Looking at Europe's recent behavioral telehealth practices for children and families impacted by neurodevelopmental disabilities

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There is a widespread lack of behavioral professionals available to support children and families affected by neurodevelopmental disabilities. As a result of limited availability, services that can be provided from a distance have developed. Telehealth is a modality that can increase access to services, lessen financial constraints, and support assessments of generalization. Using either synchronous or asynchronous components it can foster evaluation and coaching. Guidelines for usage have surfaced in North America and been integrated into the continent's existing model of behavioral care. However, in Europe where all modalities of behavioral services are fighting to receive funding, frameworks are scarce. Understanding more about telehealth in behavioral care, its various applications throughout Europe, and the local context into which it can be applicable may promote system growth. To support this cause, a scoping review of recent behavioral telehealth practices for children and families impacted by neurodevelopmental disabilities in Europe was undertaken; looking specifically to assess types of studies, their targets and outcomes, telehealth modality components, barriers, and directions for future work. Although few studies surfaced, valuable conclusions can be drawn about the model's empirical validation, creating a groundwork for sustainability, and the need for developing policy and standardized application.

Keywords: Telepractice; autism; applied behavior analysis; European Union; telemedicine

A look at recent telehealth practices for children with neurodevelopmental disabilities in Europe

Children with neurodevelopmental disabilities can present with challenges in different areas (Ritzema *et al*. 2016). Areas of struggle may include socialization, communication, cognition, and self-care (Lollar *et al*. 2012). Resulting behavioral challenges can substantially decrease the child and family's quality of life. Parents of children with neurodevelopmental disabilities may also face increased financial challenges, marital disharmony, social isolation, and reduced quality of life (Gardiner *et al*. 2018).

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Strategies grounded in the science of applied behavior analysis (ABA) have been empirically demonstrated to increase skills by using systematic teaching methods, incremental instruction, and function-based approaches (e.g. Virués-Ortega 2010). ABA-based services can involve different tactics. Some of these include a verbal behavior approach (e.g. Barbera 2007), naturalistic teaching strategies (e.g. Schepis *et al*. 1998), functional communication training (e.g. Tiger *et al*. 2008), discrete trial training (e.g. Smith 2001), and self-management (Lee *et al*. 2007). Given the complex nature of the science of behavior, practicing competently requires specialized educational courses and training (Shook and Neisworth 2005). The specialized coursework must adhere to a certain number of hours of discretionary content. This content includes ethics, professional conduct, philosophical underpinnings and principles of behavior, measurement, analysis, experimental design, behavior assessment, and behavior change procedures (Association for Behavior Analysis International, 2018). Training requirements include the completion of

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supervised practicums across numerous competency areas which take, on average, over a year to complete (Behavior Analyst Certification Board, 2021b). The rigorous nature of working towards becoming a behavioral practitioner has contributed to the dearth of people available to provide ABA services.

As a result, many children and families affected by neurodevelopmental disability cannot access ABA services. While available in parts of North America and some metropolises in other countries, there is a lack of ABA services in areas of the world (Ferguson *et al*. 2019; Salomone *et al*. 2014). In Europe there are exceedingly fewer behavior practitioners than in North America (Keenan *et al*. 2015). At the time of writing this manuscript there were 1349 registered behavior analysts across the 49 European countries targeted in this review (Behavior Analyst Certification Board 2021a, March 2). This number is further put in perspective when considering that in the small US state of Florida there are over 5000 registered behavior analysts (Behavior Analyst Certification Board 2021a, March 2). Moreover, Keenan and colleagues (2015) reviewed behavior analysis in Europe, finding that unvalidated eclectic models of treatment are commonly used. In the Czech Republic for example, it was found that behavior analytic services were scarcely available for children with autism, and that current behavior analysts were spread very thin with many practitioners working without needed resources, social support, or mentorship opportunities (Kingsdorf and Pančocha 2020).

This lack of available behavioral professionals is widespread and ongoing. For example, rural communities in the United States have historically been left with fewer or lower quality ABA services (Mello *et al*. 2016). Other places across the Middle East (Eid *et al*. 2017), Africa (Ruparelia *et al*., 2016), Asia (Manohar *et al*. 2019), and Latin America (Montiel-Nava *et al*. 2020) are also facing this challenge. As a result of the limited access worldwide, services that can be provided from a distance have developed.

History of telehealth practices

Although not coined by the field of behavior analysis, delivering ABA services from a distance has been termed telehealth. Modern telehealth practices of providing behavioral services to children and families impacted by neurodevelopmental disabilities grew out of the field of telemedicine. In the field of ABA the terms telehealth and telemedicine are often used interchangeably. However, telemedicine, is a field broadly defined as providing medical information and services over a distance by telephone or other means of broadcasting or cable, and started in the 1960s (Bashshur and Shannon 2009), but began gaining popularity in the 1990s (Perednia and Allen 1995). It originated as sending still images between patients and doctors to allow for diagnosis of ailments such as skin conditions. Ordinary telephone communication was also used to allow for therapeutic consultations with medical professionals. Other early modalities and applications included medical education and case management using short video clips, sound, or voice. Over time, application became more complex with real-time one-way or two-way video sharing for services such as psychiatric evaluations. As bandwidth has become more available and digital technologies more accessible, the field has continued to grow. Despite issues in clinical expectations, legality, and economic factors, telemedicine has still allowed medical services to become available to individuals in rural and underserved areas (Perednia and Allen 1995). Some notable examples of positive telemedicine outcomes around the world include the 237,221,884 kilometers of patient travel avoided in 2012 in Ontario (Ontario Telehealth Network, 2013), and the $15.8 million USD saved in the central United States during a 5-year period when eEmergency services were used to avoid patient transfers (Kansas Health Institute 2014). With such successes, it is not surprising that the field has evolved to include service variations.

Telehealth is one such variation. Telehealth, also known as telepractice, is now often considered to encompass training, teaching, or coaching services provided using online delivery and two-way audio-visual communication (Rispoli and Machalicek 2020). Although at the most basic level telehealth involves the movement of images and data, advanced technologies have been utilized to improve the outcomes.

