

# Journal on Developmental Disabilities Le journal sur les handicaps du développement

# JDD



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General

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## ***Welcome to the Editorial Board of the Journal On Developmental Disabilities***

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The Journal is pleased to welcome three individuals who will contribute their diversity of experiences and talents to the Journal's Editorial Board. Yani Hamdani, PhD, OT (Reg.) (Ont.), Department of Occupational Science and Occupational Therapy, University of Toronto, and the Azrieli Centre for Adult Neurodevelopmental Disabilities and Mental Health, Centre for Addiction and Mental Health (CAMH), has joined the Chief Editorial Board. Andrea Maughan, MA, MSc, BSc, from York University, and Ami Tint, PhD, CPsych (Supervised Practice), Azrieli Centre for Adult Neurodevelopmental Disabilities and Mental Health, CAMH, have joined as Associate Editors.

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### **Research Special Interest Group Annual Meeting Abstracts**

Abstracts presented at the Research Special Interest Group (RSIG) Annual Meeting in 2014, 2015 and 2016 have been published in various issues of the JoDD (<https://oadd.org/publications/journal-on-developmental-disabilities/>). RSIG Abstracts for 2017, 2018 and for subsequent years will be posted in a special section on the OADD website.

### **Thank You to Reviewers of Articles Submitted to the Journal and of Articles Eligible for the 2017 Journal Student Publication Prize**

The Journal is highly indebted to volunteers who provide critical reviews of papers submitted to the Journal and ranking of papers which are eligible for the Annual Student Publication Prize competition. Thank you everyone, and to Dr. Shelley Watson for arranging a subcommittee of the Research Special Interest Group to rank papers eligible for the Student Publication Prize.

### **Individuals Who Provided Feedback Prior to 2017, and Were Not Previously Acknowledged**

Pamela Bryden	Bruce Oddson
Eva Niechweij-Szwedo	Joan Versnel

### **Individuals Who Provided Feedback During 2017 and 2018**

Michael Arthur-Kelly	Lea Ann Lowery	Duong (Young) Ramon
Marjorie Aunos	Lynn Martin	Mélina Rivard
Robert Balogh	Hélène Ouelette-Kuntz	Shahin Shooshtari
Madeline Burghardt	Ghislain Magerotte	Samantha Stronski
Tom Cariveau	Voula Marinos	Kendra Thomson
Kelly Coons	Frances Owen	Shannon Wagner
Peter Elson	Annie Paquet	Kerri Walters
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Rachelle Hole	Maire Percy	Kristen Wirth
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## In Memoriam: Dr. Thomas Cheetham



Our dear friend and colleague, Dr. Tom Cheetham, MD, Fellow of the American Association of Intellectual and Developmental Disabilities (FAAIDD), passed away August 20, 2018 in Nashville, TN after a battle with cancer.

Tom was passionate about the healthcare of individuals with intellectual and developmental disabilities (IDD). This passion began prior to his medical school training when he and his wife were live-in house parents for a group individuals with IDD in Hamilton. He completed his family medicine residency at Oxford Regional Centre in Woodstock and became the Chief Physician until the Centre closed. In 1982 he opened his family practice in Woodstock and continued to advocate for individuals with IDD.

Subsequent appointments included being Director of the Developmental Disabilities Program at Western University in London. His other duties while at the Program as Western was the sole physician of a 24-bed dual diagnosis unit at St. Thomas Psychiatric Hospital. He was Chief Physician at Southwestern Regional Centre, and Chief Physician at Rideau Regional Centre. In the latter capacity, he worked closely with Dr. Bruce McCreary, Director of the Division of Developmental Disabilities program at Queen's University. During this time, he played an active role in development of the Canadian consensus guidelines for primary care of adults with developmental disabilities (Sullivan et al., 2011; 2018), the health care tool kits for primary care providers of people with IDD and health watch tables for selected developmental and related disabilities (Developmental Disabilities Primary Care Initiative (2011). Tom also held appointments in the Departments of Family Medicine and Psychiatry at the University of Western Ontario, Family Medicine at Queen's University, and Family Medicine at the University of Toronto. In the United States, he was an adjunct assistant professor of Psychology and Human Development at Vanderbilt's Peabody College.

Dr. Cheetham's expertise led to appointment as Medical Director at Orange Grove Center, Chattanooga, Tennessee, in 2009. After a brief period at Surrey Place Centre, he returned to Tennessee in 2011, first as Director, Office of Health Services and later as Deputy Commissioner of Health Services for the Tennessee Department of Intellectual and Developmental Disabilities. Though based in Tennessee, Dr. Cheetham never forgot his Canadian roots and he continued to be an active contributor to the Canadian consensus guidelines, the health

care tool kits and the health watch tables. He was instrumental in disseminating the tools and health watch tables in the United States and internationally. Other publications included book chapters on physical health, the use of psychotropic medications in management of problem behaviours in adults with IDD, and video training modules for health professionals.

Tom was the consummate teacher; he always had time to talk to anyone and would also take the time to make sure the issue was addressed. He was a caring and compassionate man who always seemed to be able to interact with any individual despite their level of functioning. In 2007 he was selected as the Physician of the Year by the Developmental Disabilities Nurses Association. In 2012 he was recipient of the Ontario Association on Developmental Disabilities (OADD) Award of Excellence for better understanding of, or enhancing the quality of life for, those with developmental disabilities. In 2017, he was honoured with the inaugural Surgeon General's Spirit Award for Outstanding Leadership and Service established by the American Academy of Developmental Medicine and Dentistry.

The field of IDD mourns the loss of a passionate and compassionate advocate. The Journal of Developmental Disabilities extends sincerest condolences to dear friend Shirley McMillan, his family, other friends and colleagues.

**Sources:** In part from obituaries prepared by: Jan Rosemergy, Vanderbilt Kennedy Center Director of Communication and Dissemination (<https://vkc.mc.vanderbilt.edu/notables/2018/08/remembering-dr-tom-cheetham-physician-and-advocate/>); yourlifemoments.ca, Woodstock, ON (<http://www.yourlifemoments.ca/sitepages/obituary.asp?oid=1088788>); and staff from Surrey Place Centre (<https://www.surreyplace.ca/dr-tom-cheetham-dear-friend-and-valued-colleague/>).

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## Differential Responses to Early Behavioural Intervention in Young Children With Autism Spectrum Disorders as a Function of Features of Intellectual Disability

### Abstract

*Early behavioural intervention (EBI) treatments are recognized as the most effective interventions to date for young children with autism spectrum disorders (ASD) and have a significant impact on the two domains implicated in the diagnosis of intellectual disability (ID). This study examined the role of concomitant ID features on young children with ASD's outcomes in EBI. Participants were 88 children with ASD aged 51 months on average. Their intellectual quotient (IQ, WPPSI-III), adaptive behaviours (AB, ABAS-II), and the severity of their autism symptoms (CARS) were assessed before EBI and after 12 months of EBI. Among the 55 children who met the diagnostic criteria for ID, 15 displayed such improvements in IQ and AB that they no longer met these criteria after 12 months of EBI. These children also showed improvements on the CARS, whereas the remaining 40 children with ID only improved on the CARS. Children without ID features ( $n = 33$ ) showed the greatest improvements in AB. Predictors of outcomes of EBI differed by ID status. Even at a relatively low intensity, interventions based on applied behavioural analysis may improve cognitive and adaptive functioning among young children with ASD. However, profiles that correspond to diagnoses of both ID and ASD may require further considerations in terms of intensity and duration in planning interventions.*

Intellectual disability (ID) is one of the conditions most often associated with autism spectrum disorder (ASD), with reported comorbidity rates ranging between 25 and 70% (Autism Developmental Disabilities Monitoring Network [ADDM], 2007; Chakrabarti & Fombonne, 2005; Charman et al., 2011; Fombonne, 2003; Yeargin-Allsopp et al., 2003). Learning and behavioural change for young children with ASD who meet the criteria for ID may unfold differently over the course of early intensive behavioural intervention (EIBI) compared to children who do not meet these criteria (Eldevik, Eikeseth, Jahr, & Smith, 2006; Eldevik et al., 2010; Perry et al., 2011). Furthermore, higher pre-intervention intellectual quotients (IQ) and adaptive behaviour (AB) scores are associated with greater responsiveness to EIBI (Bibby, Eikeseth, Martin, Mudford, & Reeves, 2002; Eikeseth, Smith, Jahr, & Eldevik, 2002; Harris & Handleman, 2000). Therefore ID may affect how children with ASD respond to early intervention. Moreover, because these interventions impact intellectual and adaptive functioning, they may also exert an influence

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on the diagnosis of ID in these children. In spite of the importance of programs such as EIBI for children who present features of ASD and ID, relatively few studies have examined this population's progression as a result of interventions delivered under real-world conditions (but see Eldevik et al., 2006; Peters-Scheffer, Didden, Mulders, & Korzilius, 2010)

Two criteria recognized by the American Association of Intellectual and Developmental Disabilities (AAIDD, 2010) for an ID diagnosis are significant limitations in intellectual functioning and adaptive behaviour (AB), as determined by scores two standard deviations below average ( $\pm 70$ ; taking into account the instruments' standard error of measurement) on standardized measures of intellectual quotient (IQ) and AB. Additionally, there should have been evidence of ID prior to the age of 18. Beyond these criteria, the AAIDD recommends that the observed limitations be interpreted in the context of the child's environment and other personal characteristics, and be accompanied by an analysis of his or her support needs. Recent studies report that EIBI yields positive effects on the adaptive and intellectual functioning of children with ASD (Ben-Itzhak & Zachor, 2007; Cohen, Amerine-Dickens, & Smith, 2006; Eikeseth et al., 2002; Eikeseth, Klintwall, Jahr, & Karlsson, 2012; Eldevik et al., 2010). Behavioural intervention among children with ID generally yields gains in multiple spheres of functioning such as communication, social skills, and autonomy, as well as in challenging behaviour (Buckhalt, Baird, & Reilley, 2004; Carter & Hughes, 2007). Preschoolers with both ASD and ID who receive behavioural intervention show improvements in terms of IQ and AB, but these gains may be contingent on the intensity or duration of the intervention (Eldevik et al., 2006; Smith, Eikeseth, Klevstrand, & Lovaas, 1997). Indeed, most of the studies showing sizeable benefits from EIBI are implemented at nearly optimal intensity, that is, at a rate of approximately 40 hours per week for at least 24 months (e.g., Cohen et al., 2006; Granpeesheh, Dixon, Tarbox, Kaplan, & Wilke, 2009; Matson & Smith, 2008). Similarly, studies have demonstrated gains arising from lower-intensity interventions among children with ASD and ID that were deployed over a longer period of time (e.g., 12.5 hours/week for approximately 20 months, Eldevik et al., 2006). Thus, this group of children

may not benefit under circumstances where available resources or other concerns preclude the provision of high-intensity protocols over extended periods. In order to fulfill their mandate to support to all families of children with ASD or ID within their territory with limited resources, publicly funded rehabilitation centres in Quebec must deviate from optimal EIBI implementation parameters. As an organizational compromise to best meet their clientele's needs, centres' service allocation decisions are based upon children's age and overall profile severity. Specifically, children who are older (i.e., will soon enroll in school) or present with more severe profiles will be prioritized for the centres' most intensive offering (the program investigated here), whereas younger children with less severe profiles, or whose parents have declined to receive more intensive services, will either be offered less intensive interventions or enrolled in a low-intensity parental coaching program. The presence of features of ID are not presently taken into account in the allocation of ASD services. This report therefore presents an in-depth look at a sample of children receiving a relatively low-intensity (between 16 and 20 hours per week), community-based early behavioural intervention (henceforth labelled EBI, in contrast to higher-intensity EIBI) program over a 12-month period. These children represented a subgroup of a larger and more diverse sample of children who were recruited upon requesting services at a Quebec rehabilitation centre (Rivard, Terroux, Mercier, & Parent-Boursier, 2015). While the prevalence of ID (36.8%) was documented in the larger sample at intake, the predictive role of co-occurring ID features on children's response to EBI had not been examined in the context of this longitudinal study. The present study therefore sought to contrast the outcomes of 12 months of EBI as a function of whether or not the participants met the criteria of ID according to AAIDD (2010) prior to EBI and to examine potential predictors of responsiveness to EBI.

## Methods

The research protocol for the present study was approved by the research ethics board that oversees rehabilitation centres in Quebec. Because the methodology of the larger longitudinal study within which these data were

collected was detailed in Rivard, Terroux, & Mercier (2014), only the details pertinent to the present investigation are provided here.

## Participants

The present study took place in Quebec, where public rehabilitation centres provide free services to children with ASD and their families. Of the cohort enrolled in the larger longitudinal study, only participants who had been selected to receive the most intensive service available: EBI with a target intensity of 20 hours per week, and for which data pertaining to intellectual functioning and adaptive behaviour were available at intake and after 12 months of EBI, were included in the present study. Participants were 88 children with ASD (65 male) aged between 33 and 63 months ( $M = 50.6$ ,  $SD = 4.9$ ) prior to beginning EBI. Within this group, 55 children met the criteria for ID at intake ( $M = 50.0$ ,  $SD = 5.3$ ) and 33 did not ( $M = 51.2$ ,  $SD = 4.3$ ) according to the AIDD (2010). It should be noted that in Quebec, the term global developmental delay, rather than ID, is used for children under the age of 7. Prior to intake, 47% (total hours received:  $M = 69$ ) and 65% (total hours received:  $M = 34$ ) of the children in the sample had received occupational therapy services and speech language therapy services, respectively. Additionally, 31% had participated in another form of therapy (e.g., Picture Exchange Communication System, hyperbaric oxygen therapy). To the extent that children's age, the proportion of children who had participated in each type of therapy, and the average number of hours of therapy received, did not vary as a function of whether children met the criteria for ID or not; these variables were not investigated further.

## Intervention

The EBI intervention used in the present study adopted a 1:1 child-to-therapist ratio (Lovaas, 1981; Maurice, Green, & Luce, 1996). Intervention sessions took place in a designated room at the child's day care at a rate of approximately 4 hours per day. Each session primarily employed discrete trial teaching, that is, 2- to 5-minute periods in which a specific behaviour or concept was taught, followed by approximately 5 minutes of free play. These sessions also included incidental teaching periods. As applicable, some sessions took place in

a group setting, for instance by involving the child's peers in the teaching of social skills.

The therapists who worked with the participants had an undergraduate degree in special education and had received direct training on EBI through the rehabilitation centre that employed them. They met every 2 weeks with their supervisor, a professional who had a master's degree in psychoeducational studies with a specialization in ASD, to ensure that interventions were aligned with the child's individualized intervention plan. The supervisors were themselves supervised by a licensed psychologist, a PhD and PsyD holder, with a specialization in behavioural psychology with 20 years of experience in EBI supervision. Finally, participants' parents received 21 hours of training in applied behaviour analysis (ABA) and ASD as their child began receiving EBI, and were instructed on how to help their child generalize skills learned in EBI sessions. Parents were encouraged to inform the therapist of their child's progress and express priorities or concerns they had with the intervention as it unfolded.

## Materials

Participants' intellectual functioning was measured using the Wechsler Preschool and Primary Scale of Intelligence (WPPSI-III; Wechsler, 2002). Preliminary analyses conducted on Verbal IQ, Performance IQ, and the General Language Composite score obtained from this measure mirrored those found for the full-scale IQ in both group- and individual-level analyses. In the interest of brevity, only analyses based on the full-scale IQ are reported here. Children's AB was assessed using the Parent/Primary Caregiver Form for ages 0–5 years of the Adaptive Behaviour Assessment System-II (ABAS-II; Harrison & Oakland, 2003). This tool targets adaptive skills within the Conceptual, Social, and Practical domains as recognized by the AAIDD (Schalock et al., 2010), and yields a General Adaptive Composite score. Autism symptom severity was rated by the child's therapist using the Childhood Autism Rating Scale (CARS; Schopler, Reichler, & Rochen Renner, 1988). One of the child's parents also completed the CARS; to the extent that preliminary analyses indicated that parent and therapist ratings were strongly correlated and yielded identical patterns of results, only therapist ratings are reported.

## Procedures

After having provided informed consent, each family participated in an interview during the summer months (May through August) leading up to the beginning of their child's EBI program in September. A research assistant administered a sociodemographic questionnaire to the parents and another administered the WPPSI-III to the child in another room. Both assistants were trained and supervised by a graduate student and a psychologist. Therapists from the rehabilitation centre were trained to administer the ABAS-II as a semi-structured interview with parents at the beginning of the EBI program. Therapists completed the CARS after they had observed the child for seven observation sessions, each of which lasted 1 hour. The intellectual functioning (WPPSI-III), adaptive behaviour (ABAS-II), and severity of autism symptoms (CARS) assessments were repeated after 12 months of EBI.

## Statistical Analyses

As per AAIDD guidelines, children whose IQ and AB scores were two or more standard deviations below (70) the population average were considered to meet the criteria for ID. The outcomes of children who met these criteria and those who did not at the onset of EBI were first examined and compared at the group level. Both groups' changes in IQ, AB, and autism symptom severity were computed and compared to each other by means of independent samples t-tests.

