2013). Weight management interventions in adults with intellectual disabilities and obesity: A systematic review of the evidence. *Nutrition Journal, 12*, 132. doi:10.1186/1475-2891-12-132
- Systematic Review Data Repository. (n.d.). Retrieved December 1, 2014, from <http://srdr.ahrq.gov/>
- U.S. Department of Health and Human Services. (2000). *Oral health in America: A report of the surgeon general*. Rockville, MD: U.S. Department of Health and Human Services, National Institute of Dental and Craniofacial Research, National Institutes of Health.
- Wallace, B. C., Small, K., Brodley, C. E., Lau, J., & Trikalinos, T. A. (2012). Deploying an interactive machine learning system in an evidence-based practice

center: Abstractkr. *Proceedings of the 2nd ACM SIGHIT Symposium on International Health Informatics—IHI '12*. doi:10.1145/2110363.2110464

## Author Biographies

**Alexandra Bonardi**, MHA, OTR/L, studies trends and patterns in health and service outcomes for people with intellectual and developmental disabilities and ways to improve the health status of this population. Projects include the development of preventive health screening guidelines for adults with intellectual and developmental disability, the coordination of a national process to operationalize a definition of intellectual disability that can be used for health surveillance, falls reduction, the promotion of good oral health, and access to adaptive equipment and technologies to support independent function.

**Christine J. Clifford**, MHP, is well versed in public health research methodology and underserved populations. Her projects include developing retraining for people aging with intellectual and developmental disability, social innovation for inclusion on behalf of people with intellectual and developmental disability, and the systematic review of interventions to reduce oral health disparities between adults with intellectual disability and the general population. Her experience also includes project design, data collection, and analysis, including enrollment data, claims data, and survey data.

**Nira Hadar**, PhD, has a doctorate in epidemiology. While at the Brown University Evidence Based Practice Center, she has provided technical, data, and training expertise on the Systematic Data Review Repository. In addition, to being an innovator in data collection and management, she has published articles on a variety of medical and pharmaceutical interventions.