Telehealth technologies

A myriad of telehealth technologies can be identified throughout the literature. Two major categories of telehealth technology can be distinguished based on the temporal presence and mode of participation. Synchronous, or real-time support, may involve one-on-one parent and practitioner meetings or one-on-one service delivery between the behavior practitioner and the child done via videoconferencing or telephonically. Asynchronous support, where practitioner and client do not meet at the same time, may involve the sending of videos by parents to practitioners, the independent review of videos by practitioners, the review of electronic learning materials by parents, and monitoring the attribute of a client from afar (e.g. sleep patterns). When it comes to providing synchronous didactic instruction or coaching, some studies have demonstrated success using real-time video conferencing tools like Skype (e.g. Pantermuehl and Lechago 2015), FaceTime (Boutain *et al*. 2020) and the Google platform (e.g. Pennefather *et al*. 2018). In the case of asynchronous telehealth components, some studies have demonstrated success using common file storage applications like Dropbox for video sharing (e.g. Neely *et al*. 2020), store-and-forward technologies requiring low

bandwidth where video recordings from live events and historical data are shared via a secured platform (e.g. Smith *et al*. 2017), and remote patient monitoring where a person's physiological changes are tracked and shared with a professional (Taj-Eldin *et al*. 2018). E-learning platforms and Learning Management Systems (LMS) such as Moodle, Blackboard, and Adobe Authorware have been used for independent distance learning to provide information and training to families (Meadan and Daczewitz 2015).

Telehealth encompass both high-tech and low-tech formats. However, a minimum standard of connectivity, hardware, and software is usually needed to access any such technologies. This includes, but is not limited to, a minimum download and upload speed of 1Mpbs, a laptop or desktop computer with 4 GB of RAM, and antivirus software (Lee *et al*. 2015).

Validated telehealth practices

Telehealth practices can allow for increased access to services, reduced travel time for overworked professionals and overstressed families, more cost-effective services, and provide professionals with information about how teaching has extended to the natural environment (LeBlanc *et al*. 2020). As a result, the use of telehealth practices for delivering services to children with neurodevelopmental disabilities and their families has increased (Tsami *et al*. 2019). A literature search conducted by the Behavior Change Institute revealed over 80 telehealth studies that were published since 1997 (Behavior Change Institute, 2020). A comprehensive review conducted by McLay *et al*. (2020) found that some early empirical support exists for using telehealth services to deliver interventions to children and adolescents with sleep difficulties. Additional article reviews were conducted by Akemoglu *et al*. (2020) and Unholz-Bowden *et al*. (2020) which found over 30 studies looking at using telehealth practices to train parents. Further empirical validation for behavioral telehealth exists for practices focused on training direct care staff (e.g. Rios *et al*. 2020) and supporting and coaching educators (e.g. Rosenberg *et al*. 2020). In a recent randomized controlled trial conducted by Lindgren and colleagues (2020), it was demonstrated that parents who received telehealth services teaching them to use functional communication training with their children saw much greater improvements than their control group counterparts who received treatment as usual. The value of telehealth services has been demonstrated outside the field of ABA, too. Speech-language pathology services (Molini-Avejonas *et al*. 2015) and occupational therapy services (Cason 2014) have been successfully provided via teleheath. Overall, evidence-based practices have been made more accessible to parents and schools because of telehealth (Chung *et al*. 2020). Moreover, as discussed by Unholz-Bowden et al. (2020), research shows that parents receiving telehealth services provide high ratings of treatment acceptability. Most recently, considering the COVID-19 pandemic, the focus on medical care has shifted to the telehealth model prompting extensive publications and reviews which discuss ongoing research and development of this service delivery modality (e.g. Rodriguez 2020). The pandemic has also resulted in funding for ABA services via a distance model, which was previously unpermitted in many places (Council of Autism Service Providers 2020).

However, when it comes to telehealth practices in Europe, movement forward has been slower. Raposo (2016) discussed the challenges with establishing telemedical practices in Europe being the result of a missing comprehensive legal framework around services. Some steps have been taken to support telepractice in the region, though. For example, training courses on telemedicine have been incorporated into the health practitioner education in Switzerland (Brockes *et al*. 2017) and professionals have pushed for the use and regulation of telemedicine apps across Europe (Crico *et al*. 2018). Moreover, it was recently presented that some of the European nations which are part of the Organisation for Economic Co-operation and Development (OECD), such as Belgium, Denmark, and Poland, are working towards building public funding and legislation around telehealth (Hashiguchi 2020). With another review being generated because of COVID-19 showing the European nations of Italy and Spain taking increased steps towards telehealth transitions (Bhaskar *et al*., 2020).

Europe and behavioral services

Despite the small steps being taken in some European nations towards telehealth applications, there appears to be little formal alignment between this modality and behavioral services. To understand this lag, it is necessary to consider the context of behavior analysis in the region. In most European countries behavior analysis is not a recognized, or resultantly publicly funded, profession (Kelly *et al*. 2019). Moreover, in places across the continent, like Germany for example, there has been funding for only eclectic approaches as opposed to ABA-based services (Dillenburger 2011). Europe has not seen legislation surrounding behavior analysis services and their integration into the healthcare system (Keenan *et al*. 2010). In European nations, like Italy, where ABA appears to be offered as a service for individuals with autism, its prevalence of usage is less than one third and there are not mandates surrounding its application (Borgi *et al*. 2019). In the Czech Republic, for example, behavioral services have never been widely accessible due to the lack of skilled professionals and financial constraints for the families (Kingsdorf and Pančocha 2020). In another recent study of southeastern European nations less than one half of the survey respondents were presently receiving

behavioral services for their children with autism with less than 10% accessing these services from a behaviorist (Daniels *et al*. 2017). This aligns with the previous work of Salomone *et al*. 2016 who surveyed 18 European countries to find that behavior analytic services for children with autism were not the norm. This minimal use of behavioral services in Europe is likely the result of missing legislation, funding guidelines, availability of professionals, and misinformation (Gillen and Keenan 2018). These concerns have been historically echoed when discussing the plight of children and families impacted by neurodevelopmental disabilities on the continent (e.g. Hughes and Shook 2007; Keenan *et al*. 2015; Roll-Pettersson *et al*. 2020).