Children's score changes were also analyzed at the individual level. A Reliable Change Index (RCI) was therefore computed for IQ, AB, and autism symptom severity scores for each child according to the method devised by Jacobson and Truax (1991). This measure scales differences in scores at two points in time relative to standard error of measurement of the difference, and thus helps to determine whether the magnitude of a child's score changes exceed what could be attributed to measurement error. RCI was computed as follows:

$$RCI = \frac{x_2 - x_1}{SE_{diff}} \text{ where } SE_{diff} = \sqrt{2(SE)^2} \text{ and } SE = SD_1\sqrt{1-r}$$

The standard deviation of children's scores at intake were used as an estimate of the variability in test scores within the population of children with ASD of this age group who would be selected to receive this type of EBI program. To the extent that the reliability ( $r$ ) measures typically used for these computations (test-retest reliability or internal consistency) could not be obtained on the present sample, these were estimated on the basis of internal consistency values reported in each instrument's manual (for the WPPSI-III and ABAS-II, specifically using data obtained from a sample of children with ASD). A RCI exceeding  $\pm 1.96$  on a given measure indicates that the child exhibited "reliable change," that is, a robust increase or decrease on a given score between two measurement periods. The proportions of children within each group that did or did not demonstrate reliable change was then compared by Chi square tests of independence.

As a measure of clinical significance of changes, the proportion of children who did not meet the criteria for ID at any point (Group 1), children who lost their ID status after 12 months (Group 2), and those who retained it (Group 3) were computed (for a discussion of this type of clinical cutoff-based approach to clinical significance, see Jacobson & Truax, 1991). McNemar's test was then used to determine whether the proportion of children who lost their ID status was statistically significant. A change in ID status does not necessarily imply a large change in either measure, as it is a function of a child's scores relative to the numerical thresholds used to determine ID status. Therefore, follow-up analyses examined whether changes in ID status indicated quantitatively different profiles of change over time. One-way analyses of variance (ANOVAs), followed by Tukey's HSD post hoc tests, compared T1-T0 difference scores for IQ, AB, and CARS between the three groups of children formed on the basis of clinically significant changes. Inasmuch as RCI provides standardized measure of (reliable) change, a global change score could be computed for each child by averaging together RCIs for IQ, AB, and CARS after reversing the sign for the CARS RCI (such that positive RCIs indicated improvement). This average RCI could then be used to

identify which scores (IQ, AB, CARS) at intake were most predictive of overall changes noted over the 12 months of EBI through correlational analyses followed by hierarchical regression analyses. However, visual inspection of these data suggested different relations between both IQ and AB on the one hand, and average RCI on the other, as a function of level of functioning (ID status) at intake. It was therefore deemed prudent to analyze potential predictors of EBI outcomes separately for children who did and did not meet the criteria for ID at intake.

Based on Cohen's recommendations (1988), effect sizes between  $d = .02$ – $.04$  and  $R^2$  or  $\eta^2 = .01$ – $.05$  were considered small; between  $d = .05$ – $.07$  and  $R^2$  or  $\eta^2 = .06$ – $.13$ , moderate; and  $d = .08$  and  $R^2$  or  $h^2 = .14$  or greater, large.

## Results

### Changes in Assessment Scores as a Function of ID status

Table 1 shows average full-scale IQ, AB, and CARS scores before and after 12 months of EBI. Before EBI, 62.5% (55) of participants met the criteria for ID. This group showed a 6-point increase in IQ, 2-point increase in AB, and 1.6 point decrease in CARS scores after 12 months. On average, children who did not meet the criteria for ID showed a 6.2-point increase, a 11.5-point increase, and a 2.6-point decrease on these same measures, respectively. The latter group showed significantly greater gains in AB,  $t(86) = 4.798$ ,  $p < .001$ ,  $d = 1.06$ ; the two groups did not differ in terms of change in IQ or CARS scores,  $t(86) = 0.09$ ,  $p = .927$ ,  $d = 0.02$  and  $t(81) = -0.688$ ,  $p = .389$ ,  $d = -0.20$ .

Table 1. Children's Test Scores Before and After 12 Months of EBI as a Function of Pre-treatment ID Status

		All children ( <i>N</i> = 88)		Meeting ID criteria ( <i>N</i> = 55)		Not meeting ID criteria ( <i>N</i> = 33)	
		T0	T1	T0	T1	T0	T1
	<i>N</i>	<i>M</i> ( <i>SD</i> )	<i>M</i> ( <i>SD</i> )	<i>M</i> ( <i>SD</i> )	<i>M</i> ( <i>SD</i> )	<i>M</i> ( <i>SD</i> )	<i>M</i> ( <i>SD</i> )
WPPSI-III							
Full-scale IQ	88	60.5 (16.8)	66.6 (19.9)	49.7 (6.9)	55.7 (15.0)	78.6 (12.4)	84.8 (12.2)
Verbal IQ	88	63.2 (12.9)	66.5 (14.5)	56.0 (5.3)	58.4 (8.8)	75.3 (12.9)	80.0 (11.9)
Performance IQ	88	70.8 (19.1)	77.5 (22.3)	59.3 (9.9)	66.8 (18.5)	90.1 (14.6)	95.5 (15.9)
General language composite	88	63.8 (17.7) <sup>a</sup>	69.8 (19.1)	53.4 (7.9)	60.6 (14.9)	80.4 (16.4)	85.1 (15.3)
ABAS-II							
Global adaptive composite	88	57.7 (11.8)	63.2 (17.1)	51.1 (6.7)	53.1 (10.5)	68.6 (10.2)	80.1 (11.6)
Conceptual	88	60.7 (12.2)	68.1 (17.1)	54.4 (7.6)	58.0 (11.2)	71.2 (11.2)	84.9 (10.8)
Social	88	63.0 (13.8)	68.5 (17.7)	55.6 (7.9)	58.5 (12.1)	75.3 (12.6)	85.1 (12.2)
Practical	88	58.9 (12.1)	61.6 (15.6)	52.5 (8.2)	52.8 (10.9)	69.7 (9.5)	76.2 (10.4)
CARS							
Total	83	34.6 (8.2)	32.7 (8.9)	38.2 (6.7)	36.6 (7.9)	28.0 (6.5)	25.4 (5.5)

Note. T0: At intake, before EBI; T1: after 12 months of EBI. WPPSI-III = Weschler Preschool and Primary Scale of Intelligence - III, ABAS-II = Adaptive Behavior Assessment System - II, CARS = Childhood Autism Rating Scale.

<sup>a</sup> The general language composite for two children was below the scoring threshold at T0, therefore N = 86.

The proportions of children who demonstrated reliable score increases or decreases over time, or no change, as per RCI computations are shown in Table 2. In order to achieve a RCI of  $\pm 1.96$ , a child would need to demonstrate a 11.42-point change in IQ, a 3.21-point change in AB, or a 8.02-point change in autism symptom severity. A greater proportion of children who did not meet the criteria for ID at the beginning of the study showed reliable improvements in terms of AB than those who initially met the criteria for ID. However, the two groups did not differ in terms of the proportion of children demonstrating reliable changes in IQ or autism symptom severity scores (see Table 2).

Fifteen (27.27%) of the children who initially met ID criteria experienced such improvements in terms of IQ and AB that they no longer met ID criteria after 12 months of EBI. The decrease in the proportion of children who met ID criteria after 12 months of EBI was statistically significant,  $\chi^2(1, 88) = 13.07, p < .001$ . None of the children who did not meet ID criteria at intake did so after 12 months of EBI. Means for each group at T0 and T1 are displayed in Figures 1 through 3. All three groups differed in their changes in IQ,  $F(2, 85) = 24.25, p < .001, \eta^2 = .36$ . The children who lost their ID status showed

IQ gains over three times larger than those who did not meet the criteria for ID at any point in time, who in turn showed higher gains than those who retained the ID status. Changes in AB also varied across groups,  $F(2, 85) = 16.41, p < .001, \eta^2 = .28$ . Children who did not meet the criteria for ID and those who lost their ID status did not differ, but both showed greater gains than children who retained the ID status. The groups did not differ in terms of their improvements on the CARS,  $F(2, 80) = 0.95, p = .391, \eta^2 = .02$ .

### Predictors of Outcomes of EBI

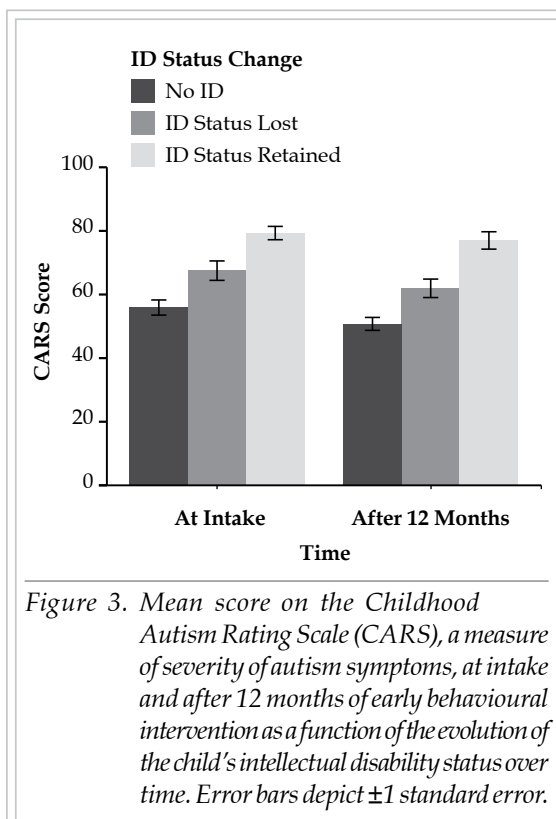
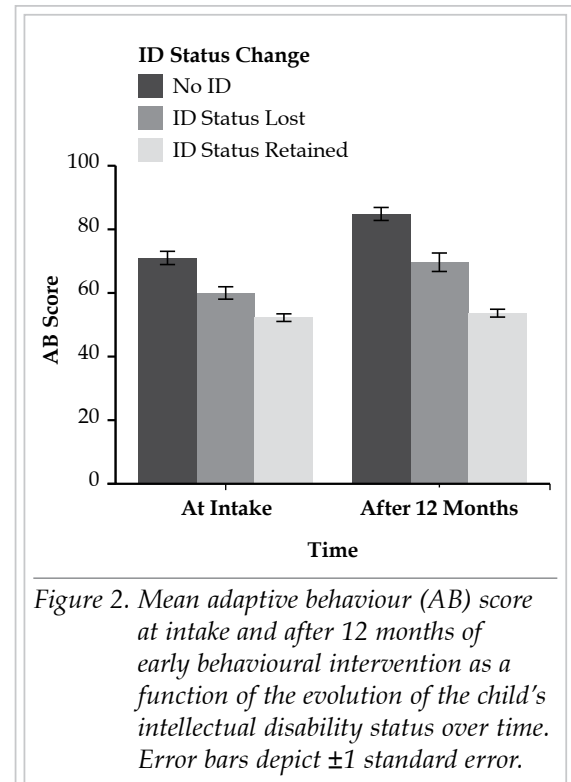
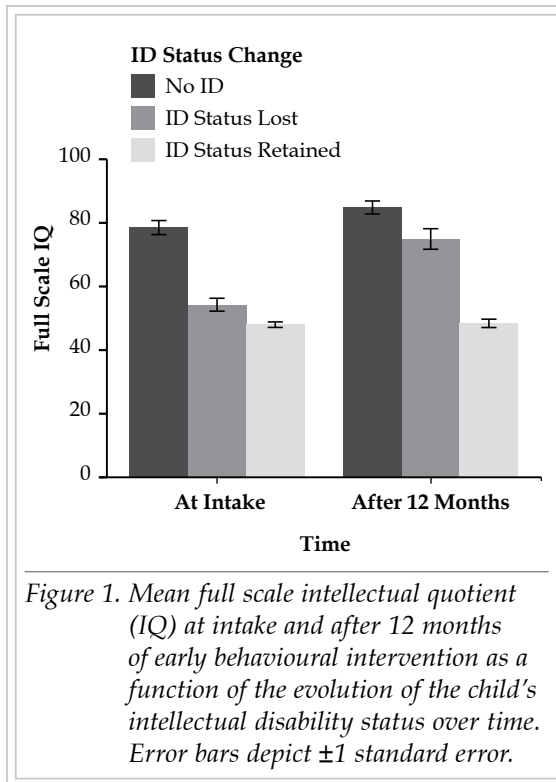
Figure 4 on page 12 depicts children's average RCI, where a positive index denotes improvement, as a function of intake scores. Both IQ and AB appeared to be non-linearly related to average RCI: specifically, a visual inspection of these graphs suggested that these relationships may differ as a function of ID status. In contrast, autism symptom severity appeared linearly related to average RCI.

Table 3 on page 12 presents correlations between RCI and children's characteristics at intake as a function of group membership. Among children who met the criteria for ID at

Table 2. Proportion of Children Showing Reliable Increase or Decrease, or no Change, Based on ID Status at Intake

		All children (N = 88)		Meeting the criteria for ID (N = 55)		Not meeting the criteria for ID (N = 33)		$\chi^2$
		n	%	n	%	n	%	
IQ	Decrease	1	1.1%	0	0%	1	3.0%	$\chi^2(2, 88) = 0.66^a, p = .282$
	No change	59	67.0%	39	70.9%	20	60.6%	
	Increase	28	31.8%	16	29.1%	12	36.4%	
AB	Decrease	17	19.3%	14	25.5%	3	9.1%	$\chi^2(2, 88) = 21.57, p < .001$
	No change	27	30.7%	24	43.6%	3	9.1%	
	Increase	44	50.0%	17	30.9%	27	81.8%	
CARS	Decrease	9	10.8%	21	9.3%	4	13.8%	$\chi^2(2, 88) = 0.52^a, p = .662$
	No change	73	88.0%	24	88.9%	25	86.8%	
	Increase	1	1.2%	9	1.9%	0	0.3%	

Note. A RCI of  $\pm 1.96$  was used to identify reliable score increases or decreases. IQ = intellectual quotient, AB = adaptive behaviour; CARS = Childhood Autism Rating Scale (autism symptoms).<sup>a</sup> Fisher's exact test.



intake, IQ correlated positively with average RCI, whereas autism symptom severity scores showed a significant negative association with average RCI. Age and AB scores were uncorrelated with average RCI. Thus, a hierarchical linear regression was conducted with T0 IQ and autism symptom severity scores entered in successive steps. On its own, IQ explained 7% of the variance in RCIs. The addition of autism symptom severity scores significantly improved the model's prediction. When both variables were entered into the model, autism symptom severity scores, but not IQ, significantly contributed to the prediction. Among children who did not meet the criteria for ID, baseline IQ and AB scores were significantly and negatively correlated with average RCI, whereas age and autism symptom severity scores were not associated with RCI. IQ explained 14% of the variance in RCI. The addition of AB scores to the regression model significantly improved this prediction by 20%. Both variables significantly contributed to the predictive strength of the final model.

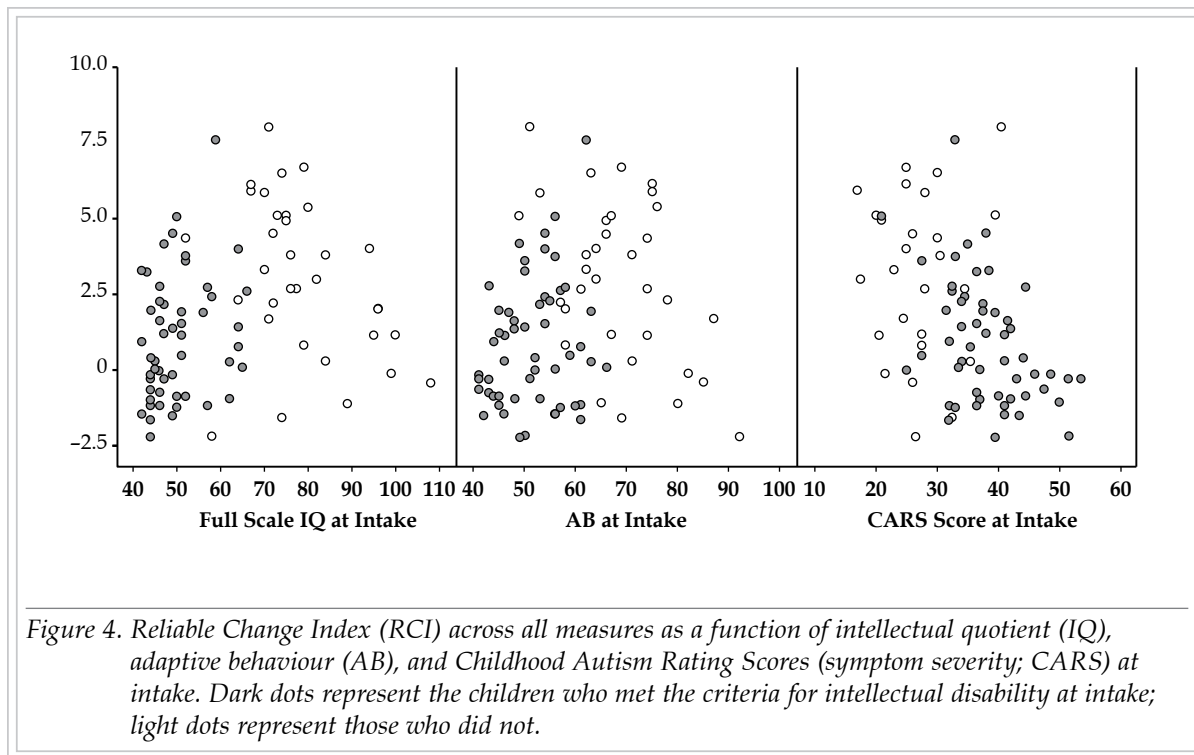


Figure 4. Reliable Change Index (RCI) across all measures as a function of intellectual quotient (IQ), adaptive behaviour (AB), and Childhood Autism Rating Scores (symptom severity; CARS) at intake. Dark dots represent the children who met the criteria for intellectual disability at intake; light dots represent those who did not.