Given the overwhelming research which supports that evidence-based practices for children with autism are based on the science of ABA (Grigorenko *et al*. 2018), it is not surprising that in the United States these treatments have become the standard of practice. In accordance with the recommendations of the American Academy of Pediatrics (Myers and Johnson 2007), U.S. Surgeon General (U.S. Surgeon General, 2001), and others, the accessibility of services has increased throughout the nation. As a result, funding for ABA services has steadily grown over the past decades, to include access via insurance, state or federal funds, public education, and voucher or scholarship programs (Harvey *et al*. 2010). While there is a recognized challenge, even in the U.S., with placing the funding responsibility for ABA services on the shoulders of education or medicine (Pollak 2016), guidelines have still been developed to help provide access to care. These regulations have helped to later establish a framework for increasing the availability of services to those in more rural or underserved regions. The process of evaluating the context of need and the development of, or transition to, telehealth has been made explicit by several governmental agencies like the Center for Disease Control (CDC) (National Center for Immunization and Respiratory Diseases (NCIRD) and Division of Viral Diseases 2020), telehealth task forces like the Council of Autism Service Providers (CASP) (e.g. Council of Autism Service Providers 2020), and professional journals (e.g. Rodriguez 2020).

As made evident earlier, more needs to be done in Europe to support the needs of families impacted by neurodevelopmental disabilities. There are projects underway to drive the allocation of funding and development of practitioner training to support behavioral services in the region (Roll-Pettersson *et al*. 2020). However, to date, little has been done to integrate telehealth services into any European model of care for behavioral services. Perhaps better understanding the telehealth system in behavioral care, its various applications throughout Europe, and the local context into which it can be applicable will support system growth and better serve families in need.

Research aim

Countless telehealth studies have been conducted looking at the efficacy of delivering intervention services to children and training staff and parents via this methodology. However, it appears that there is less empirical support for the practice in European countries, which may be responsible for its slow growth. To inform the development of better practices and guidelines for integrated models of care for telehealth services in Europe, a review of telehealth practices for providing services to children and families impacted by neurodevelopmental disabilities in Europe was undertaken. Looking at the literature aimed to answer these questions:

1. What type of studies on providing telehealth services to families of children with neurodevelopmental disabilities in Europe have been conducted in the last five years?
2. What were the main behaviors targeted for change and intervention strategies used?
3. What were the main telehealth components?
4. What barriers were faced?
5. What future work needs to be done to support children and families impacted by neurodevelopmental disabilities in Europe looking to access behavioral services via a telehealth model?

Method

Numerous literature reviews of telehealth have been conducted in recent years (e.g. Ferguson *et al*. 2019; Meadan and Daczewitz 2015; Neely *et al*. 2021; Tomlinson *et al*. 2018). As a result, this literature search was targeted. A scoping review of the literature was conducted. This method was chosen given the relatively limited nature of the topic. It was anticipated that too few studies would be found to enable a quantitative analysis. As a result, this more rapid method for summary was selected.

Eligibility criteria

Studies were included in the review if they reported on the use of a telehealth modality to deliver behavior-analytic interventions to support families impacted by neurodevelopmental disabilities in Europe. Only recent studies, those published since 2015, were included. Given the novelty of telehealth practices and behavior analytic services in the region, it was anticipated that searching prior to 2015 would not result in more studies. This decision was additionally supported by the reported mandate for widespread telemedicine services in Europe which was presented in 2014 (eHealth Stakeholder Group 2014). As well, studies needed to be published in the English language and involve caregivers in the telehealth model. There were no restrictions on the specific telehealth modality used or the age of the participants. However, studies were excluded in the review if they were review articles themselves,

Table 1 Search terms.

Diagnosis	Autism, neurodevelopmental disability, neuro-developmental, ASD, autism spectrum disorder, developmental disability.
Participants	Children, family, parent, caregiver, adolescent, child.
Mode of delivery	Telehealh, telepractice, remote service, synchronous, telemedicine, telecare, e health, video consult, electronic consult, e consult, virtual consult, remote consult, videoconference.
Location	Europe, EU, European Union, OECD, Albania, Andorra, Armenia, Austria, Azerbaijan, Belarus, Belgium, Bosnia, Herzegovina, Bulgaria, Croatia, Cyprus, Czech, Denmark, Estonia, Finland France, Georgia, Germany, Greece, Hungary, Iceland, Ireland, Italy, Kazakhstan, Kosovo, Latvia, Liechtenstein, Lithuania, Luxembourg, Malta, Moldova, Monaco, Montenegro, Netherlands, Macedonia, Norway, Poland, Portugal, Romania, Russia, Serbia, Slovakia, Slovenia, Spain, Sweden, Switzerland, Turkey, Ukraine, United Kingdom, England, Scotland.

"how-to" guides that did not evaluate the use of an intervention, used strategies that were not grounded in the science of behavior (e.g. music therapy, psychiatry), focused on only using telehealth to facilitate a diagnosis or follow-up care, or reported on survey data in the absence of intervention.

Information sources and search strategy

The Web of Science, Scopus, and PsycInfo databases were searched. Since the focus of this review was on intervention-based telehealth services provided to families impacted by neurodevelopmental disabilities in Europe a combination of terms and keywords relevant to the diagnosis (neurodevelopmental disability), participants, mode of delivery (telehealth), and location (Europe) were used (see Table 1 for the lists of keywords). Appendix 1 provides one of the detailed searches. Google Scholar was additionally used to search for titles from reference lists and previously conducted review studies. However, no new articles surfaced.

Selections of sources of evidence

Search results, after being filtered for the language and journal article requirements, if needed, then underwent title review. After title review the remaining articles were combined and duplicates were removed. After the subsequent abstract screening, very few articles remained. During the title review and abstract screenings, the authors looked for the key words as well as indications that the articles involved training caregivers and using behavior analysis. Those left underwent full-text review. Permanent products were generated by the first author of all searches and screenings at each level and for each database. The permanent products were shared with the second author for review and confirmation.

Data extraction and synthesis

A form for data extraction was created based on the summary of characteristics charts provided in published telehealth literature reviews (e.g. Ferguson *et al.* 2019; Unholz-Bowden *et al*. 2020), and the form used by the authors in a previous literature review study (Pančocha and Kingsdorf 2021). Data extracted included first author and year of publication, and then the 15 targeted components of: (1) location, (2) strategies used via telehealth, (3) child dependent variables (behaviors targeted for change), (4) caregiver dependent variables (behaviors targeted for change), (5) study design, (6) participant characteristics of children, (7) participant characteristics of caregivers, (8) asynchronous telehealth components used, (9) synchronous telehealth components used, (10) technology components used, (11) use of an *in situ* (in-person) component, (12) whether or not cultural competence was demonstrated (i.e. the intervention was driven by local norms or values), (13) children's outcomes, (14) caregivers' outcomes, and (15) social validity outcomes. The data extraction was conducted by the first author and checked by the second author for 33% of the studies, a total of 30 targeted components. The agreement percentage for the coding of data extraction was compared for each targeted component and was calculated by dividing agreements by agreements plus disagreements and multiplying by 100. The intercoder agreement was 100%.