Table 3. Bivariate Correlations and Hierarchical Regression Analyses Predicting Reliable Change Indices From Children's Characteristics at Intake

	Meeting ID criteria (N = 55)				Not meeting ID criteria (N = 33)			
	<i>r</i>	<i>p</i>			<i>r</i>	<i>p</i>		
Age	.09	.521			.24	.184		
IQ	.30	.025			-.38	.031		
AB	.24	.084			-.46	.006		
CARS	-.44	.001			.00	.987		
Predictor	$\Delta R^2$	$\beta$	<i>F</i>	<i>p</i>	$\Delta R^2$	$\beta$	<i>F</i>	<i>p</i>
Step 1	.07		3.77	.058	.14		5.13	.031
IQ		.26		.058		-.38		.031
Step 2 <sup>a</sup>	.21		9.42	.003	.20		9.25	.005
(IQ)		.14		.292		-.36		.022
CARS		-.40		.003				
AB						-.45		.005

Note. IQ = intellectual quotient, AB = adaptive behaviour; CARS = Childhood Autism Rating Scale (autism symptoms).<sup>a</sup> The predictor entered in the second step differed by group (CARS scores for those meeting the criteria for ID, AB for those who did not).

## Discussion

This study focused on the evolution of the clinical profiles of 88 children with ASD who participated in an EBI program consisting of approximately 20 hours per week of intervention services provided by a public rehabilitation centre. At the beginning of the intervention, 55 children (62.5%) met the diagnostic criteria for ID. When assessed with the same two standardized measures after 12 months of services, 15 of them had made such improvements that they no longer met the criteria for ID. A closer inspection indicated large IQ gains and modest but significant AB gains in this group, similar to Eldevik et al.'s (2006) observations among children with ASD and ID who had participated in 12.5 hours per week of EBI on average. These results suggest that EBI, even when offered in community settings and with relatively low intensity, can yield positive results among children with a dual diagnosis of ID and ASD. It should nevertheless be noted that although children who met ID criteria prior to intervention improved significantly as a whole, 70% of those 55 children still met these criteria after 12 months of EBI. This subgroup showed no statistically significant changes in either IQ or AB. All children, regardless of ID status, showed comparable improvements in autism symptom severity, however.

The fact that a number of children meeting the criteria for ID made statistically (i.e., in terms of absolute scores on standardized tests) and/or clinically significant (i.e., relative to diagnostic criteria) gains is encouraging inasmuch as previous studies found that early deficits in children diagnosed with global developmental delay tend to persist into the school years, at which point a diagnosis of intellectual disability is made (e.g., Shevell, Majnemer, Platt, Webster, & Birnbaum, 2005). Relatedly, the developmental gap in terms of IQ and AB between children with ASD and their typically developing peers tends to persist or worsen among those on a waiting list for services (Authors, 2015) or receiving control interventions (Klintwall, Eldevik, & Eikeseth, 2015; see also Eldevik et al., 2010). Furthermore, among those receiving EBI, IQ and AB at intake were both related to intervention outcomes (Eldevik et al., 2010). In contrast, results from the present study suggest that an early diagnosis of global

developmental delay (or indicators of ID present at an early age) may be somewhat fluid if children meeting the diagnostic criteria for ID gain access to behavioural intervention services during the critical preschool years. These data may be relevant to service providers who must determine whether children with ASD and indicators of ID are likely to benefit from interventions, and to parents for whom a label of global developmental delay could be a cause for concern for the child's prognosis.

Among children's intake assessment scores, previous research had identified IQ as a strong predictor of gains in EBI, and reported positive associations between IQ or AB and intervention outcomes (e.g., Ben-Itzhak & Zachor, 2007; Harris & Handleman, 2000; Magiati, Charman, & Howlin, 2007; Magiati, Moss, Charman, & Howlin, 2011; Perry et al., 2011). Here, the measures collected at intake showed different and somewhat non-linear patterns of association with 12-month outcomes as a function of whether children met the criteria for ID. For those who did not, IQ and AB were negatively correlated with overall gains. The philosophy of EBI should be to prioritize areas where the child presents the most difficulty (Rivard & Forget, 2012; Rivard et al., 2016; Smith, 2010, 2011). The closer a child's IQ and AB to average levels, the more emphasis would be placed on other aspects of the child's development and social functioning. Gains on these dimensions might then not be captured by the measures used here. In contrast, among children who met the criteria for ID, IQ was weakly but positively linked with overall improvement. Importantly, CARS scores more strongly and negatively predicted outcomes for this group. Thus children with more severe profiles did not improve as much, perhaps because they required a more intensive intervention. Alternatively, interventions for these children may have prioritized more salient and urgent difficulties, such as the presence of challenging behaviours. Thus, the strength and direction of associations between pre-intervention measures and children's outcomes reported in the literature may be a function of the variability in levels of functioning represented in a given sample.

One limitation of this study is its lack of inclusion of a control or comparison group. However, the outcomes observed here for EBI

were previously contrasted with those of a minimally intensive (1 hour/week of therapist support) parental coaching program offered by the same rehabilitation centre (Rivard et al., 2014). Furthermore, the principal goal of the present study was to compare children with different ID statuses who received the same type of treatment. On this note, while all children received EBI at a comparable intensity and under similar conditions, it is possible that some of the differential outcomes noted in the present study could be attributable to other therapies (e.g., occupational therapy, speech therapy) that some families may have elected to pursue in parallel with EBI offered by the rehabilitation centre. While pre-EBI enrolment in these other therapies did not vary as a function of group, data pertaining to these supplemental interventions were not collected during or after EBI. Another important limitation is that children were assessed by individuals who were aware of the intervention, such that expectancy effects cannot be ruled out. Nevertheless, the improvements observed here are generally consistent with those reported in the EBI literature (Eikeseth et al., 2002, 2012; Eldevik et al., 2006; Remington et al., 2007). Additionally, neither parents, therapists, nor research assistants were aware of the specific goals of this study, which was embedded in a more extensive longitudinal investigation during which various other measures were administered.

It should also be noted that the constraints and resource limitations under which Quebec rehabilitation centres typically operate limited the extent to which our study could demonstrate the potential effectiveness of EBI, particularly for children with ID or more severe clinical profiles generally. Indeed, because of the extensive waiting lists for EBI, most of the participants were older than recommended for optimal outcomes (Makrygianni & Reed, 2010; Reichow, Barton, Boyd, & Hume, 2012; Virués-Ortega, 2010) and were only able to receive 12 months of EBI before beginning school, at which point EBI services are discontinued (c.f. 20 months of lower intensity EBI in Eldevik et al., 2006). It is possible that if these children had been offered EBI sooner, or for an extended duration, they would have shown gains comparable to their peers. This would be the case if, as we speculate, challenging behaviours and other barriers to learning in these types of inter-

ventions had to be addressed first. Measures of challenging behaviour, or records of each child's intervention plan, could not be collected to verify this point. Relatedly, it is possible that more varied or sensitive measures of children's outcomes could have identified some benefits in of EBI in other areas among children who did not show appreciable gains in terms of IQ, AB, or symptom severity.

This study furthers our understanding of the effects of EBI on young children with ASD and ID. After 12 months of intervention, several children had made statistically significant improvements in terms of intellectual and adaptive functioning, two spheres of development associated with ID. These results support the idea that even relatively low-intensity behavioural interventions offered as universally available public services could benefit children with ASD, including those who present with ID. These improvements may, in turn, lead to increased autonomy and an improved quality of life for these children and their families. However, the fact that children with ID had lower AB gains as a whole and showed heterogeneous change over time profiles, as well as the suggestion that different measures best predict outcomes for children with and without concomitant ID, confirms the importance of taking this dual diagnosis into account when devising intervention plans in early childhood. This information may be especially pertinent in a context where resources are limited and the goal is to provide beneficial (if not optimal) assistance to a very large number of individuals, such that it is necessary to match service intensity to intake profiles and anticipated needs. Indeed, children who present features of ID along with ASD may derive greater benefits, or benefit in larger numbers, from EBI services offered more intensively (i.e., up to 30–40 hours per week) and over longer periods (i.e. for more than 12 months) than what is currently provided.

## Key Messages From This Article

**People with disabilities.** You deserve services that are suited to your specific strengths and needs. We are trying to understand why some children with ASD do not get the same benefits from early intervention as others do so that we can provide better services for everyone.

**Professionals.** Some children with ASD who also show features of ID, especially those with more severe symptoms, may not benefit as much from established early intervention services.

**Policymakers.** Additional resources may be needed to help children with ASD who also show features of ID, or present with more severe symptoms, to benefit optimally from early intervention.

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## The Role of Social Enterprise in Creating Work Options for People With Intellectual and Developmental Disabilities

### Abstract

*It has been broadly acknowledged that supported, inclusive employment can promote independence, quality of life and social integration for people with intellectual and developmental disabilities (IDD). Despite the ongoing development of the individual placement and support model of supported employment, competitive employment rates for this population remain low. Social enterprise is a means of enhancing employment participation through the creation of jobs and job training in competitive community businesses in the "3<sup>rd</sup> sector." This study used a case study methodology to examine practices of successful social enterprises for workers with IDD that led to both sustainable business ventures and promote desired social outcomes. Four Ontario and one Alberta work integration social enterprises (WISE) provided the primary data sources. Cross-case analysis revealed a number of tensions and challenges as agencies strive to meet provincial employment standards while running successful businesses and honouring worker need for choice and social inclusion. Results include identification of best practices and key outcome indicators for successful ventures at individual, business and community levels.*

Considerable attention has focused on efforts to improve the persistently low employment participation of people with intellectual and developmental disabilities (IDD) (Ellenkamp, Brouwers, Embregt, Joosen, & van Weeghel, 2016; Santilli, Nota, Ginevra, & Soresi, 2014). These efforts are rooted in principles of normalization (Wolfensberger, 2011) and human rights (United Nations, 2006), and the role of inclusive employment in promoting independence, quality of life and social integration (Kocman & Weber, 2016; Lysaght, Petner-Arrey, Howell-Moneta, & Cobigo, 2016).

One approach to improving employment participation is work integration social enterprise (WISE), also known as social firms, affirmative businesses, social cooperatives, and social purpose businesses. WISEs are a specific type of social enterprise with a primary purpose of providing job training and/or creating jobs for work-disadvantaged populations (Chan, Ryan & Quarter, 2017); O'Connor & Meinhard, 2014). WISEs have dual social and revenue generation goals, producing goods and services for trade in the regular marketplace while also striving to improve economic and social conditions for populations who face challenges with self-sufficiency (Quarter, Mook, & Armstrong, 2009). These populations

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#### Keywords

social enterprise,  
social inclusion,  
social integration,  
employment

include people who have encountered poverty and/or homelessness, marginalized youth, Aboriginal communities, ethnic minorities, women, seniors, and people with disabilities, including mental, physical, and intellectual disabilities (Kidd & McKenzie, 2014).

In the IDD field, non-profit organizations have a long history of entrepreneurship and employment activities for their clients. This has often taken the form of sheltered workshops. Once commended for providing productive work and a safe and social environment, in the 1980s there was a movement away from sheltered workshops due to a philosophical shift toward social inclusion, normalization, and social role valorization (Caruso & Osburn, 2011; Galer, 2014; Katz, 2014). In Ontario, the social enterprise model for people with IDD started to appear in the mid-1990s and early 2000s (Lysaght, Krupa, & Bouchard, 2015). A recent policy move by the Province of Ontario to advance a social enterprise strategy (O'Connor & Meinhard, 2014) as well as a 2015 imperative by the Ministry of Community and Social Services (MCSS) that transfer funding agencies should discontinue sheltered workshops (MCSS Community Supports Policy Branch, 2015) has garnered further interest in the social enterprise model.

Currently, the most accepted intervention strategy to improve employment outcomes is supported employment, and in particular the individual placement and support model (Kirsh, Krupa, Cockburn, & Gewurtz, 2010). Supported employment programs seek to place workers in competitive workplaces, train them relative to specific job demands, accommodate performance differences, and provide ongoing support (Bond, 2004). Issues related to choice and social inclusion are difficult to address in this model, due to the inherent problem of matching a worker with a limited skill set to available jobs in a competitive and shrinking job market. In fact, few programs have used worker satisfaction and inclusion as outcome metrics (Lysaght, Cobigo, & Hamilton, 2012). Recently the wisdom of supported employment as the primary or exclusive entry point to employment has been questioned, based on the failure of this model to adequately meet the needs of a diverse population (Hall, 2009; Lysaght et al., 2016; Wilton, 2004). In this context WISEs are emerging as another alternative.

Questions have been raised as to the role of WISEs relative to their contributions to civil society and community integration for people in marginalized groups (Cooney, Nyssens, O'Shaughnessy, & Defourny, 2016) including those with IDD. For example, within these enterprises there exist no guidelines to ensure that current imperatives around fair and inclusive employment are met. This means that there are no standards to guarantee that workers are provided opportunities to build connections with the larger community, exercise choice relative to work options, build competitive work skills, or increase their access to and success in entering other community work options. In Ontario, the WISE sector is highly diverse due to a broad range of operational models, forms of incorporation and funding sources (Brouard, McMurtry, & Vieta, 2015), and this diversity is evident in the intellectual disability sector.

While the benefits of WISE as a means of enhancing the employability of marginalized populations in Canada have been reviewed (Lysaght & Krupa, 2011; Krupa, Lysaght, Vallee, & Brown, 2013), their contributions to the lives of people with IDD and the local communities in which they live and work have not been widely examined. Studies in Europe and the United Kingdom have demonstrated the contributions of WISE to engaging marginalized groups and moving them towards conventional employment and higher incomes (O'Connor & Meinhard, 2014), while some Canadian studies have focused on the social and economic benefits of WISE (Chan, 2016; Chan, Ryan & Quarter, 2017). Most of these studies, however, have focused on mental health populations, and there has been limited focus on WISEs that provide long term employment rather than transitional programming. The present study was intended to enhance understanding of the advantages and challenges associated with IDD-focused WISEs and to promote ongoing refinement and evolution of the model within the Canadian context.

## Methods

Using a concurrent, multiple case study approach (Yin, 2014), we identified five "cases" – individual WISEs purposefully selected to inform the key research questions, including:

1. What conceptual models/philosophies have guided the development of WISEs for persons with IDD?
2. What factors have contributed to promoting and defining these enterprises?
3. What policies and structures contribute to or detract from the establishment of WISEs?
4. What factors contribute to the success and survival of WISEs?
5. What strategies promote choice, independence and social inclusion for workers?
6. What does social inclusion look like in these varied contexts?

The study was reviewed and received ethical clearance by the Queen's University Health Sciences Research Ethics Board.

## Data Sources

Cases were purposively selected from a national catalogue of WISEs for people with IDD developed in this study's first phase (Lysaght et al., 2015). Included enterprises had been established for a minimum of 2 years and served a primary population of persons with IDD. Selection of the WISEs was intended to create a maximum diversity sample, and as such the cases were varied based on funding sources, historical development, geographical location, type of commercial activity, and wage structure in order to create a rich and varied snapshot of the WISE sector. The focus of the study was on enterprises operating in the Province of Ontario, but one out-of-province (i.e., Alberta) WISE was selected to provide a point of comparison (since employment policies are largely set at the provincial level). A profile of the selected WISEs appears in Table 1.

## Data Collection

Each individual case study involved a range of data sources, including interviews, organizational documents, observation, and focus groups. Interviews of about 30 minutes were conducted with employees with IDD, supervisory staff, board members, and customers and followed semi-structured interview protocols developed for each participant category. Longer interviews (120 minutes or more) were

held with business managers and executive directors. Key documents reviewed included articles of incorporation, annual reports, balance sheets, publicly available web pages and marketing materials.

## Data Analysis

As recommended by Yin (2014) for multiple case designs based on replication logic, each case was analyzed independently using line-by-line review of interview transcripts, observation reports, and key company documents. The analysis dissected emergent descriptive and explanatory themes that we identified relative to the research questions. Case summaries were developed and data display matrices of emergent themes were constructed. Findings relative to each case were provided to the staff of each WISE, and discussed in a collaborative manner to both share our observations and confirm interpretations. The second stage of analysis compared and contrasted the emergent theme tables using cross-case comparison. Common trends, points of tension and challenges, and unique approaches were identified across cases. The initial thematic findings were presented to members of our partner organization, the Ontario Disability Support Network (ODEN), a provincial network of employment service providers (ODEN 2018). Discussion with this group assisted interpretation and organization of findings.

## Results

The cross-case analysis yielded eight major themes that summarize the primary findings. These themes highlight key issues that emerged as being salient to the use of social enterprises as a means of addressing the employment needs of people with IDD.

## Structural Models

Business structures influence how philosophy and values are operationalized and how goals and objectives are achieved. In addition, legal business structures protect individuals involved in the enterprise from potential liabilities and risks associated with the work of the WISE, and influence the nature of the relationships with a sponsor or affiliated organizations. The WISEs

Table 1. Profile of Participating WISEs

Case #	Incorporation Model	Nature of Business(es)	Setting	Annual Revenues*
1	Co-operative	<ul style="list-style-type: none"> <li>• Baked goods production</li> <li>• Catering</li> <li>• Coffee kiosks</li> <li>• Toy sterilization</li> </ul>	Large urban; multi-site	\$190,000
2	Multi-business organization under overarching structure and board of directors. Arm's length relationship to an IDD social service organization.	<ul style="list-style-type: none"> <li>• Wood furniture production</li> <li>• Restaurants/cafés</li> <li>• Copying/printing</li> <li>• Recycling</li> <li>• Furniture restoration</li> <li>• Packaging</li> <li>• Farm workers</li> <li>• Printing business</li> </ul>	Rural – regional municipalities	\$505,000
3	Multiple businesses operated by parent IDD service provider, not separately incorporated, but with separate books. One manager for all businesses reports to agency executive director, who reports to board of directors.	<ul style="list-style-type: none"> <li>• Wood products</li> <li>• Gift shop</li> <li>• Event catering</li> <li>• Janitorial contracts</li> <li>• Coin laundry</li> <li>• Rental income properties</li> </ul>	Small urban setting; multiple locations	\$342,100
4	Single business operated by parent IDD service provider, not separately incorporated, but with separate books. Manager reports to agency executive director and board of directors	<ul style="list-style-type: none"> <li>• Laundry services</li> <li>• Linen rentals</li> </ul>	Small urban setting; single location	\$825,000
5	Suite of businesses operated by parent disability social service organization. Director of social enterprise and employment oversees businesses, each with its own cost centre	<ul style="list-style-type: none"> <li>• Airport cart retrieval</li> <li>• Bottle depot</li> <li>• Bottle pick up service</li> <li>• Paper recycling</li> </ul>	Large urban setting; multiple locations	\$1,587,000

\* Drawn from annual reports for 2014–15

studied here operated under three distinct structures: agency-owned and operated; cooperative; and not-for-profit corporation. Most operated a number of distinct businesses.

In addition to their legal business structures, some had also been granted charitable status. To be eligible for charitable status a WISE must have activities that further a charitable purpose, specifically implementing activities that relieve unemployment and other social and economic conditions associated with disability. In the case of co-operatives, there is a requirement that the worker “partners” will be involved in management or decision-making.

### **Emergence from the Legacy of the Past**

WISEs for people with IDD face the unique challenge of distancing themselves from sheltered employment, particularly if sheltered workshops were operated by the particular agency in the past. A range of business strategies were identified as countering the spurious association with sheltered workshops. These included: producing high quality, desirable goods or services that meet a recognized or potential market need, and thus are valued in the community; presenting a positive public image of people with IDD and their work roles; creating integrated work structures, where workers with and without IDD work side-by-side; providing opportunities for interactions between workers with IDD and the general public; and finally, paying wages at minimum wage or industry standards.

Where WISEs had emerged from former sheltered workshops, re-creating the enterprise to be seen as market-based, real employment required creating alternative options for legacy workers with IDD who were not interested in or able to work to business expectations. Such shifts could result in unresolved tensions for WISE developers, particularly when large numbers of former workers needed to be re-deployed. For example, when former sheltered workshop workers moved to day programs, many were left with diminished income, empty days and lack of productive engagement. Such situations presented a crisis point, challenging the underlying mission of enhancing the productive lives and incomes of people with IDD.

### **Worker Support Models**

The WISEs in this study demonstrated a number of approaches to providing worker support, and various means of dealing with the costs associated with that support. Overall, four general support strategies were identified:

**Trained agency staff provide worker support on a daily, ongoing basis.** This approach is based on the assumption that specialized training and expertise in rehabilitation strategies can best address the behavioural and support needs of workers with IDD.

**Business/trade specialists provide daily worker support.** In this model, trade specialists (e.g., skilled cooks, carpenters, landscapers) supervise workers. In all instances, these were individuals who lacked social service training, but were keenly sensitive to individual needs and how to nurture capacity while holding workers to expected standards of the trade or business.

**Natural supports combined with support of trained agency staff.** This involved support by co-workers, normally those without disability. In one of the WISEs, trained agency staff assisted with worker development and support; however, a portion of the workforce did not have a disability, and worked alongside those with IDD. These non-disabled workers saw themselves as providing collegial guidance and encouragement to the co-worker with IDD, but not responsible for their supervision.

**Natural supports combined with limited job coaching.** In this approach, workers without disabilities in an integrated workforce provided support to workers with IDD as needed, but without regular involvement of support professionals. Parent agency job coaches, who provide supports across all their employment programs, stay in touch with the workers and provide follow along supports, typically intervening where a worker is struggling, needs to learn a new skill, or desires to move into a community setting.

Trained support worker/job coach salaries were covered by the parent agency staffing budget in most cases but there was a movement in some settings to cover at least part through business revenues. When business or trade specialists

provided the worker oversight their salaries were covered by business revenues. In all instances where coworkers without disabilities provided natural supports, those wages were covered by business revenues.

## Compensation Structures and Strategies

The compensation approach was typically driven by agency philosophy, the incorporation structure, and the productive capacity of the workers. Overall, the enterprises sought fair and equitable means of compensating individuals and to provide some level of choice to workers and their families. Several different compensation approaches were observed, and often several were used within a single organization. The means of compensation included minimum wage, market-based wages, contractual pay, profit share, work incentive pay, piece rates, and training stipends (see Table 2). Managers expressed concerns, however, most

arising at the intersection between business philosophy and mission, and the need for an enterprise to be financially sustainable. Overall, the wage issue was the most volatile one emerging in agency discussions, but pay rates were not a major focus for workers themselves.

## Worker Engagement

While three WISEs actively engaged families in decision making and other mission-supporting work, two also developed structures to systematically and directly engage worker voice through the creation of a worker advisory committee. Sample contributions of committee members included discussing health and safety initiatives, developing surveys on key issues, and weighing in on the philosophy and goals of the organization. The cooperative WISE provided the most fully-developed structure for worker participation, and by definition this incorporation model depends on the collective involvement of workers in work decisions and

*Table 2. Compensation Strategies*

<i>Method</i>	<i>Business(es)</i>	<i>Description/Example</i>	<i>Tensions</i>
Minimum wage	2, 3, 4, 5	Entry level wage matches provincial minimum wage; rate increases with experience.	Requirement that all workers be paid at least minimum wage rate typically excludes those who are not sufficiently productive to merit this pay; tension between desire of business to be inclusive and engage many and need for business to be sustainable.
Market-based wage	5	Wages are competitive or close to competitive for the nature of the work. May exceed minimum wage.	As with minimum wage.
Worker contract	2	Worker agrees to perform certain work for a set compensation. For example, a farm worker may be contracted to clean animal stalls and feed livestock bi-weekly for an agreed weekly rate.	Some workers respond well to this arrangement, which allows them to work without deadlines and to target task completion rather than timeliness. If the weekly pay, when calculated as an hourly rate, averages below minimum wage, the business may be seen as violating minimum wage laws.

*continued on following page*

Table 2. Compensation Strategies (continued)

<i>Method</i>	<i>Business(es)</i>	<i>Description/Example</i>	<i>Tensions</i>
Profit share	1	Available only within registered co-operatives, profits realized over and above operating expenses are shared proportionally by member workers based on the number of hours each works during the month in question. Compensation in this instance was calculated as a portion of monthly sales (e.g., 33% of sales were divided by workers) with the percentage reviewed regularly.	This model generates a group work ethic wherein all are responsible for the success (and pay rates) of the business. Members determine who should be admitted to the business, and who will be hired as support workers. In the case observed here, wages were typically lower than minimum wage; however, workers have increased control over their work situation.
Work incentive	4	Workers receiving provincial benefits (e.g., ODSP) are paid an hourly "incentive" for work that supplements the benefits received.	Most workers in the instances where this was observed reported being satisfied with receiving a small regular cheque on top of their disability benefits; in fact, some had limited awareness of what they were being paid. While a low pay rate allows a business to employ more workers, the practice diminishes the view of the work as "real employment" and places the business in violation of provincial law.
Training Wage or Stipend	3, 4	Similar to a work incentive, workers receive a below-minimum wage hourly rate while in a training phase.	This rate is justified as serving to reward workers who are not competitive in terms of vocational skills or productivity. This payment arrangement is considered defensible when it is time limited and associated with skill development, and where the pay rate increases as skills advance.
Piece rate	2	Workers are paid for the number of products completed, such as items packed or furniture items produced.	Rewards stronger workers who produce well, and is based on the notion of fair or proportional reward based on capacity. This practice was observed only in a few businesses that employed less productive workers who were committed to holding a job. Perceived by most as sheltered work.

processes. In this setting, workers with IDD were part of decision making for hiring, product development, work routines and policies. Some workers were also recognized as leaders and were supported in developing entrepreneurial skills.

## Advancing Inclusion

Social inclusion for workers was a central goal for all participating WISEs, and three primary means of achieving inclusion of workers were identified. In the first approach, the workforce consisted of a mix of workers with IDD and other workers with or without disabilities. At one WISE it was noted that there was no distinction in the business between the workers with IDD and others, and that co-worker and public perceptions of people with disabilities have been enhanced:

It has been great. Everyone accepts – everyone likes these guys big time. But they can see how hard they work, how much dedication they put into it.

In the second approach (sometimes seen in combination with the first), inclusion was achieved by having contact with the public in the natural course of the work. This contact was realized in the context of work activities such as serving the public in a retail sales context, doing landscaping, recycling pick-ups or other tasks in community spaces, or simply traveling on public transit as part of the job.

In the third approach enhanced social participation occurred in workers due to their increased incomes, and their heightened level of social confidence built on the job. For example, workers, when talking about how they used their income spoke of shopping, attending movies, going out for coffee with friends, participating in public events, and saving up for special things like travel.

In addition, the WISEs prided themselves on the impact the business as a whole made on the local community, such that the work of the WISE broke down stereotypes, built sustainable relationships between the company and the community, addressed social and economic gaps in the local community and built goodwill towards the IDD population by making them a positive force within the community.

## Career Progression

A range of options related to promoting worker access to a variety of jobs, work-related opportunities and career advancement were present across the case studies.

Most of the WISEs included a suite of business ventures. The creation of new business opportunities and new types of jobs was one important way to ensure that people with IDD can find work that is a match for their interests, strengths and needs, and was seen in WISEs that ran multiple businesses. In rural communities, it was also reported to be the only source of new job development in the region. Business creation required that WISE leaders were highly entrepreneurial and continuously attentive to local opportunities related to products and services. For example, one rural WISE identified an opportunity to integrate workers into existing farm operations in the region, and to serve as the employer for the workers in a brokerage type of relationship. Enterprises also found ways to create novel growth experiences for workers that could help build skills and enhance job satisfaction.

There were several examples of efforts to develop jobs with higher levels of responsibility, and titles and compensation to reflect this responsibility. These included a type of foreman position that had workers with IDD oversee attendance, holiday schedules and related work routines, and also the potential in one workplace for workers with IDD to move to general labourer positions reflecting a higher level of experience and work performance. One WISE created an opportunity for some of their workers to travel to France to visit social firms built around a similar model to their own. There was some pushback from families, however, when enterprises tried to move workers into higher level positions. In some instances families were reported to be uncomfortable with their family members earning higher pay rates which they believed would compromise the financial security of disability benefits. In one instance, where part of the workforce was unionized, family members had concerns about the risks of union membership.

Movement to jobs in the conventional workforce is supported within these WISEs, but

direct services to support this transition was typically seen as out of the scope of the WISE mandate. The WISEs did not routinely collect data related to movement to other mainstream work. Within these WISEs workers with IDD who were motivated to seek employment in the conventional workforce were typically connected to social agencies with job placement and support services. Each WISE provided examples of workers moving to mainstream jobs, and at least one indicated that some of their workers also hold part-time jobs in the broader community workforce.

It was noted that some attempts by workers to move to conventional jobs had not been successful. Issues related to the general lack of suitable employment and difficulties with transportation, particularly within rural areas, were also raised. There was evidence that the workers experience a high level of social belonging within the WISE and that families may push-back when they perceive that this social stability will be disrupted. Overall there were few explicit incentives for leaving the WISE to pursue community employment.

### **WISE Connection With the Provincial Social Services Mandate**

The agencies that supported the WISEs studied here universally reported that by creating a WISE they were furthering their agency mission and satisfying the trust placed in them by the social services funding source. Social enterprise allowed them to expand their activities and serve more people than would be possible if operating only with provincial funding.

WISEs initially created and supported through provincial funding were sometimes able to secure outside loans to grow the enterprise. However, provincial funding was critical to all as a means of providing the salaries for key staff who provided direction and leadership and facilitated sustainability. Also critical was support from provincially funded small business development agencies, well-sourced boards of directors, input from university researchers and mentoring from existing WISEs.

All Ontario WISEs used provincial funding for at least one key staff person (e.g., manager or

director) or a portion of the workload of upper management agency staff (e.g., director of programming, chief financial officer, marketing and outreach manager) who supported the WISE in addition to their other responsibilities. A portion of support worker salaries was also paid through agency funds for job coaching, supervision, or movement to supported employment. All agencies had evolved their bookkeeping to delineate the resources used by the WISE, including staff and space, and to run separate balance sheets for their financial operations.

There were challenges to working within the system, particularly in Ontario, where most of the WISEs existed in a space that lay between social agency and competitive business. Thus, the employer-employee relationship was complicated by the need to maintain individual support plans for workers, and to hire through the provincial program admission system. There were other scenarios, based on the form and history of incorporation, where the WISE businesses were seen as a separate entity operated at arm's length from a social service agency, and the persons with IDD were seen as workers only. This was clearly the case in Alberta, where the WISE could hire workers into any of their businesses using a varied recruitment system of agency referrals and community job postings, and an informal hiring system to maintain a desired mix of people with IDD and others.

## **Discussion**

In the IDD sector, the competitive employment market is becoming increasingly difficult to navigate, given the changing nature of job demands, the shift to non-permanent work, and increasing socio-economic demands on employers (Santilli et al., 2014). Yet work remains a primary context for skill development, socialization, and establishment of an adult identity and self esteem (Santilli et al., 2014). WISEs provide one option for meaningful work involvement and job preparation. This study provided the opportunity to unpack some of the successes, challenges and tensions associated with WISEs in the IDD sector. The emergent data provided a number of insights relative to our research questions, and suggested directions for further development.

### **What conceptual models/philosophies have guided WISE in the IDD sector?**

A core feature of the WISEs in this study was their belief in the value of and the right to work for people with IDD. Tensions arose when WISEs were forced to exclude some workers in an effort to operate like a business, with minimum expectations for worker performance. Such situations created major turning points for some WISEs, but helped them move towards a business orientation. WISEs also espoused the need to empower workers. Some focused on empowerment by building work skills and confidence, while others developed meaningful strategies to incorporate worker voice and valorization through their evaluative processes, boards and structured worker meetings.

In most of these cases, the WISE was seen as a stepping stone to community based supported employment, or a hybrid that created the conditions for successful work, and provided supports from within. In one case the WISE emerged from an initiative developed by a person with IDD and evolved as an entrepreneurial worker cooperative. While supported employment was endorsed by the WISEs, the model was not seen as feasible in rural communities where jobs are rare and need to be created, or for workers with exceptional challenges in meeting competitive standards.

### **What factors have contributed to promoting and defining WISE in the IDD sector? What policies and structures contribute to or detract from the establishment of WISEs?**

Provincial funding has been foundational to the success of all the WISEs studied. Flow-through funding supported salaries for managerial positions, support workers, and in some cases, worker stipends. The need to serve more constituents was a key driver behind implementation of these WISEs. Critical funds for equipment purchase or real estate acquisition derived from external foundations and loans were leveraged on the existing infrastructure and stability gained through the provincial funding support.

A central feature that dominates discussion in this arena is the approach to compensation.

Most of the WISEs studied had a commitment to paying workers at least minimum wage, aligned with principles of normalization (Wolfensberger, 2011) and the social value placed on the labour of those with IDD. This commitment comes with certain perils, as particularly in this sector, payment at minimum wage may threaten the very existence of the WISE, or the upholding of WISE goals of employing people who are the most marginalized and vulnerable. While this tension shaped the nature and direction of growth, and threatened some of the core visions of the businesses involved due to the need to move some workers out of employment streams, it also helped define the legitimacy of the businesses.