Three tables were created to summarize the components. Table 2 displays the technical components of the studies including the location, dependent variables, design, and participant characteristics. Table 3 displays the training strategies and telehealth components. Table 4 displays the various outcomes for the participants, the assessment of cultural competence, and social validity. All the components are also discussed qualitatively below.

Results

Selection of sources of evidence

A total of six publications were included in the review. The decision was ultimately made to include the one relevant thesis found, given the very limited amount of research on this topic. A PRISMA flow diagram was created using the PRISMA Flow Diagram Generator tool (http://prisma.thetacollaborative.ca/). The diagram in Figure 1 shows the screening and retrieval process throughout the literature search.

Table 2 Technical components of the studies.

Article	Location	Design	Child Participants	Caregiver Participants	Child Behavior Targets	Caregiver Behavior Targets
Dai *et al*. 2018	Albania	Treatment and control group comparison.	29 children with autism aged 18–70 months.	29 mothers.	None.	Scores on knowledge quizzes and the Early Intervention Parenting Self-Efficacy Scale.
Griffiths 2020	Albania	Non-concurrent multiple baseline design (MBD) across participants.	Three children with autism aged 4-8 years.	Three mothers.	Mands (requests).	Implementation of a mand training procedure as assessed via a fidelity checklist.
Guðmundsdóttir *et al*. 2017	Iceland	MBD across people for one participant and across settings for the other.	Two children with autism aged 4-5 years.	Two families, mothers were the main participants.	Request or social attending.	Teaching episodes which included correct implementation of the taught DANCE procedures.
Guðmundsdóttir *et al*. 2019	Iceland	MBD across child behaviors replicated across parents.	Three children with autism aged 3-4 years.	Three families, mothers were the main participants.	Communication response.	Teaching episodes which included correct implementation of the taught DANCE procedures.
Marino *et al*. 2020	Italy	Randomized control trial. Random assignment 2 group comparison (training with or without telehealth component).	23 children with autism aged 30 months to 10 years.	42 parents completed the study, 23 mothers and 19 fathers.	None.	Pre- and post-training assessments using the Home Situation Questionnaire (HSQ-ASD) and the Parental Stress Index (PSI/SF).
Tsami *et al*. 2019	Greece, Turkey, Russia, Ukraine, Mexico, Saudi Arabia, Costa Rica, USA.	MBD across participants (parent and child dyads).	12 children with autism aged 3-13 years.	14 parents completed the study, 12 mothers, 1 brother, and 1 father.	Challenging behaviors (e.g. aggression, self-injury, property destruction, etc..) and mands.	Implementation of functional assessment and functional communication training procedures.

Table 3 Training and telehealth components of the studies.

Article	Strategies used via Telehealth	Asynchronous Components	Synchronous Components	Telehealth Technology	In-situ Component
Dai *et al*. 2018	Independent learning activities using videos, vignettes, manuals, and tests.	Video recordings of the teaching material.	Telephone interviews to answer questions about the teaching material presented in the recordings.	DVD player, recordings, telephone.	Yes
Griffiths 2020	All 4 components of the Behavioral Skills Training (BST) procedure (instructions, modelling, rehearsal, feedback).	File sharing of the teaching materials and videos for clinician assessment.	Video conferencing for BST training.	Computer or smartphone, Skype, Zoom, and Dropbox	No
Guðmundsdóttir *et al*. 2017	Training package using explanation, modelling, positive and corrective feedback, written summary of teaching procedures, and reviewing data with the caregivers.	Sessions were recorded and later watched and scored by a clinician.	Video conferencing for trainings and assessments.	Laptops, desktop, webcams, Skype, closed file system, encrypted website, video camera.	Yes
Guðmundsdóttir *et al*. 2019	Training package using explanation, modelling, positive and corrective feedback, written summary of teaching procedures, and reviewing data and self-monitoring videos with the caregivers.	Sessions were recorded and later watched and scored by a clinician.	Video conferencing for trainings and assessments.	Laptops, webcams, Skype, closed file system, encrypted website.	Yes
Marino *et al*. 2020	Coaching as individualized ABA therapy, engaging parents in behavioral interactive play activities with their young children.	None.	Video conferencing for one-to-one behavioral parent training and coaching.	Web platform within G-Suite, video conference tools.	Yes
Tsami *et al*. 2019	Coaching, prompting, prompt fading, explanation.	Sessions were recorded and later watched and scored by a clinician.	Video conferencing for trainings and assessments.	Laptop, desktop, phone, iPad, Vidyo (video conference software).	No

Characteristics of Sources of Evidence

Study location and design

The goal of this study was to evaluate research that was conducted in Europe. Despite aiming to inclusively search all countries in Europe, the only European countries with results were Iceland (Guðmundsdóttir *et al*. 2017; Guðmundsdóttir *et al*. 2019), Greece, Turkey, Russia, Ukraine (Tsami *et al*. 2019), Albania (Dai *et al*. 2018; Griffiths 2020), and Italy (Marino *et al*. 2020). Four of the studies (67%) used single subject experimental designs, all versions of a multiple baseline design (Griffiths 2020; Guðmundsdóttir *et al*. 2017; Guðmundsdóttir *et al*. 2019; Tsami *et al*. 2019). Two of the studies (33%) used group designs: randomized controlled trial and a group comparison (Dai *et al*. 2018; Marino *et al*. 2020).

Participants

A total of 72 children completed participation across the six studies. Despite making the search criteria inclusive of all neurodevelopmental disabilities, all the children in the studies had diagnoses of autism. The children ranged in age from 18 months to 13 years old. A total of 95 parents completed participation in the studies. Of those 95 participants, 23 identified as male (22 fathers and one brother) and 72 identified as female

Table 4 Outcome components of the studies.