The nature of government disability benefit regulations and their role in shaping the nature of the WISE workforce also produces human resources challenges. Because many workers with disabilities and their families limit employment exposure in order to avoid disruptions to their disability income (Saunders & Nedelec, 2014), WISEs typically operate with a largely part-time workforce, and lack back-up to cover for worker absences. While the Ontario government and others have sought to remove barriers to work, it was clear in our findings that many participants and their families retained concerns about risk, and made choices that may not have supported optimal levels of work engagement. Further attention to this issue, in particular with respect to how it might uniquely manifest for people with IDD, is an important area of further study.

Finally, an ongoing tension that existed only in Ontario WISEs relates to complying with provincial social service requirements as a service provider while functioning in an employer-employee relationship with their workforce.

### **What factors have contributed to the success and survival of WISEs in this sector?**

Universally, the nature and growth of the WISEs was based in opportunism – with a visionary leader or grassroots manager seeing an opportunity to serve a market niche or to purchase or bid on a local business or real estate opportunity. Tolerance for risk was a core feature of some of the larger WISEs. The cases

studied revealed the importance of WISEs nurturing local resources, connections and community support as they considered possible ventures and went through the challenges of creating and growing their businesses. In Alberta, the provincial decision to assign bottle return/recycling facilities to community agencies as a revenue source is a major support to the WISE, while in Ontario the local business case for a recycling firm led to municipal support for a WISE venture. Overall, government policy appeared to have a major role in decisions made by WISEs, and the direction of their growth, development, and in some cases, failure.

For a WISE to succeed, a sustainable business plan and sound operational procedures are essential (Cooney et al., 2016; Kidd & McKenzie, 2014). All WISEs in this study noted the importance of securing business advice. The presence of skilled trades people as supervisors in some businesses both enhanced the quality and marketability of the product or service being delivered, and contributed to the perception of the positions as real work.

### **What strategies promote choice, independence and social inclusion for workers? What does social inclusion look like in these varied contexts?**

There are a number of ways that people can achieve inclusion that relate to the employment context itself, including doing work that fits one's goals and preferences, feeling competence in the work role, and having the sense that one is contributing to a greater good (Lysaght & Cobigo, 2014). There are also explicit strategies demonstrated through this study that represent different forms of inclusion through WISE, such as creating an integrated workforce, structuring the work so as to ensure community contact, and building a socially desirable product or service that feeds the sense of pride and contribution in workers. A taxonomy of social participation that has been advanced by Levasseur, Richard, Gauvin, and Raymond (2010) proposes a continuum by which inclusion strategies might be interpreted, progressing from doing activities that prepare one for being with others, to presence in a community without direct interaction, and ultimately to

shared experience, collaboration and contribution. The strategies to encourage social inclusion for workers that were employed by the firms in this study spanned the full continuum described in this model, often including more than one approach.

## **Conclusions and Recommendations**

Based on the findings of this study, the following findings are of note:

- WISEs can serve as a valuable means for IDD agencies to support their missions. By leveraging the agency resources (equipment, expertise) supported through provincial funding, agencies are creating WISEs to provide opportunities for greater numbers of citizens with IDD, while advancing the social inclusion mandate beyond what would be possible if entrepreneurial strategies were not used.
- Guidance and support are needed to assist organizations who wish to undertake this approach to employment. This may be in the form of seed funding, legal counsel, assistance in making important choices about business models, ongoing business advice, and guidance as to how transfer funds can legally and reasonably be used. The sector should consider how to grow this knowledge base if WISEs are to be encouraged as a viable option.
- While guidelines and standards are important to ensure that WISEs satisfy best practices, it is important that they have opportunity for innovation, creativity and flexibility so that new business ventures respond to the local environment and specific missions and policies that govern WISE development.
- Evidence suggests the need for a *business orientation* rather than a *service orientation* in the way WISEs operate. Strategies for providing support while allowing WISEs to make sound business decisions, take risks, and do independent worker recruitment are vital.
- Finally, it is critical that ongoing evaluation of the varying business models and approaches to compensation be studied, along with stud-

ies of worker outcomes. This evidence base is necessary to build support for WISE, and to guide ongoing development such that WISEs effectively promote engagement and social inclusion for adults with IDD.

## Key Messages From This Article

**People with disabilities.** Working in a social enterprise can be a good choice for many people with disabilities. In choosing to work in a social enterprise, it is important to find a good match for your skills and interests, fair working conditions, the amount of support you need, and plenty of opportunity for continued career growth.

**Professionals.** WISE success requires incorporation of sound business principles, willingness to take on some level of risk, and use of a business (rather than social service) orientation

**Policymakers.** WISEs are a potentially valuable strategy for helping agencies move more people into employment. Evidence shows that use of explicit strategies can build social inclusion through WISE.

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## Building Employers' Capacity to Support Vocational Opportunities for Adults With Developmental Disabilities: A Synthesis Review of Grey Literature

### Abstract

*Although a significant body of literature has focused on employment supports, knowledge of beneficial components of vocational services for adults with developmental disability (DD) from the perspective of employers is lacking. To our knowledge there is no synthesis review of grey literature focused on services, supports and strategies that foster employers' capacity to support employment for adults with DD, including autism spectrum disorder (ASD). This review builds on our review of peer-reviewed literature in this area, and may reveal information and ideas not reported in academic materials. Accommodation in the workplace was a main theme that emerged in numerous documents included in this review. In addition, employment support and job coaching was identified as vital to supporting employment for people with ASD and DD. Education, awareness training and recruitment also emerged as main themes in the documents. The evidence from this synthesis review provides a strong platform to understand the literature focused on services, supports and strategies that foster employers' capacity to support employment for adults with ASD and DD, including information broadly available outside of academic contexts.*

Research has emphasized several approaches to improving opportunities for valued and meaningful work for individuals with developmental disability (DD). Approaches consisted of partnerships between employers and employment service providers, and networking opportunities between employers (Crawford, 2012; Human Resources Development Canada, 2002). In a study done in the United States (U.S.), effective practices to promote integrated employment included building collaborations and partnerships between employers and DD agencies capacity through information sharing, open communication, ongoing training, and technical assistance (Timmons, Bose, & Hall, 2013). A recent U.S. review of practices at vocational rehabilitation agencies identified promising practices for vocational rehabilitation, including using a standardized process to refer individuals with DD to vocational rehabilitation, state legislature to increase funding for long-term supported employment and decrease wait lists, and inter-agency collaboration (Burns, Haines, Porter, Boeltzig-Brown, & Foley, 2013).

Job consultants who serve individuals with DD employ a greater percentage of strategies in areas related to career

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planning and assessment, and facilitating transition after hire as compared to job search strategies (Migloire, Butterworth, Nord, Cox, & Gelb, 2012). Recently, the Canadian Association for Supported Employment (2014) and its partners identified nine best practices for employing people with DD, including: choice and control, paid employment, partnership, full inclusion, job search, individualized, natural supports, long term support, and continuous quality improvement.

Although a significant body of literature has focused on employment supports, knowledge of beneficial components of vocational services for adults with DD from the perspective of employers is lacking (Nicholas & Roberts, 2012; Richards, 2012; Van Wieren, Reid, & McMahon, 2008). It is important to consider the perspective of employers, who have the ultimate control over hiring individuals with DD. We recently completed a knowledge synthesis of peer-reviewed literature in this area (Rashid, Hodgetts, & Nicholas, 2017), which revealed that relatively limited research has focused on employer perspectives. A significant amount of information is now available online. As such, for this review we have specifically focused on exploring grey literature, which may expose us to a wide range of information and new ideas that may not be reported in published materials (Simkhada, Knight, Reid, Wyness & Mandava, 2004). Grey literature is mainly comprised of unpublished documents, such as conference proceedings, dissertations, bibliographies, and government reports/documents (Alberani, Pietrangeli, & Mazza, 1990).

## Methods

### Search Strategies

A Google Scholar search was conducted with the assistance of a research librarian to identify documents focused on services, supports and strategies to help build employers' capacity to support vocational opportunities for adults with DD and/or autism spectrum disorder (ASD). Based on the literature we found in our synthesis review of peer-reviewed research, we also specified epilepsy in our search strategy. The search strategy was restricted to English language, grey literature and pdf files. As

recommended by the research librarian, we initially reviewed the first 50 documents within each search to maintain consistency across our Google searches. If new information continued to emerge as we reviewed these 50 documents, we planned to review more documents until no new information emerged. However, a review of results past the first 50 within each search was not required because no new information was emerging.

### Inclusion/Exclusion of Studies

This review focused on services, supports and strategies to help build employers' capacity to support vocational opportunities for adults with DD, ASD and epilepsy (hereafter referred to collectively as DD, unless findings were specific to a diagnostic group). We included all documents that met the following inclusion criteria: (1) focused on supporting individuals with DD, (2) considered employers and/or colleagues' perspectives, (3) written in English, and (4) consisted of grey literature, including personal websites and blogs. One reviewer screened the documents retrieved from the search. The full texts of all the relevant documents were retrieved for review. Documents were grouped into three categories: include, exclude, or unsure. Both reviewers (MR & SH) met to review the full texts for the "unsure articles" with a final decision to either include or exclude each article.

### Data Extraction and Analysis

A standardized data extraction form was used to extract data from all included documents. The main components of the form were study publication information (e.g., year of publication, country) and findings. One reviewer (MR) extracted data and a second reviewer (SH) checked the entries to make sure that the content extracted was accurate. Extraction discrepancies were resolved by consensus. Descriptive study information and detailed findings from each document are presented in Table 1. A thematic analysis was conducted, which revealed four emergent themes from the documents. These are summarized below.

## Results

### Description of Included Studies

The electronic Google search yielded 150 potentially relevant documents for screening. After title and abstract review (when present), all 150 documents were selected for full review with 19 documents meeting the inclusion criteria after full-text review. General study characteristics are presented in Table 1. The majority of documents ( $n = 9$ ) were produced in the

United States, five documents were produced in Canada, three documents were produced in the United Kingdom (UK), and two documents contained information related to the European Union. Although not necessarily evident based on document titles in Table 1, the review revealed that employee diagnosis varied across the included documents, with 12 documents focusing on unclassified disabilities, six documents focusing on ASD, and one document focusing on epilepsy.

Table 1. Findings From the 19 Grey Area Resources Included in the Study

<i>Authors (Year); Location</i>	Owens (2010); USA
<i>Source</i>	HR Magazine
<i>Title</i>	Hiring employees with autism: as more people with autism enter the workforce, employers are tapping a new source of talent
<i>Salient Findings</i>	<p>The document focuses on discussing Ken's Kids, Inc. (now Ken's Krew) in Home Depot stores.</p> <p>What is needed for employment programs to be successful:</p> <p>(1) support from store management and store associates; (2) consistent support from job coaches; (3) sensitivity training for store managers.</p> <p>The Home Depot's district managers conduct orientation and training so store managers can uphold company policies and explain procedures to new associates.</p> <p>Ken's Kids job coaches are paid by the program. After 3 months, the amount of coaching starts to gradually decrease, but coaches continue to conduct spot checks on workers every 2 to 6 weeks, depending on the individual.</p>
<i>Authors (Year); Location</i>	Kurtz & Jordan (2008); Canada
<i>Source</i>	Institute for Community Inclusion
<i>Title</i>	Supporting individuals with Autism Spectrum Disorders: Quality employment practices.
<i>Salient Findings</i>	<p>Employment specialists assist employers with job training, fine-tuning job supports and accommodations, and answering questions that arise from supervisors or coworkers. They can also serve as a liaison between work and home to maximize communication between the employee and employer.</p> <p>Employment staff can help employers provide customized supports for workers who might need extra attention around predictability and routine. Some effective strategies and resources include a visual schedule, the use of personal digital assistants, reviewing a schedule with the employee regularly, discussing changes in routine or personnel in advance, and extra support during periods of transition or personnel changes.</p>

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Table 1. Findings From the 19 Grey Area Resources Included in the Study (continued)

<i>Authors (Year); Location</i>	Iyer & Masling (2015); USA
<i>Source</i>	Curb cuts to the middle class initiative
<i>Title</i>	Recruiting, hiring, retaining, and promoting people with disabilities
<i>Salient Findings</i>	<p>This resource guide includes a list of federal agencies working together to ensure employers have the tools and resources they need to recruit, hire, retain, and promote people with disabilities.</p> <p>The following strategies and resources were listed to assist employers in the process of hiring and retaining employees with disabilities: local American job centres, forming community partnerships, disability awareness training, workplace mentoring programs, employee resource groups, job coaches, and employee onboarding programs.</p>
<i>Authors (Year); Location</i>	North East Community Partners for Inclusion (2005); Canada
<i>Source</i>	*
<i>Title</i>	Guide to hiring persons with disabilities for Saskatchewan employers
<i>Salient Findings</i>	<p>This document provided strategies and resources for hiring persons with disabilities. Co-worker education, supported employment agencies, job coaching, disability awareness training for staff, on-the-job training, and job carving were listed as employment supports and strategies for employers.</p> <p>The guide suggested co-worker education should comprise disability sensitivity training and information on how to support co-workers with disabilities.</p> <p>Provincial supportive employment agencies in Saskatchewan offer funding for employment supports, including job coaches, while federal programs offer funding to cover costs such as wages and other related employer expenses.</p>
<i>Authors (Year); Location</i>	Martin et al. (2014); USA
<i>Source</i>	Institute for Corporate Productivity (i4cp)
<i>Title</i>	Employing people with intellectual and developmental disabilities
<i>Salient Findings</i>	<p>Introductory work programs, which are designed to introduce workers with intellectual and developmental disabilities to the workplace, were positively reviewed by the majority of surveyed respondents. These programs provide organizations with on-location work trials, job shadowing, internships, and/or long-term work exchanges.</p> <p>Employment support agencies can provide a range of services to assist employers, including transportation, on-the-job support, pre-employment training, job carving, and information on workplace accommodations.</p>

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Table 1. Findings From the 19 Grey Area Resources Included in the Study (continued)

<i>Authors (Year); Location</i>	Fredeen et al. (2012); Canada
<i>Source</i>	Labour market opportunities for persons with disabilities
<i>Title</i>	Rethinking disability in the private sector
<i>Salient Findings</i>	<p>This document discussed employment support strategies for private organizations.</p> <p>Disability awareness training for staff and collaboration with community partners were determined to be valuable strategies for employers looking to support staff members with disabilities.</p> <p>Community partners can assist employers with post-placement support where support can be for the employer, the employee, or both. The duration of their services varies, depending on the needs of the employer and employee.</p> <p>Once a candidate is hired, community partners can also assist with onboarding and training activities. These services include job coaching, advice concerning accommodations and information on programs and resources useful for both employer and the employee.</p>
<i>Authors (Year); Location</i>	Dunst et al. (2015); USA
<i>Source</i>	Kansas University Center on Developmental Disabilities and the Beach Center on Disability
<i>Title</i>	What every employer needs to know: Key success factors for hiring people with disabilities.
<i>Salient Findings</i>	<p>This document discusses the importance of collaboration between employers/HR staff and employees during the workplace accommodation process.</p> <p>This document also highlights an employment support outcome measure entitled the Self-Determined Career Development Model (SDCDM). The SDCDM is specifically designed for use by adolescents and adults with disabilities, to support job and career-related goal setting and attainment and promote more positive employment and career development outcomes. A facilitator is integral to model implementation. It is critical for people with disabilities to be able to communicate about their needed accommodations, but also to have employers who are willing to work collaboratively and in partnership.</p>

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Table 1. Findings From the 19 Grey Area Resources Included in the Study (continued)

<i>Authors (Year); Location</i>	Katz et al. (2012); USA
<i>Source</i>	Disability and Work
<i>Title</i>	Strategies to support employer-driven initiatives to recruit and retain employees with disabilities
<i>Salient Findings</i>	<p>This document discusses three employment support programs for persons with disabilities: (1) Bridges to Bridges; (2) Connecticut Industry-Specific Training and Placement Program; (3) Project SEARCH</p> <p><i>Bridges to Bridges</i> acts as an intermediary business between employers and service providers. The program provides employers with evaluations of, and recommendations for, local service provider organizations. Through this program, employers are able to access a pool of quality, pre-screened employees with disabilities to meet their workforce needs without continually navigating the public and private social services and workforce systems for employee sourcing.</p> <p><i>Connecticut Industry-Specific Training and Placement Program</i> offers grants to Community Rehabilitation Provider agencies to partner with a major employer on a disability employment initiative. Grant funding comes from Connecticut's allocation of federal vocational rehabilitation funds. The grants are for two years and the amount of each grant is based on the projected number of people employed.</p> <p><i>Project SEARCH</i> offers supervisors training on the program model and common disability employment needs.</p>
<i>Authors (Year); Location</i>	Alberta Works (2014); Canada
<i>Source</i>	*
<i>Title</i>	Recruiting and employing persons with disabilities.
<i>Salient Findings</i>	Community partners are an excellent resource as they understand business' talent needs and can help employers recruit and support talented people with disabilities.
<i>Authors (Year); Location</i>	Chénier & Vellone (2012); Canada
<i>Source</i>	The Conference Board of Canada
<i>Title</i>	Employers' toolkit: Making Ontario workplaces accessible to people with disabilities.
<i>Salient Findings</i>	<p>Community organizations can assist employers by providing expertise related to accessibility, accommodations, or training (e.g., disability awareness/sensitivity training), candidate pre-screening, job coaching, pre-employment training, and information on best practices in recruitment and retention of people with disabilities at little or no cost to the employer. These community-based organizations include recruitment centres, community living centres, and employer networks.</p> <p>Umbrella employment support organizations such as Job Opportunity Information Network (JOIN), connect employers with appropriate community agencies to support the process of hiring and employing people with disabilities. In doing so, the organization assists employers with the following: (1) accessing job candidates; (2) connecting with other employers to hear the benefits of hiring people with disabilities; (3) mentoring job-ready people with disabilities.</p>