Article	Cultural Competence	Child Outcomes	Caregiver Outcomes	Social Validity Outcomes
Dai *et al*. 2018	Materials created with parent input and in native language. Local clinicians with US collaboration. Most videos locally created.	None.	Small improvement in the telehealth treatment group over control group for the test scores and Early Intervention Parenting Self-Efficacy Scale.	Caregivers rated the social validity of the information in each training module. Ratings were on average over 4 (5-point scale).
Griffiths 2020	Materials and training provided in native language but from the US. Intervention goals were created in collaboration with the parents.	All children's mands increased.	Fidelity of their implementation of the mand training procedure increased.	The social validity questionnaire data collected were an average of 4.7 or higher (5-point scale).
Guðmundsdóttir *et al*. 2017	One parent was taught in her native language and one parent was not. Intervention goals were created in collaboration with the parents.	All children's skills increased.	Parent delivery of teaching episodes increased.	None.
Guðmundsdóttir *et al*. 2019	All parents were taught in their native language. Intervention goals were created in collaboration with the parents.	All children's skills increased.	Parent delivery of teaching episodes increased.	Full information on social validity assessment procedures not provided in this article. Only positive anecdotal social validity reports discussed.
Marino *et al*. 2020	No information provided.	None	Parents in the telehealth group had statistically significant positive effects over the control group on their reported stress levels and the perceptions of their children's behavior as reported in the HSQ-ASD and PSI/SF.	None.
Tsami *et al*. 2019	Trainings were provided in either the native language or with a trained interpreter. Intervention goals were created in collaboration with the parents.	All children's problem behaviors decreased and mands increased.	Fidelity of the FCT and FA procedures for all but one parent had at least 90% accuracy across sessions.	Social validity means via Treatment Acceptability Rating Form were high.

(all mothers). The Tsami *et al*. (2019) study had two male participants, one father and one brother. However, the father and the brother in that study only participated in one generalization probe and were therefore not trained in the intervention. Two fathers participated in the Guðmundsdóttir *et al*. (2017) study, but only for the assessment and goal setting process, not the training. The remaining male participants were part of the Marino *et al*. (2020) study; and were fathers that participated fully in the intervention. Therefore, in five out of the six studies (83%) the main parental participants were mothers.

Dependent variables

A mix of direct and indirect assessments of intervention effectiveness was used across the six studies. Four of the studies (67%) included measures of both the caregiver and the child behaviors (Griffiths 2020; Guðmundsdóttir *et al*. 2017; Guðmundsdóttir *et al*. 2019; Tsami *et al*. 2019). All four studies (67%) measured the children's communication responses (e.g. requests or mands). Tsami *et al*. (2019) additionally measured the challenging behaviors of the children (e.g. aggression). Five of the studies (83%) collected direct assessment data on the behaviors of the caregivers (Dai *et al*. 2018; Griffiths 2020; Guðmundsdóttir *et al*. 2017; Guðmundsdóttir *et al*. 2019; Tsami *et al*. 2019). A fidelity checklist was used to assess the caregivers' applications of the mand protocol in the Griffiths study (2020). Treatment fidelity data were also collected in the Tsami *et al*. (2019) study on the caregivers' procedural fidelity when implementing the functional assessment (FA) and functional communication training (FCT) procedures. The Guðmundsdóttir *et al*., studies (2017; 2019) collected caregiver data on the number of teaching episodes that they delivered to their children.

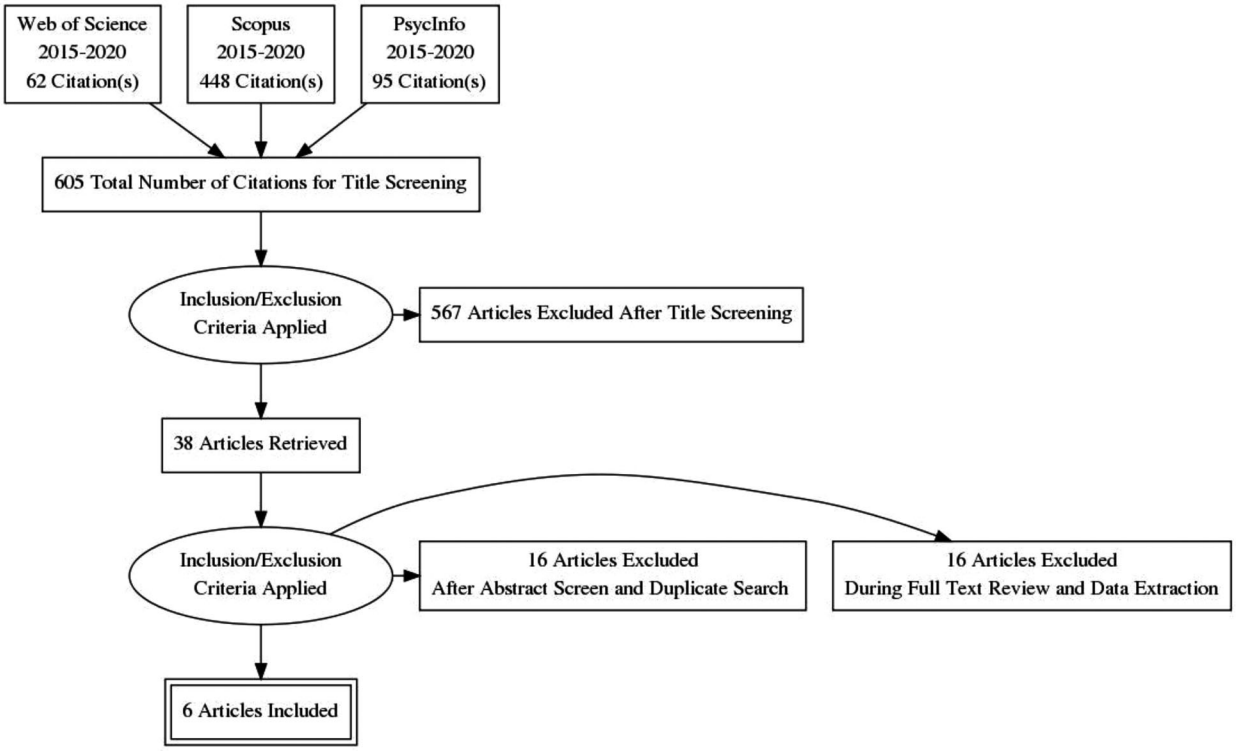

Figure 1. Screening and retrieval process.

In the Dai *et al*. (2018) study, caregiver data were collected on pretest and posttest knowledge assessments. That study additionally collected indirect data on the caregiver-completed assessment tool of the Early Intervention Parenting Self-Efficacy Scale. Similar self-assessment scales were used in the Marino *et al.*, (2020) study: The Home Situation Questionnaire (HSQ-ASD) and the Parental Stress Index (PSI/SF).

Strategies used during telehealth

This article review centered on assessing family-focused behavioral interventions which involved a component delivered via a telehealth modality. Therefore, telehealth was not the intervention in these studies, but rather, a mode for delivering at least a portion of education, training, or intervention to families. The telehealth components used several different strategies. In four of the studies (67%) it was made clear that the parents were taught specific intervention strategies/teaching protocols such as the use of naturalistic teaching methods (Guðmundsdóttir *et al*. 2017; Guðmundsdóttir *et al*. 2019), FA and FCT procedures (Tsami *et al*. 2019), and mand (request) training procedures (Griffiths 2020). In these studies, ABA-based training techniques were used to train the caregivers. In the Guðmundsdóttir *et al.*, studies (2017; 2019) a specific methodology for training the parents was not stated. However, they appeared to use components of behavioral skills training by incorporating explanation, modelling, positive and corrective feedback, and a written summary of teaching procedures. The Guðmundsdóttir *et al.*, studies (2017; 2019) also included a data review component with the parents during the video conferences including a self-monitoring video review of the parent behaviors in the 2019 study. The Griffiths study (2020) explicitly stated the usage of behavioral skills training, outlining the use of all four steps of instructions, modelling, rehearsal, and feedback. The Tsami *et al.*, study (2019) provided procedural explanations to the parents and then relied on coaching with prompting and prompt fading. In the Marino *et al.*, study (2020) the telehealth component involved coaching parents during individual ABA therapy sessions with their child. The targets or procedures were not made clear but were said to involve assisting parents in interactive play activities with their children. The study by Dai and colleagues (2018) used telehealth to deliver only parent education. The education procedures included the independent learning activities of watching videos, completing activities, vignettes, manuals, and tests.

Telehealth modality and technology

All the studies used a telehealth modality in some way. However, four of the studies (67%) did some in-situ initial assessment or follow-up (Dai *et al*. 2018; Guðmundsdóttir *et al*. 2017; Guðmundsdóttir *et al*. 2019; Marino *et al*. 2020). Regardless, telehealth was the main method of intervention delivery.

Five of the six studies (83%) used both asynchronous and synchronous modalities (Dai *et al*. 2018; Griffiths 2020; Guðmundsdóttir *et al*. 2017; Guðmundsdóttir *et al*. 2019; Tsami *et al*. 2019). All the

studies used synchronous modalities. These included combinations of real-time video conferencing and telephone calls; with most of the studies using real-time video conferencing (Griffiths 2020; Guðmundsdóttir *et al*. 2017; Guðmundsdóttir *et al*. 2019; Marino *et al*. 2020; Tsami *et al*. 2019). The most used asynchronous modality was the use of video recordings, specifically videos sent by the caregivers for the clinicians to review and vice versa (Dai *et al*. 2018; Griffiths 2020; Guðmundsdóttir *et al*. 2017; Guðmundsdóttir *et al*. 2019; Tsami *et al*. 2019). In the Guðmundsdóttir *et al*. (2017; 2019) and the Tsami *et al*. (2019) studies, though, the teleconference sessions were recorded, and the videos were later watched and scored by the clinicians. Several different hardware components were utilized. Combinations of laptops, desktop computers, smartphones, tablets, and webcams were needed for family participation in five out of the six studies (83%) (Griffiths 2020; Guðmundsdóttir *et al*. 2017; Guðmundsdóttir *et al*. 2019; Marino *et al*. 2020; Tsami *et al*. 2019). The one exception was the Dai *et al*. (2018) study that provided DVDs for the parents to review and conducted audio-only interviews. Video-conferencing software included commonly known tools like Skype, Zoom, Vidyo, and Google platforms. Asynchronous file-sharing was done using Dropbox, the Google platform, and proprietary encrypted systems.

Cultural competence

Since a goal of research, especially research which involves working with families, should be respecting the cultural values and norms of research participants (Bal and Trainor 2016), the decision was made to do a cursory assessment of the cultural competence displayed in the studies. Based on previous evaluations of cultural competence in parent training research (Pančocha and Kingsdorf 2021), the following components were combined to assess this variable: match between participant native language and information delivery, the development of the intervention with input from the participants and using interventionists that were part of the local community. These components were selected based on the limited cultural information provided in most of the studies. Only the Dai *et al*. (2018) study explicitly appeared to meet all these criteria. The Guðmundsdóttir *et al*. (2017; 2019) studies appeared to teach in the local language, develop intervention goals with the input of the families and possibly use an interventionist from the local community. However, the last component, the location of the interventionist in relation to the study participants, was not made clear in either article. It should also be noted, that the Guðmundsdóttir *et al*. (2017) study stated that one of the parental participants was not trained in her native language, but still fluent in the language of instruction. The Tsami *et al*. (2019) study either provided training in the native language or used translators, and incorporated families into the intervention development. However, it did not appear that the interventionists were currently part of the local community. The Griffiths study (2020) appeared to provide the intervention in the local language and did translate needed materials. The interventionist did not appear to currently be part of the local community. However, it appeared that the items to target for mand training for each child were selected by the parents. The Marino *et al*. (2020) study did not provide any information regarding these criteria.

Outcomes

All the studies reported favorable outcome measures. Both Guðmundsdóttir *et al*. (2017; 2019) studies demonstrated that parents' skills of using a naturalistic teaching intervention and their children's communication skills increased as assessed via either visual analysis or percentage of nonoverlapping data (PND). The Tsami *et al*. (2019) study demonstrated high treatment integrity data (90% or higher). Child problem behaviors (e.g. aggression) decreased and mands (requests) also increased as assessed via visual analysis. The Marino *et al*. (2020) study conducted numerous statistical tests comparing the telehealth and control groups. Overall, there was a statistically significant positive effect of the telehealth intervention in terms of parents' stress levels, perception of the disruptive and noncompliant behavior of their children, coping with their children in cases of inadequate behaviors, as well as in the influence on their children's behavior. The Griffiths (2020) study demonstrated via visual analysis increased fidelity in parents' use of mand (request) teaching procedures and increases in child mands from baseline to intervention for all dyads. The Dai *et al*. (2018) study had the least favorable outcomes with the repeated measures ANOVA comparing the telehealth and control groups and showing only minimal differences.