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Table 1. Findings From the 19 Grey Area Resources Included in the Study (continued)

<i>Authors (Year); Location</i>	European Commission (2012); Europe
<i>Source</i>	*
<i>Title</i>	Supported employment for people with disabilities in the EU and EFTA-EEA good practices and recommendations in support of a flexicurity approach.
<i>Salient Findings</i>	<p>This document focuses on vital elements such as job coaching, disability training for staff, and long-term on-the-job support as effective strategies for supporting employment for people with disabilities.</p> <p>Job coaches were considered a key support for employers.</p> <p>Recommendations for employers hiring a job coach include providing the support worker with an adequate caseload, reasonable remuneration, quality training, and knowledge of employer competency requirements.</p>
<i>Authors (Year); Location</i>	The National Autistic Society (2011); United Kingdom
<i>Source</i>	*
<i>Title</i>	Employing people with autism: A brief guide for employers
<i>Salient Findings</i>	<p>This article suggests that appointing a mentor, collaborating with employment support agencies, employing a job coach, and arranging ASD awareness training for staff are viable methods for supporting employees with ASD and their employer.</p> <p>The role of employment service advisors in providing information on schemes and job programs were also discussed. These services can provide employers and employees with wage subsidies, workplace supports, interview support, advice on accommodations, assistance with transportation, adaptive aids, and employment support staff.</p>
<i>Authors (Year); Location</i>	Westbrook et al. (2012); USA
<i>Source</i>	Campbell Systematic Reviews
<i>Title</i>	Adult employment assistance services for persons with autism spectrum disorders: Effects on employment outcomes
<i>Salient Findings</i>	<p>This document highlights the importance of job coaching. Specifically, the use of behavioural techniques such as functional behavioural assessment, response cost procedure, positive reinforcement, social skills training, prompt fading, task analyses, and task preference assessments were reported as effective job supports.</p> <p>Natural supports, such as the use of co-workers to provide on-site training, social skills training, or transportation, have also been found to be effective in reducing the amount of training provided by the employment specialist.</p> <p>Long-term vocational support services were critical for sustaining employment and should involve both employees and employers.</p> <p>Though short-term vocational supports were important in helping individuals with ASD learn how to perform work tasks, long-term supports were needed for people with ASD to sustain employment and adapt to changes in the work.</p>

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Table 1. Findings From the 19 Grey Area Resources Included in the Study (continued)

<i>Authors (Year); Location</i>	European Commission (2014); Europe
<i>Source</i>	*
<i>Title</i>	Results of four pilot projects on employment of persons with autism
<i>Salient Findings</i>	<p>Informing and training colleagues in the workplace as well as concerned persons in a wider environment of the private and public sector are important for successful integration of a person with ASD in workplace. The documents also discussed methods of raising awareness, which included workshops, individual meetings and trainings of managers and mentors, producing guidelines for work with persons with ASD, and contacts with employers to ensure consistency between business goals and corporate social responsibility initiatives.</p> <p>Job coaches and other experts, such as psychologists, special pedagogues, speech therapists etc., can assist employers with providing workplace accommodations.</p>
<i>Authors (Year); Location</i>	The National Autistic Society (2004); United Kingdom
<i>Source</i>	*
<i>Title</i>	The undiscovered workforce
<i>Salient Findings</i>	<p>There are financial and practical support programs in the UK to assist employers who hire individuals with ASD. Government supported agencies provide employers with information on financial assistance and incentives as well as any support which is available in the local area.</p> <p>Employment agencies can assist employers by providing on-the-job support to employees and offering professional advice, support, and training to managers and staff. Agencies can also help employers maximize the skills of employees with an ASD by assessing the work environment, advising on any adaptations or adjustments, and advising on the recruitment process. Agencies can assist employers by setting up work experience placements, job tasters (i.e., &gt; 6 month job trials), short-term contracts, and permanent contracts.</p>
<i>Authors (Year); Location</i>	Epilepsy Action (2013); United Kingdom
<i>Source</i>	*
<i>Title</i>	Work and epilepsy
<i>Salient Findings</i>	<p>This document addresses the issue of reasonable adjustment or accommodation that is needed to be done while employing a person with epilepsy. This will ensure that the employee with epilepsy is not disadvantaged compared to other employees.</p> <p>Services such as Disability Employment Advisers provide a range of support, advice and information to people with disabilities. They can offer help if people are looking for a job, looking for training or need help to keep a job.</p>

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Table 1. Findings From the 19 Grey Area Resources Included in the Study (continued)

<i>Authors (Year); Location</i>	Office of Disability Employment Policy (2008); USA
<i>Source</i>	*
<i>Title</i>	Survey of employer perspectives on the employment of people with disabilities
<i>Salient Findings</i>	<p>This document focuses on some of the challenges employers faced while hiring people with disability and services available for people with disabilities.</p> <p>One-Stop Career Centers are operated by state and local agencies and are designed to provide a full range of assistance to job seekers and employers in one location. The centers offer training referrals, career counselling, job listings, and other employment-related services.</p> <p>The Job Accommodation Network (JAN) was another service discussed in this document. Its mission is to facilitate the employment and retention of workers with disabilities by providing information on job accommodations, entrepreneurship, and related subjects.</p>
<i>Authors (Year); Location</i>	The Job Accommodation Network (2009); USA
<i>Source</i>	*
<i>Title</i>	Employers' practical guide to reasonable accommodation under the Americans with Disabilities Act
<i>Salient Findings</i>	<p>This document reveals that, according to the American Disability Act, an employer may be required to provide a temporary job coach to assist in the training of a qualified individual with a disability as a reasonable accommodation, barring undue hardship. An employer also may be required to allow a job coach paid by a public or private social service agency to accompany the employee at the job site as a reasonable accommodation.</p>
<i>Authors (Year); Location</i>	Fesko & Butterworth (2001); USA
<i>Source</i>	Institute for Community Inclusion
<i>Title</i>	Conversion to integrated employment: Case studies of organizational change
<i>Salient Findings</i>	<p>This document highlights the use of job coaching as an effective employment strategy for both employers and employees.</p> <p>The document also discusses strategies to make employee work experiences more meaningful, such as using a co-worker as support during conflict and switching places with a worker, allowing him or her to act as job coach.</p>

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## Theme 1: Accommodation

Job accommodation was a main theme, occurring across ten documents. The majority of documents indicated that job accommodations play a vital role in increasing productivity and the overall development of people with DD. Job accommodations are perceived to help create equal employment opportunities so individuals with disability have opportunities for the same achievements as other employees (Chénier & Vellone, 2012; The Job Accommodation Network, 2009). Two documents, published by Alberta Works (2014) and the Office of Disability Employment Policy (2008), suggested that a major challenge that employers face when hiring individuals with disability is a lack of knowledge about the financial costs associated with accommodations. In addition, employers often overlooked effective hiring strategies such as a centralized accommodation fund and reassignment (Office of Disability Employment Policy, 2008). It is vital to note that documents showed low or no costs associated with employing a person with disability. For example, Fredeen, Martin, Birch, and Wafer (2012) reported that 57% of people with disability required no accommodation, and 37% of people with disability required only one accommodation, which cost the employer approximately \$500.

Owens (2010) highlighted some of the most common requests for establishing reasonable accommodations. These included modification of testing and training materials, policies, and equipment. Another document that specifically focused on guidelines for employers hiring people with disability in Saskatchewan, Canada incorporated an entire chapter on job accommodation and its significance. This chapter provided in-depth information to employers about the value and advantages of job accommodation. The main focus of this chapter was on three main factors of job accommodation including, "modification of schedules and duties, removal of barriers through changes to the work environment, and providing supports including the purchase of assistive technology" (North East Community Partners for Inclusion, 2005, p. 26). In addition, Chénier & Vellone (2012) reinforced that it is vital that the employee's needs are heard when determining accommodations, as they have the best know-

ledge about what kind of accommodation may be most needed and beneficial in that particular organization or company.

## Theme 2: Employment Support

A document of the European Commission (2012) delineated employment support into two subgroups: Job Assistance and Individual Support at the Workplace. Eight articles included in this review discussed the effectiveness of employment support for individuals with DD (Alberta Works, 2014; Dunst, Shogren, & Wehmeyer, 2015; European Commission, 2012; Fesko & Butterworth, 2001; Iyer & Masling, 2015; North East Community Partners for Inclusion, 2005; The National Autistic Society, 2011; Westbrook et al., 2012). Employment support was viewed as a practical employment approach and associated with positive outcomes for both employees and employers (Fesko & Butterworth, 2001; North East Community Partners for Inclusion, 2005). Supported employment refers to both principles and models that promote the integration of persons with disabilities into community workplaces with competitive pay. An array of supports may be provided, such as assistance with determining job matches with skills and interests, finding a job, and maintaining a job (Rashid et al., 2017). Supported employment was cost and time effective in relation to pre-screening of potential employees (North East Community Partners for Inclusion, 2005). The English, Scottish and Northern Irish Association of Supported Employment estimated that 30,000 individuals with a disability were involved in some kind of supported employment services, and that employers prefer to hire individuals with disability through supported employment (European Commission, 2012). However, studies from other jurisdictions suggested that there was lack of knowledge about disability related employment support. For example, 63% of employers from food services, and art and entertainment industries reported that a lack of knowledge about supported employment for individuals with disability was a major challenge (Alberta Works, 2014).

The significance of job coaching for people with DD was discussed in seven documents included in this review (European Commission, 2012; Fesko & Butterworth, 2001; Fredeen et al.,

2012; Iyer & Masling, 2015; Katz, O'Connell, & Nicholas, 2012; North East Community Partners for Inclusion, 2005; Westbrook et al., 2012). Documents included in our review indicated that job coaching plays a vital role in improving outcomes related to employing individuals with disability (Iyer & Masling, 2015). Some of the documents (North East Community Partners for Inclusion, 2005) indicate that long-term employment initiatives that included job coaching play a substantial role in creating a positive relationship between employer and employee. Moreover, numerous documents suggest that agencies that are involved in providing employment services to people with disability need to be involved in providing ongoing vocational rehabilitation (job coaching) and training activities to enhance job opportunities and success (European Commission, 2012; Fredeen et al., 2012; Katz et al., 2012). Agencies that were involved in providing supports for employers and employees with disability endorsed job coaching to provide assistance well suited for the specific needs of the potential employee. Job coaching was viewed as the backbone of achieving the required performance standard (European Commission, 2012).

### Theme 3: Education and Awareness Training

Ten documents discussed the value of education and awareness training for employers, colleagues, and individuals with disability (Alberta Works, 2014; Chénier & Vellone, 2012; European Commission, 2012; Fredeen et al., 2012; Iyer & Masling, 2015; Martin, Jamrog, Lykins, & Davis, 2014; North East Community Partners for Inclusion, 2005; Office of Disability Employment Policy, 2008; The Job Accommodation Network, 2009; Westbrook et al., 2012). One document stated that disability awareness training was vital in relation to cost efficiency, added to the value of workplace inclusivity, and attracted more individuals with disability to the workplace (Iyer & Masling, 2015). Disability awareness training enabled in-depth understanding and created a sensitive environment, which increased openness and willingness to hire and accept people with disability in the workforce (North East Community Partners for Inclusion, 2005). In addition, disability education and training

was recommended to employers who have not yet employed a person with disability (Martin et al., 2014). One document showed that most companies were eager to hire people with disability. However, there was constant need to provide education and disability awareness training to employers to improve inclusion of individuals with disability in the workforce, and especially to maximize their potential (Alberta Works, 2014; Fredeen et al., 2012). The lack of education and training for people with disability has been identified as a major barrier for their employability (Alberta Works, 2014), and changing attitudes in the workforce through education and awareness training has been suggested as a way to enhance everyone's comfort (Chénier & Vellone, 2012). In fact, one report identified ASD awareness training as the most effective and successful strategy for employees with ASD (Westbrook et al., 2012). It is evident from the findings in this review that increasing disability education and awareness increases the number of companies that recruit individuals with DD, and plays a significant role in decision making related to hiring employees with disability (Office of Disability Employment Policy, 2008).

### Theme 4: Recruitment

Another theme that emerged throughout the documents addressed the recruitment of individuals with disability. This theme was evident in ten documents (Alberta Works, 2014; Chénier & Vellone, 2012; European Commission, 2012; Fredeen et al., 2012; Iyer & Masling, 2015; Katz et al., 2012; Martin et al., 2014; North East Community Partners for Inclusion, 2005; The Job Accommodation Network, 2009; The National Autistic Society, 2004). One document estimated that 40% of individuals with disability were recruited through employee referral and 52% were recruited through newspaper advertisements targeted at people with disability (North East Community Partners for Inclusion, 2005). It has also been recommended that successful recruitment may be done through collaboration with community-based partners (Iyer & Masling, 2015), or programs such as the Workforce Recruitment Program (WRP) or Skills Training Partnership (STP) (Chénier & Vellone (2012). These programs were developed to help recruit motivated young people with disability into the

labour force, and help employers support hiring employees with disability into their businesses (Chénier & Vellone, 2012; Martin et al., 2014; The Job Accommodation Network, 2009). Many companies, such as Walgreens, Lowe's, Toys R Us, and Proctor and Gamble, are actively involved in establishing connections and streamlining recruitment processes in an effort to bring more individuals with disabilities into their companies (Katz et al., 2012), suggesting increasing interest in hiring individuals with disabilities into the current labour market. Alternatively, one document included in this review stated that, despite employment opportunities, individuals with ASD are tremendously disadvantaged in the process of recruitment (The National Autistic Society, 2004).

## Discussion

This synthesis review of grey literature reveals several key findings. Accommodation in the workplace was a main theme that emerged in numerous documents. It became evident that accommodation for employing individuals with DD was perceived to be most effective in supporting overall productivity and individual growth in the workplace. In addition, employment support and job coaching were identified as vital supports that need to be part of employment for people with DD. Education, awareness training and recruitment also emerged as main themes in the documents. In addition, it is vital to note that some of our findings are in line with the results from our recent review of the peer-reviewed literature related to enhancing employer capacity to support employment of individuals with DD (Rashid, Hodgetts, & Nicholas, 2017). Existing peer-reviewed literature indicated that employers perceive disability awareness training and education played a significant role in building their capacity to supporting individuals with DD in meaningful work opportunities (Howlin, Alcock & Burkin, 2005). These findings highlight the need for further enhanced investment of available resources, supports, and services to improve vocational outcomes for individuals with DD. Our findings related to supported employment have also been acknowledged by existing peer review literature, which highlights the importance of supported employment when employing individuals with DD. For example,

Unger (1999) reported that supported employment was deemed to be the most effective strategy for employees with DD. Furthermore, we found that accommodations were vital for overall growth and success of an individual with DD in the workplace, and, consistent with the grey literature, generally associated with low costs. However, employers often have a misconception that employment accommodations for people with DD are expensive, so employers are often reluctant to employ people with DD (Hernandez et al., 2008).

As evidenced by the cohesive themes that emerged from our data, we found that the substantive content was very similar across the documents reviewed. In addition to the key themes that emerged in our results, there were some redundancies across documents that did not amount to major themes. For instance, retention of employees with DD in the workforce was documented in three reports (Chénier & Vellone, 2012; Iyer & Masling, 2015; Katz et al., 2012). Based on the similarity across reports retrieved from a variety of organizations, we suggest that one comprehensive and openly available document may be a good use of resources, instead of so many independent organizations creating their own. We believe that the creation and dissemination of a gold standard best practices guide would be ideal for employers who are hiring people with DD.

Findings from this review point to vital gaps in the current grey literature in relation to employing individuals with DD. Numerous documents have been produced by various organizations, such as government agencies working to enable employment opportunities for people with DD (Iyer & Masling, 2015), employment support perspectives (North East Community Partners for Inclusion, 2005), and the perspectives of friends and family members of individuals with disability (Fredeen et al., 2012). However, there were no documents that focused on the direct perspective of individuals with DD. Their perspectives need to be incorporated into future literature. Articulation of experiences and identification of gaps and needs based on first-hand experiences will offer employers insight that will help them develop better employment opportunities, refine or develop training programs to best meet needs, inform priorities in policy development and

employment support services. Therefore, our literature review highlights the need for future research that helps understand the lived experiences of people with DD in relation finding a job. Importantly, this research should creatively incorporate perspectives of people across the spectrum of disability, including those who are minimally verbal, those with "hidden" versus visible disability, and those with intellectual impairment. In-depth research will enable us to gain first-hand information from participants and accordingly develop policies and programs that will lead to better employment capacity for individuals with DD.

The majority of the documents reviewed in this study do not emphasize strategies for better communication amongst employers, co-workers and employees, nor the resulting positive impact of increased communication for employers and their business growth. Only one document addressed elements for effective communication strategies and communication supports for employees with DD (Chénier & Vellone, 2012). There were few documents that highlighted the importance and effectiveness of solid communication in a general manner in the workplace (Dunst et al., 2015; Fesko & Butterworth, 2001; Westbrook et al., 2012). In addition, only one document indicated that people with ASD may have difficulty communicating and often are unable to read facial expressions or other non-verbal cues (The National Autistic Society, 2004). The lack of emphasis on communication skills and lack of focus on developing communication strategies relevant to employing people with DD emerges as a gap in this literature. Hence, we recommend that more research be conducted on developing communication strategies that may be appropriate when employing individuals with DD.

## Strengths and Limitations

A main limitation of this review was that we had limited the search to English language and there may be documents in other languages that may have been missed. Despite these limitations, this review has numerous strengths. A thorough search was conducted with an experienced librarian. In addition, the search was diverse in nature as it included varied docu-

ments such as government reports, policy related documents, tool kits for employers and other informative documents related to the needs and demands of employing individuals with DD.

## Conclusions

The evidence from this synthesis review provides a strong platform to understand the accessible grey literature focused on services, supports and strategies that foster employers' capacity to support employment for adults with DD. It reinforces the value of sustained, and potentially increased, funding for programs and services that are well suited to employers' needs for hiring people with DD.

## Key Messages From This Article

**People with disabilities.** Individuals with DD and ASD will benefit from this review by gaining knowledge about what is available for them in the existing literature about vital topics such as recruitment, job coaching and employment support.

**Professionals.** This work will effectively summarize existing grey literature for professionals who are often busy to read every documents in-depth which exists about building capacity development for people with disabilities.

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## Estimating the Prevalence of Children Who Have a Developmental Disability and Live in the Province of British Columbia

Disclaimer: All inferences, opinions and conclusions drawn in this paper are those of the authors, and do not reflect the opinions or policies of the Data Steward.

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### Abstract

*Administrative health data for 1986 to 2013 was used to identify the prevalence of childhood developmental disability in British Columbia. A total of 26,320 children who have a developmental disability were found in the data set. Prevalence increased for all categories of developmental disability over the 27-year period. Following 2010, the rate of increase in prevalence for all categories of developmental disability slowed. The increase in prevalence was greatest for autism spectrum disorder. Prevalence data is important to have for planning programs and services for people who have a developmental disability.*

This study was conducted to estimate the prevalence of children who have a developmental disability in the province of British Columbia. Information on the prevalence of developmental disability is important for planning long-term policies and services. In addition, statistics are critical in monitoring the well-being of people with disabilities. As Recommendation 9 of the World Health Organization/The World Bank World Report on Disability states: "Research is essential for increasing public understanding about disability issues, informing disability policy and programmes, and efficiently allocating resources" (*World Report on Disability*, 2011, p. 267).

However, there is little information available on the prevalence of developmental disability in Canada or internationally (Fujiura, Rutkowski-Kmitta, & Owen, 2010). Moreover, using the data that do exist is hampered by the differing definitions and terms used to describe developmental disability. Terms such as intellectual disability, developmental disability, mental retardation and neurodevelopmental disability are all used in the literature and may be poorly defined. Despite these drawbacks, some prevalence data are available, providing a rough sketch of this neglected area.

In a meta-analysis of 52 population-based studies from 27 countries, Maulik, Mascarenhas, Mathers, Dua, and Saxena (2011) found that the highest prevalence of developmental disability occurred in low income countries and in child/adolescent age groups. The overall prevalence for high-in-

come countries was 0.92%, the overall prevalence for the child/adolescent population across the 52 studies was 1.83%.

Hogan, Msall, and Drew (2006) reported an overall prevalence of developmental disability in the United States of 13.9 per 1,000 in children aged 5–17 years. In Metropolitan Atlanta, Van Naarden Braun et al. (2015) found that the prevalence of eight year old children with an intellectual disability in 2010 was 13.0 per 1,000 and the prevalence of eight year old children with autism in 2010 was 15.5 per 1,000. Maenner et al. (2016) used the U.S. National Survey of Children's Health (NSCH) and the National Health Interview Survey (NHIS) to determine the prevalence of intellectual disability among children. They found that prevalence varied from 5.7 per 1,000 (NHIS) and 5.9 per 1,000 (NSCH) for 2- to 5-year-olds to 15.0 per 1,000 (NSCH) and 15.9 per 1,000 (NHIS) for 14- to 17-year-olds.

There is evidence that the number of children diagnosed with a disability and particularly with a developmental disability is increasing in developed countries (de Graaf et al., 2011; Halfon, Houtrow, Larson, & Newacheck, 2012; Houtrow, Larson, Olson, Newacheck, & Halfon, 2014; Reichman, Corman, & Noonan, 2008; Shin et al., 2009). Houtrow et al. (2014) analyzed the U.S. National Health Interview Survey datasets from 2001 to 2011. They found that overall the prevalence of childhood disability increased by 15.6% between 2001 and 2011. However, in the same ten year period, they also found that the percentage of disability cases due to a physical health condition declined by 11.8% while the percentage of cases due to neurodevelopmental or mental health conditions increased by 20.9%.

In Canada, there is very little information on the prevalence of developmental disability. Over the five year period of 1998 to 2003, Ouellette-Kuntz et al. (2009) estimated a prevalence of intellectual disability in Manitoba of 4.7 per 1,000 population. They found a prevalence of 11.1 per 1,000 for children aged 10–14 years and 10.9 per 1,000 for children aged 5–9 years old. Bradley, Thompson, and Bryson (2002) found a 7.18 per 1,000 prevalence of mental retardation among teenagers living in the Niagara region of Ontario. In 2009/10, administrative data from Ontario were used to obtain

an estimate of 0.78% of the population as adults with a developmental disability (Lunsky, Klein-Geltink, & Yates, 2013).

The prevalence of children with a developmental disability in British Columbia is unknown. Crude estimates of the number of people with a developmental disability in British Columbia can be assumed from service information from both the B.C. Ministry of Education (for children) and Community Living B.C. (for adults). B.C. Ministry of Education data for 2005/2006 indicate that there were 2,457 students with a moderate to severe/profound intellectual disability; 2,593 students with autism; 2,751 students with a mild intellectual disability; and 16,702 students with a learning disability enrolled in public schools (B.C. Ministry of Education, 2006). This is a total of 24,504 students (4.3% of the total number of students) with some level of learning difficulties enrolled in public schools in 2005/2006 in British Columbia. This does not include students enrolled in private schools, home-schooled or in the public system but without an assessment or identification and therefore likely underestimates the total number of school-aged children who have a developmental disability.

In British Columbia, Community Living B.C. currently provides services to over 20,000 adults with a developmental disability (Community Living B.C., n.d.). This number represents approximately 0.53% of the adult population of British Columbia in 2016. However, Community Living B.C. most likely does not provide services to all adults who have a developmental disability in British Columbia, so this estimate is also likely an underestimate of the actual prevalence.

One under-used tool for obtaining estimates of prevalence is administrative health databases (Lin et al., 2013; Lin et al., 2014). Administrative data have some unique advantages. The most significant advantage is the size of the data set (Jutte, Roos, & Brownell, 2011). The availability and large size of the data sets makes the use of administrative data very cost effective; improves the generalizability of the findings; reduces problems associated with selection bias (Jutte et al., 2011); and provides the ability to select varying comparison groups (Glasson & Hussain, 2008).

This study used health administrative data from the province of British Columbia to estimate the prevalence of children with developmental disabilities. This information will be useful for future planning of programs and services within British Columbia, and may serve as an estimate for other provinces and territories within Canada.

## Materials and Methods

As part of a larger population level study of the health of parents and siblings of children with developmental disabilities, data were collected on the prevalence of children who have a developmental disability in British Columbia. Data were obtained from Population Data B.C. (Population Data B.C., 2018).

For this study three data bases were linked. Linkages were done by Population Data B.C. staff and provided to the researchers as data extracts developed in accordance with the request of the researchers and the agreement of the B.C. Ministry of Health and Population Data B.C. ("Data Linkage," 2013) The researchers were allowed access to the data extracts only via the Population Data B.C. Secure Research Environment. The data extracts cannot be removed from that environment and are only available to researchers for a time limited period (Pencarrick-Hertzman, Meagher, & McGrail, 2013). The linked data files for this study were:

1. A central consolidation file providing demographic information on all individuals in British Columbia. The file contains information on birth dates and sex of each individual in the study as well as neighbourhood income deciles and quintiles. ("British Columbia Ministry of Health, (1986–2014): Consolidation File (MSP Registration and Premium Billing). Population Data B.C. Data Extract. B.C. Ministry of Health," 2016).
2. The Medical Services Plan (MSP) payment file that contains administrative information for all fee-for-service care provided by physicians in British Columbia. The file includes the date of each visit to a physician, the diagnostic code (ICD-9), the Health Authority and Health Service Delivery Areas where the visit occurred, and the subsidy code indicat-

ing whether payments to the physician were subsidized through provincial programs and the amount of the subsidy ("British Columbia Ministry of Health, (1985–2014): Medical Services Plan (MSP). Population Data B.C. Data Extract. B.C. Ministry of Health," 2016).

3. The hospital separation file with information on all hospitalizations, including the date of admission and discharge and the diagnostic codes (ICD-9 and ICD-10) ("British Columbia Ministry of Health, (1985–2014): Discharge Abstract Database (Hospital Separations). Population Data B.C. Data Extract. B.C. Ministry of Health," 2016).

Data were linked using unique and study specific codes that allow people to be anonymously identified across databases. The B.C. Ministry of Health approved access to and use of the data through Population Data B.C. Ethics approval was granted by the University of Victoria Human Research Ethics Board.

The resulting data extracts provided information on number of people in British Columbia aged 0–19 years old at any time between 1985 and 2014 who had received a diagnosis of a developmental disability. This diagnosis was either a primary diagnosis when these children used medical services or a secondary diagnosis (i.e., a child could be visiting a doctor for treatment for a common childhood illness and the doctor also noted developmental disability as a secondary diagnosis).

Identification of children with a developmental disability used the algorithm developed by Lin et al. (2013). Children aged 0–19 were identified by ICD-9 codes in MSP files and ICD-10 codes in hospital separation files. Identification required at least two occurrences of the ICD-9 codes identifying developmental disability in MSP data, or at least one occurrence of developmental disability identified by ICD-9 or ICD-10 codes in hospital separation data between 1985 and 2014. Data were collected according to ICD-9 and ICD-10 codes on a range of developmental disabilities including fetal alcohol syndrome (FAS), autism spectrum disorder (ASD), Down syndrome and rarer chromosomal abnormalities. This paper used the term fetal alcohol syndrome rather than fetal alcohol spectrum disorder because the ICD-9 and ICD-10 codes are for

fetal alcohol syndrome. There are no codes for fetal alcohol spectrum disorder.

For this study, prevalence was operationalized as proportion of children (defined as aged 0–19) with a developmental disability in British Columbia in each year of the data for the years 1986 to 2013. Thus, a person could have been born in 1970, but would still be a child in the years 1986, 1987, 1988 and 1989. Prevalence information for 1985 and 2014 was omitted due to the fact that data was only available for a portion of each of those two years.

Data from the data extract were also examined for sex of the child, income quintile at birth, and health authority at birth. For descriptive statistics, children with a developmental disability were compared to a cohort of children born between 1990 and 1995 who did not have a developmental disability.

## Results

A total of 26,320 children who have a developmental disability were found in the data set. Developmental disabilities were categorized into types (Table 1) according to the ICD-9 and ICD-10 codes used.

Following the initial identification of a developmental disability diagnosis, individuals were

grouped according to a single diagnosis. For example, individuals with a dual diagnosis of Down syndrome and ASD were categorized with Down syndrome and removed from the ASD group, and individuals categorized as Down syndrome and mild mental retardation were categorized with Down syndrome and removed from the Mild group. The following four broad diagnostic groups were formed: autism spectrum disorder (ASD), fetal alcohol syndrome (FAS), Down syndrome and other developmental disabilities (Table 2, following page).

Over half of children with a developmental disability were diagnosed with autism spectrum disorder (58.19%).

Following sorting into diagnostic groups, the data were further analyzed to obtain information on the prevalence of each category of developmental disability for the years 1986–2013 (Table 3, page 51). Prevalence of children with a developmental disability was calculated as a percent of the total number of children in British Columbia (“Provinces age sex population totals,” n.d.).

In 2013 the prevalence of children with a developmental disability in British Columbia was estimated at 2.42% of the total population of children in British Columbia. This number is greater than prevalence estimates reported previously in the literature for either the United

*Table 1. Total Number of Children With a Particular Developmental Disability Diagnosis 1985–2014*

<i>Type of Developmental Disability</i>	<i>Total Number</i>
Autism Spectrum Disorder	15,533
Down Syndrome	2,370
Dual Diagnosis of Down Syndrome and Autism Spectrum Disorder	218
Dual Diagnosis of Down Syndrome and Fetal Alcohol Syndrome	7
Mild Mental Retardation	3,511
Moderate Mental Retardation	869
Severe Mental Retardation	884
Profound Mental Retardation	311
Unspecified Mental Retardation	4,381
Chromosomal Abnormalities (excluding Down syndrome)	1,880
Fetal Alcohol Spectrum Disorder	1,844

*Table 2. Grouped Categories and Numbers of Children Who Have a Developmental Disability in British Columbia 1985–2014*

<i>Category of Developmental Disability</i>	<i>Number of Children Identified</i>
Autism Spectrum Disorder	15,315
Down Syndrome	2,370
Fetal Alcohol Syndrome	1,837
Other Developmental Disabilities (including chromosomal abnormalities but excluding Down syndrome)	6,798

States or Canada. Similar to studies from the United States, prevalence of the total number of children with a developmental disability in British Columbia increased over time. The increase was from 0.44% in 1986 to 2.42% of the total number of children in British Columbia in 2013. The range of percent increase each year for children with a developmental disability was 0.01–0.10. The smallest increases occurred in the years 2010–2013, indicating a recent slowing in the increase in the number of children with a developmental disability in British Columbia.

Prevalence of each of the four categories of developmental disability also increased with time. The largest increase was for children with ASD. The prevalence of children with ASD increased from 0.14% in 1986 to 1.49% in 2013. The smallest increase over time was for children with Down syndrome.

Table 4 (see page 53) shows information regarding some descriptive statistics for each of the four categories of children with a developmental disability compared to children born in 1990–1995 who did not have a developmental disability. Significant differences occurred in the number of males and females between the four disability groups and the group without developmental disabilities. Each of the disability groups had higher numbers of males. The difference was particularly great for individuals with ASD (78% of children with ASD were male).

Using the year of birth (not the year of diagnosis), additional comparisons were made between the children with a developmental disability and the children without a developmental disability regarding income and location within

British Columbia (Health Authority) at birth. Income and Health Authority data were not available for all of the children with a developmental disability, indicating that either the children were not born in British Columbia or data was missing for some other reasons. Data were available for 72% (income quintile at birth) and 74% (Health Authority at birth) of children with ASD, 65% (income quintile at birth) and 68% (Health Authority) of children with Down syndrome, 63% (income quintile at birth) and 69% (Health Authority) of children with FAS and 49% (income quintile at birth) and 53% (Health Authority) of children within the Other category.

Significant differences were found in income quintiles at birth between the disability groups and the comparison group. This difference was particularly large for individuals with FAS (45% of children with FAS were in the lowest income quintile at birth).

There were also significant differences in place of birth between the groups, again, particularly evident for individuals with FAS. Thirty-three percent of children with FAS were born in the Northern Health Authority, while only approximately 6.3% of the provincial population lives in that geographic region (“Northern Health,” n.d.). When Health Service Delivery Areas (HSDAs) were examined, it was found that 26% of children with FAS were born in the Northern Interior HSDA.