In addition to the above measures, specific social validity or caregiver rating scales of intervention effectiveness and acceptability were targeted in three of the six studies (50%) (Dai *et al*. 2018; Griffiths 2020; Tsami *et al*. 2019). The Dai *et al*. (2018) and Griffiths (2020) studies both used a 5-point Likert-scale questionnaire to assess parent agreement with statements regarding the intervention and had average agreement ratings over 4. The Tsami *et al*. (2019) study used a similar tool, a 7-point Likert scale where higher scores also indicated a higher level of acceptability and had over 80% of the statements receive average agreement ratings of 5 of higher. The Guðmundsdóttir *et al.*, studies (2017; 2019) did not describe social validity assessment procedures. However, the 2019 study did mention that there were positive anecdotal social validity reports. This was like the Marino *et al*. 2020 study,

which did not appear to collect social validity data but did mention anecdotal reports from parents on the value of the training.

Overall evaluation

Specific steps were not taken to evaluate the quality of the studies included in this review, or the overall effectiveness of telehealth interventions for families impacted with autism in Europe. These decisions were made given the small amount of research on this topic. However, certain conclusions can be drawn from looking at this small battery of research. Only one of the studies (17%) used the gold standard of research design: a randomized controlled trial (Marino *et al*. 2020). All the studies had relatively small sample sizes. The single subject experimental design studies did use control measures to demonstrate experimental control by using iterations of the multiple baseline design (Griffiths 2020; Guðmundsdóttir *et al*. 2017; Guðmundsdóttir *et al*. 2019; Tsami *et al*. 2019). However, again, the limited number of participants in those studies as well as the Dai *et al*. (2018) study makes the generalizability of the research questionable. While interobserver agreement data were collected in all the studies which used single subject designs, the amount and quality of data collected were not enough for validation in some cases (Griffiths 2020; Guðmundsdóttir *et al*. 2017; Guðmundsdóttir *et al*. 2019). Also, while most of the articles were peer-reviewed, the inclusion of a thesis (Griffiths 2020) should be considered when evaluating research quality.

Barriers in application of telehealth

Several barriers were noted in the research reviewed. In the Guðmundsdóttir *et al*. (2017; 2019), Tsami *et al*. (2019), and Dai *et al*. (2018) studies, high-speed internet access was a challenge for participants. Therefore, the technology usage was kept simple. As a result, in the Dai *et al*. (2018) study the decision was made to use DVD recordings that were mailed to the families and telephone calls occurred without video. The level of telehealth utilized in the study could be questioned, though. The impact of connectivity was specifically assessed after each video-conferencing session in the Tsami *et al*. (2019) study, but still impacted several of the families' abilities to remain in the study. Attrition was a pervasive barrier across all studies. The Dai *et al*. (2018), Marino *et al*. (2020), and Tsami *et al*. (2019) studies all specifically discussed large losses of participants during their research. Another issue was that most of the studies seemingly struggled with recruiting male caregivers, specifically fathers, to actively participate. This could be a barrier to providing the most generalizable outcomes in the home setting. Lastly, most of the studies noted that there was collaboration with professionals based in North America to support their work. While collaboration is valuable, its persistent need throughout the studies highlights Europe's barrier of access to high quality information and services grounded in the science of behavior and its telehealth modality.

Discussion

Research questions

The goal of this research was to assess the current literature base on behavioral telehealth services for children with neurodevelopmental disabilities in Europe. This review of studies found that a limited number of current works exist in this area. However, of the studies available, Iceland and Albania were the most targeted locations. Children with autism and their mothers were typically the participants. The intervention strategies for training caregivers to implement procedures more often, or with fidelity, frequently used components of behavioral skills training or coaching, to increase their children's communication responses. Single-subject methodology was used most to assess increases in these measurable behaviors. Video conferencing was the most used telehealth component and had a high level of acceptability. Overall, outcome data, when collected on children and caregiver behaviors were positive. However, several barriers and opportunities for future research exist.

Limitations

A few key limitations exist in this review. One of the main considerations is that only studies in the English language were targeted. However, in most of the countries targeted within Europe the main language spoken is not English. Therefore, studies published in local-language journals were likely missed. While it is important that research is made available in local languages to local communities, also having research available in the global language of English supports the goals of widespread healthcare access for diverse populations throughout the European Union (EU) region (Ledoux *et al*. 2018). With European citizens generally being covered for a broad range of healthcare services within and between countries in the EU, it stands to reason that similar services should be available (Busse *et al*. 2011). Efforts to build a more unified system of telehealth care in the region are likely to be influenced by research conducted within the area and accessible to everyone via a common language.

An additional limitation of this review may be the narrow date range used for the search. With only very recent publications targeted for this review important outcomes of previous studies may have been overlooked. However, this decision was made based on the influx of recent reviews on the broader topic of telehealth as well as the constantly evolving world of technology. This is to say, that older studies may have already been discussed in review articles and may no

longer hold relevance in the face of the rapidly changing digital world.

Future research

As discussed by Rispoli and Machalicek (2020), there is a body of quality single-subject research looking at the use of telehealth practices in the behavioral sciences. However, the field is only emerging, needs more empirical validation, and should support ongoing evaluation of constantly advancing technology. In the case of Europe specifically, there appears to be even less research in this arena. While active telepractice programs have roots in Europe, such as the University of Tromso in Norway's teleconsultation department (Perednia and Allen 1995), Italy's history of experimental telemedicine (Pisanelli *et al*. 1995), and the Balkans' 18-year-old telemedicine center (Latifi 2012), the use of a remote model for behavioral services appears scarce. Only six studies were found during a search of the relevant recent literature. Therefore, there is plenty of room for future research.

When it comes to addressing some of the reoccurring barriers found here, future work might consider looking at creating incentive programs to sustain families' participation in telehealth services. Research suggests that incentives can increase the participation of individuals in studies even when commitment requirements are difficult (Guyll *et al*. 2003). However, another route to consider when addressing this same barrier is doing an initial assessment of how much time and effort families can contribute and having plans in place to shape parent participation throughout the study (e.g. systematically increase the amount of time committed each week). Shaping procedures have been used to increase the participation behaviors of individuals in different contexts (e.g. Athens *et al*. 2007). The specific issue of male caregiver involvement in parent training interventions is something that persists across the literature (Fabiano 2007). Some strategies that should be incorporated into future intervention-based telehealth research are treating fathers as co-parents, increasing fathers' knowledge and awareness of the intervention participation possibility, training researchers/clinicians to facilitate father-inclusive practices, and upholding high expectations of fathers at the start of the intervention (Lechowicz *et al*. 2019). Digital inequality, as lack of access to reliable high-speed internet, needed hardware, and resultantly information, is an issue that has historically hindered more rural and disadvantaged populations (McLaren and Zappala 2002). Future work in behavioral interventions that are delivered to families via telehealth should continue to take into consideration the bandwidth needed to access certain technological modalities, alternative ways to access information in the case of internet malfunctions, and even technology confidence prior to intervention (Salomone and Maurizio Arduino 2017).

In addition to the barriers that were noted, other areas of concern were raised that could be addressed in the future. For one, the study quality needs to be more rigorous to build a solid foundation of behavioral telehealth in Europe. Future work should consider increasing the number of study participants, increasing data collection methods to include more outcome measures focusing on direct data collection, extended methods to provide data validation, and increased assessments of social validity to see the impact of the intervention on not just perceived effectiveness but later applications of skills after training. Similar recommendations were echoed in previous reviews (e.g. Akemoglu et al. 2020). The area of cultural competence, although looked at only minimally here, is another area for future consideration. Upcoming telehealth interventions should work to form partnerships with their study participants and community; including them in the planning to support intervention buy-in and recruiting local interventionists to support sustainability of the outcomes. Increased reports of these dimensions (i.e. richer descriptions of participant and clinician characteristics, settings, and intervention development) in the research will also help support the external validity.

Lastly, an area that was not looked at in any of these studies, but might be valuable to explore in the future, is the impact of the telehealth model on clinicians. With the noted issues of access to qualified behavior analysts in large parts of Europe, such as in the Czech Republic (e.g. Kingsdorf and Pančocha 2020), those currently working in the field are often spread thin and at increased risk of burnout (Plantiveau *et al*. 2018). Creating more widespread and validated models of behavioral telehealth can help ease some of the burden. Research could investigate changes in the number of clients seen over a period, the number of cases held, the differences in intensity of services (e.g. visits/sessions per week) across models, and perhaps even an evaluation of cost-effectiveness.

Conclusions

Overall, the impact of future telehealth research in Europe might not only facilitate the growth of the modality, but also bolster support for the science of behavior analysis. As discussed earlier, in Europe behavior analysis is fighting a war against misinformation and unvalidated treatment practices. Embracing a telehealth model could increase access to care, improve family outcomes, and foster dissemination. The modality multiplies the reach of the few qualified practitioners who are working on the ground providing traditional in-person services. If created systematically and sustainably their impact can be spread across greater distances. Establishing research-driven behavioral telehealth guidelines on the continent could be the

catalyst for future legislation surrounding comprehensive access to ABA-based services.

It appears that telehealth is a viable option for delivering behavior analytic support to families impacted by neurodevelopmental disabilities in Europe. While there is limited research in this region, what is available seems to align with what has been found in North America. Our conclusions here echo that there is a high level of parental acceptability for behavioral telehealth (Unholz-Bowden *et al.*, 2020), that providing services even in the face of low-speed internet conditions is possible with the right practices (Oberleitner *et al*. 2004), and that behavioral improvements are possible for both caregivers and children (Akemoglu *et al*. 2020). The match between the conclusions of our study and the works of others in North America, as well as additional work that has demonstrated the ability of telehealth applications to be culturally adapted to the broader world context (Sivaraman and Fahmie 2020), bodes well for the future of behavioral telehealth in Europe. The need, empirical validation, and groundwork for sustainability already seem to exist- so now the work on policy, procedures, and future research needs to happen.

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Appendix 1.

The search strategy used in the web of science database

Set	Results	Save History / Create Alert · Open Saved History	Edit Sets	Combine Sets (AND / OR) Combine	Delete Sets Select All / Delete
# 9	45	#7 AND #3 AND #2 AND #1 **Refined by: DOCUMENT TYPES:** (ARTICLE) *Indexes=SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC Timespan=2015-2020*		☐	☐
# 8	62	#7 AND #3 AND #2 AND #1 *Indexes=SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC Timespan=2015-2020*	Edit	☐	☐
# 7	7,285,028	#6 OR #5 OR #4 *Indexes=SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC Timespan=2015-2020*	Edit	☐	☐
# 6	4,518,710	**ALL FIELDS:** (Lithuania OR Luxembourg OR Malta OR Moldova OR Monaco OR Montenegro OR Netherlands OR Macedonia OR Norway OR Poland OR Portugal OR Romania OR Russia OR Serbia OR Slovakia OR Slovenia OR Spain OR Sweden OR Switzerland OR Turkey OR Ukraine OR United Kingdom OR England OR Scotland) *Indexes=SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC Timespan=2015-2020*	Edit	☐	☐
# 5	3,873,698	**ALL FIELDS:** (Albania OR Andorra OR Armenia OR Austria OR Azerbaijan OR Belarus OR Belgium OR Bosnia OR Herzegovina OR Bulgaria OR Croatia OR Cyprus OR Czech OR Denmark OR Estonia OR Finland OR France OR Georgia OR Germany OR Greece OR Hungary OR Iceland OR Ireland OR Italy OR Kazakhstan OR Kosovo OR Latvia OR Liechtenstein) *Indexes=SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC Timespan=2015-2020*	Edit	☐	☐
# 4	622,984	**ALL FIELDS:** (Europe OR EU OR "European Union" OR OCED) *Indexes=SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC Timespan=2015-2020*	Edit	☐	☐
# 3	69,387	**ALL FIELDS:** (Telehealth OR telepractice OR "remote service" OR "synchronous" OR "telemedicine" OR "telecare" OR "e health" OR "video consult" OR "electronic consult" OR "e consult" OR "virtual consult" OR "remote consult" OR "videoconference") *Indexes=SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC Timespan=2015-2020*	Edit	☐	☐
# 2	1,449,494	**ALL FIELDS:** (Children OR family OR parent OR caregiver OR adolescent OR child) *Indexes=SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC Timespan=2015-2020*	Edit	☐	☐
# 1	50,139	**ALL FIELDS:** (Autism OR "neurodevelopmental disability" OR neuro-developmental OR ASD OR "autism spectrum disorder" OR "developmental disability") *Indexes=SCI-EXPANDED, SSCI, A&HCI, CPCI-S, CPCI-SSH, BKCI-S, BKCI-SSH, ESCI, CCR-EXPANDED, IC Timespan=2015-2020*	Edit	☐	☐
				AND / OR Combine	Select All / Delete