## Discussion

Based upon population level administrative data, this study provided information on the prevalence of children who have a develop-

Table 3. Prevalence of Children (aged 0–19) With a Developmental Disability in British Columbia 1986–2013

Year	Autism Spectrum Disorder (percent of total children in B.C.)	Down Syndrome (percent of total children in B.C.)	Fetal Alcohol Syndrome (percent of total children in B.C.)	Other Developmental Disability (percent of total children in B.C.)	Total Number of Children with a DD (percent of total children in B.C.)	Total Number of Children in B.C.	Absolute Percent Increase in Children With a DD (year over year)
1986	1,139 (0.14)	512 (0.06)	104 (0.01)	1,884 (0.23)	3,639 (0.44)	823,031	
1987	1,348 (0.16)	561 (0.07)	132 (0.02)	2,075 (0.25)	4,116 (0.50)	828,773	0.06
1988	1,606 (0.19)	608 (0.07)	170 (0.02)	2,266 (0.27)	4,650 (0.55)	842,206	0.05
1989	1,879 (0.22)	655 (0.08)	225 (0.03)	2,468 (0.29)	5,227 (0.61)	858,240	0.06
1990	2,247 (0.26)	715 (0.08)	285 (0.03)	2,693 (0.31)	5,940 (0.68)	878,769	0.07
1991	2,638 (0.30)	801 (0.09)	345 (0.04)	2,903 (0.33)	6,687 (0.75)	892,328	0.07
1992	3,084 (0.34)	847 (0.09)	425 (0.05)	3,099 (0.34)	7,455 (0.81)	915,654	0.06
1993	3,577 (0.38)	903 (0.10)	502 (0.05)	3,346 (0.36)	8,328 (0.89)	938,321	0.07
1994	4,162 (0.43)	972 (0.10)	583 (0.06)	3,595 (0.37)	9,312 (0.97)	963,490	0.06
1995	4,756 (0.48)	1,034 (0.11)	685 (0.07)	3,799 (0.39)	10,274 (1.04)	984,505	0.07
1996	5,415 (0.54)	1,086 (0.11)	756 (0.08)	3,971 (0.40)	11,228 (1.12)	1,004,230	0.08
1997	5,972 (0.59)	1,142 (0.11)	829 (0.08)	4,116 (0.41)	12,059 (1.19)	1,016,272	0.07
1998	6,572 (0.65)	1,188 (0.12)	902 (0.09)	4,254 (0.42)	12,916 (1.27)	1,016,791	0.08
1999	7,185 (0.71)	1,212 (0.12)	965 (0.10)	4,379 (0.43)	13,741 (1.36)	1,012,793	0.09
2000	7,813 (0.77)	1,255 (0.12)	1,054 (0.11)	4,502 (0.45)	14,624 (1.45)	1,008,481	0.09
2001	8,458 (0.84)	1,305 (0.13)	1,161 (0.12)	4,641 (0.46)	15,565 (1.55)	1,005,216	0.10
2002	9,083 (0.91)	1,354 (0.14)	1,241 (0.12)	4,747 (0.48)	16,425 (1.65)	994,836	0.10

continued on following page

Table 3. Prevalence of Children (aged 0–19) With a Developmental Disability in British Columbia 1986–2013

Year	Autism Spectrum Disorder (percent of total children in B.C.)	Down Syndrome (percent of total children in B.C.)	Fetal Alcohol Syndrome (percent of total children in B.C.)	Other Developmental Disability (percent of total children in B.C.)	Total Number of Children with a DD (percent of total children in B.C.)	Total Number of Children in B.C.	Absolute Percent Increase in Children With a DD (year over year)
2003	9,727 (0.99)	1,403 (0.14)	1,314 (0.13)	4,882 (0.50)	17,326 (1.76)	984,133	0.11
2004	10,437 (1.07)	1,444 (0.15)	1,394 (0.14)	4,982 (0.51)	18,257 (1.87)	976,030	0.11
2005	11,125 (1.15)	1,491 (0.15)	1,475 (0.15)	5,089 (0.52)	19,180 (1.97)	971,449	0.10
2006	11,770 (1.21)	1,546 (0.16)	1,552 (0.16)	5,159 (0.53)	20,027 (2.06)	970,121	0.09
2007	12,377 (1.28)	1,593 (0.16)	1,608 (0.17)	5,221 (0.54)	20,799 (2.15)	968,341	0.09
2008	12,994 (1.34)	1,654 (0.17)	1,631 (0.17)	5,260 (0.54)	21,539 (2.23)	967,538	0.08
2009	13,512 (1.40)	1,703 (0.18)	1,662 (0.17)	5,259 (0.54)	22,136 (2.29)	966,920	0.06
2010	13,899 (1.44)	1,736 (0.18)	1,672 (0.17)	5,262 (0.54)	22,569 (2.33)	966,860	0.04
2011	14,140 (1.46)	1,817 (0.19)	1,669 (0.17)	5,286 (0.55)	22,912 (2.37)	966,255	0.04
2012	14,328 (1.49)	1,892 (0.20)	1,696 (0.18)	5,305 (0.55)	23,221 (2.41)	963,780	0.04
2013	14,293 (1.49)	1,951 (0.20)	1,684 (0.18)	5,316 (0.55)	23,244 (2.42)	960,083	0.01

mental disability from 1986 to 2013 in the province of British Columbia. This information has not been available previously and will be important in future research and in program planning and funding decisions.

Using administrative data, this study found that in 2013 the prevalence of children with a developmental disability was estimated at 2.42% of the total population of children in British Columbia. The administrative health data also indicated that the prevalence of children with a developmental disability increased in British Columbia between 1986 and 2013 by

1.98%, largely due to an increase in the prevalence of children with ASD. However, there is also evidence that the increase in prevalence of children who have a developmental disability has slowed between 2010 and 2013.

For studies using administrative data, findings depend upon the data inputs which build the registry files (Broemeling et al., 2009). Therefore, limitations of the study findings include possible problems with the completeness and quality of the administrative data. In British Columbia, data from alternative funding arrangements (i.e., salaried physicians and nurse practitioners) and

Table 4. Comparison of Children With a Developmental Disability to Children Without a Developmental Disability in British Columbia

Variable	Autism Spectrum Disorder (birth data 1986–2013)	Down Syndrome (birth data 1986–2013)	Fetal Alcohol Syndrome (birth data 1986–2013)	Other Developmental Disabilities (birth data 1986–2013)	Comparison Cohort (children without a developmental disability born 1990–1995)
<b>Sex</b>					
Female	3,324 (22.02%)*	1,087 (45.86%)#	794 (43.22%)*	2,912 (45.32%)*	131,600 (48.91%)
Male	11,772 (77.98%)	1,281 (54.05%)	1,043 (56.78%)	3,510 (54.62%)	137,463 (51.09%)
<b>Income Level at Birth</b>					
Lowest	2,694 (24.22%)*	353 (22.70%)	520 (44.83%)*	944 (27.65%)*	57,527 (22.08%)
2 <sup>nd</sup>	2,391 (21.49%)	327 (21.03%)	243 (20.95%)	757 (22.17%)	55,449 (21.28%)
3 <sup>rd</sup>	2,187 (19.66%)	324 (20.84%)	144 (12.41%)*	683 (20.01%)	53,388 (20.49%)
4 <sup>th</sup>	2,124 (19.09%)	290 (18.65%)	134 (11.55%)*	439 (15.79%)*	50,919 (19.54%)
Highest	1,728 (15.53%)+	261 (16.78%)	119 (10.26%)*	491 (14.38%)\$	43,264 (16.61%)
<b>Health Authority at Birth</b>					
Number (% of total births of children with a DD in the province)					
Interior	1,982 (17.53%)*	273 (16.87%)	215 (16.86%)	522 (14.53%)	41,779 (15.53%)
Fraser	3,867 (34.20%)^	605 (37.39%)	172 (13.49%)*	1,215 (33.83%)**	96,120 (35.72%)
Vancouver Coastal	2,611 (23.09%)	360 (22.25%)	153 (12.00%)*	916 (25.50%)*	60,228 (22.38%)
Island	2,078 (18.38%)*	240 (14.83%)	312 (24.47%)*	583 (16.23%)	43,229 (16.07%)
Northern	769 (6.80%)*	140 (8.65%)	423 (33.18%)*	365 (9.91%)	27,718 (10.30%)

\* significantly different from the comparison group ( $p < .0001$ )# significantly different from the comparison group ( $p = .0036$ )+ significantly different from the comparison group ( $p = .0029$ )\$ Significantly different from the comparison group ( $p = .0059$ )^ Significantly different from the comparison group ( $p = .0009$ )\*\* Significantly different from the comparison group ( $p = .0137$ )

First Nations medical systems are not included in the administrative data set. Therefore children using only these services will not have been included in this study.

Data used in this study rely upon diagnoses provided by physicians. Conclusions about the accuracy of diagnostic coding vary widely in the literature from good (Henderson, Shephard, & Sundararajan, 2006; Jetté, Reid, Quan, Hill, & Wiebe, 2010) to poor (Farzandipour, Sheikhtaheri, & Sadoughi, 2010; Jensen, Cooke, & Davis, 2013; Peabody, Luck, Jain, Bertenthal, & Glassman, 2004; Stausberg, Lehmann, Kaczmarek, & Stein, 2008) depending upon location of the study, disease or treatment examined, use of ICD-9 or ICD-10 codes and the study design. Use of medical coding for administrative data is also complicated by the problem of co-morbidity (Jensen et al., 2013; Kirby, 2002; Quan, Parsons, & Ghali, 2002). Kirby (2002) reported that mental retardation, cerebral palsy, hearing and vision impairment and epilepsy often co-occur but also that conditions that are co-morbidities are less likely to appear in medical records. In addition, diagnoses are restricted to the labels provided by ICD-9 and ICD-10 codes. The codes for fetal alcohol syndrome may particularly restrict the range of diagnoses possible as the definition that accompanies the ICD-9 and ICD-10 codes is fetal alcohol syndrome, not fetal alcohol spectrum disorder. Therefore, given the nature of administrative data, it is likely that this study under-estimates the prevalence of developmental disability, particularly of FASD.

## Conclusion

In conclusion, this study found that 2.42% of children in B.C. in 2013 had a developmental disability. This population level information can help to address the lack of data regarding those who have a developmental disability and is important for future planning of programs and services for this vulnerable population. As Fujiura et al. (2010) stated: "What gets counted gets noticed."

## Key Messages From This Article

**People with disabilities.** It is important to know the number of children who have a developmental disability so that programs and services can be planned appropriately.

**Professionals.** The overall prevalence of developmental disability in B.C. increased between 1986 and 2013. The prevalence of each of the four categories of developmental disability used in this study increased, with the largest increases in the ASD category.

**Policymakers.** Policy and services should be based upon up-to-date information on the prevalence of developmental disability, estimates of this information are available through administrative data.

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## Promoting Testicular Self-Examination and Awareness Amongst Young Men With Intellectual Disabilities: A Parallel Intervention Randomized Study

### Abstract

*Testicular cancer is the most common type of cancer affecting males between 15 and 34 years of age. Men with intellectual disabilities have an increased risk of testicular cancer, often presenting late. We compared educative teaching (Teaching group) with an educative pictorial leaflet (Leaflet group) using a multi-media questionnaire to assess participants' knowledge, skills, and behaviour. Evaluation included demonstration of self-examination skills using an anatomically correct model. Adults with intellectual disabilities were involved as co-researchers at all stages from planning to writing-up the study. Both educational methods led to significant improvements in knowledge and skills, maintained 6 months after the intervention. At one-week, direct teaching had significantly greater effect on knowledge and skills than being given a leaflet. By six months improvements in knowledge, skills and behaviour were similar, without significant increase in health-related or general anxiety in either group. Young men with intellectual disabilities benefited from health education in group teaching sessions and through information provided pictorially in leaflets to read at home. This has wider implications for health promotion, where information is often withheld to avoid anxiety.*

Testicular cancer accounts for around 1% of all male cancers (Le Cornet et al., 2014) and is the most common type of cancer affecting adolescent and young males of European ancestry (Cook et al., 2010). Global testicular cancer incidence has increased since the 1970s, with highest rates in Western and Northern Europe, Australia and New Zealand, and Northern America, although there are variations between countries and between different ethnic and racial groups (Huyghe, Matsuda, & Thonneau, 2003). In the United Kingdom, testicular cancer incidence rates increased from 3.3 per 100,000 person-years in 1973–1977 to 5.4–5.8 per 100,000 person-years in 1998–2002, compared to 4.6 per 100,000 person-years in Canada in 1998–2002 (Chia et al., 2010). Testicular cancer incidence continues to increase worldwide (Znaor, Lortet-Tieulent, Laversanne, Jemal, & Bray, 2015), and is predicted to rise to 10 per 100,000 person-years in the United Kingdom by 2035 (Smittenaar, Petersen, Stewart, & Moitt, 2016).

There appears to be an association with intellectual disability (intellectual developmental disorder) [IDD for short] (Patja, Eero, & Iivanainen, 2001; Patja, Pukkala, Sund, Iivanainen,

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participatory research

& Kaski, 2006; Thornhill et al., 1988), genetic abnormalities (Dexeus, Logothetis, Chong, Sella, & Ogden, 1988; Sakashita, Koyanagi, Tsuji, Arikado, & Matsuno, 1980) and Down syndrome in particular (Hafeez, Singhera, & Huddart, 2015; Hill et al., 2003; Thornhill et al., 1988; Yang, Rasmussen, & Friedman, 2002). Barr, Gilgunn, Kane, and Moore (1999) reported that 10.8% of men with IDD have some abnormality of the testis. Eleven per cent of testicular cancers develop in men with a history of undescended testis (cryptorchidism) (Braga, Lorenzo, & Romao, 2017); indeed cryptorchidism is the most significant known risk factor for testicular cancer (Cook et al., 2010), and is more common among those with IDD (Cortada & Kousseff, 1984). Testicular cancer prevalence is higher in males with Down syndrome, even after accounting for a higher cryptorchidism rate, (Hasle, Clemmensen, & Mikkelsen, 2000; Miller, 1970). Additional postulated contributory factors include gene hyperexpression and higher levels of follicular stimulating hormone (Lambalk & Boomsma, 1998; Satge et al., 1997), and lifestyle factors including obesity, high fat diets, increased body size and low levels of physical exercise (Lerro, McGlynn, & Cook, 2010; Sasco et al., 2011).

The 5-year net-survival rate for testicular cancer is 96% (Canadian Cancer Society, 2017), with poorer prognosis for those presenting at a later stage of the disease (Cancer Research UK, n.d.). In addition to improving prognosis, early detection may reduce the physical and psychosocial distress associated with prolonged anti-cancer treatment (Shanmugalingam, Soultati, Chowdhury, Rudman, & Van Hemelrijck, 2013). Approximately 95% of testicular cancers present with a painless testicular lump or swelling (Rudberg, Nilsson, Wikblad, & Carlsson, 2005). However, of the twenty individuals with Down syndrome and testicular cancer reviewed by Dieckmann, Rube and Henke (1997), eight presented with metastatic disease, one had a giant tumour, and two had locally advanced disease, with testicular abnormalities being discovered incidentally by carers or healthcare professionals.

Although testicular cancers, particularly seminomas and teratomas, are highly sensitive to

radiotherapy and chemotherapy, men with IDD do not always receive comprehensive treatment for more advanced disease; for example, Miki, Ohtake, Hasumi and Moriyama (1999) report a case in which “neither chemotherapy nor radiotherapy were performed due to his severe mental retardation.” Evidence of inequitable access to healthcare is recognized in the U.K. 2013 *Confidential Inquiry into Premature Mortality in People with Intellectual Disabilities*, which recommended people with IDD should have access to the same investigations and treatments as others, whilst recognizing that such services may need to be delivered differently (Heslop et al., 2013).

Barriers to accessing healthcare, along with communication difficulties, pose challenges to the detection of early signs and symptoms of testicular cancer in individuals with IDD (Hanna, Taggart, & Cousins, 2011). There is debate about the role of routine testicular self-examination (TSE) in reducing delays in presentation among men in the general population and to a lesser extent among men with IDD. The U.S. Preventative Services Task Force discourages routine screening for testicular cancer in asymptomatic males due to lack of evidence of impact on mortality rates (Lin & Sharangpani, 2010), whereas the European Association of Urology recommends routine TSE for individuals with testicular cancer risk factors (Albers et al., 2010). Men in the general population have poor awareness of testicular cancer and TSE (Saab, Landers, & Hegarty, 2016b), and even opponents of testicular cancer screening by routine TSE accept the importance of educating young men about symptoms and signs to facilitate early presentation (Austoker, 1994; Buetow, 1996; Riley et al., 1998; Westlake & Frank, 1987). Riley et al. (1998) recommend health promotion should occur during childhood and adolescence, with an emphasis on positive health awareness rather than a ritualized “check for cancer.” Given the association of IDD and testicular cancer, and delays in diagnosis, investigations and treatment (Hanna et al., 2011), the case for promoting education about testicular cancer and TSE is particularly strong for men with IDD (Satge et al., 1997).

Overcoming the challenges in providing health promotion and education for people with IDD requires the use of accessible communication methods which are effective from the perspective of the individual with IDD (Bollard, 2002). These include visual representations alternative or supplementary to written or verbal content, and personalized support to optimize the participation of the individual with IDD (Boardman, Bernal, & Hollins, 2014). Hurtado et al. (2014) compared different formats in use with people with IDD and found stand-alone pictures were easier to understand than formats presenting information in more than one medium, such as words illustrated with pictures as used in easy read leaflets.

Research into accessible communication methods around testicular cancer and TSE for men with IDD is scant. One recent systematic review found 11 studies, utilizing a variety of strategies, to increase men's awareness of testicular cancer and their intentions to perform TSE (Saab, Landers, & Hegarty, 2016a; Saab, Landers, & Hegarty, 2016b). Although one included study reported the use of an American Sign Language video to meet the needs of deaf study participants (Folkins et al., 2005), none focused on men with IDD or the adaptations required to make health material accessible to this population.

There is a need to establish whether educating men with IDD can enable them to recognize early symptoms and signs of testicular disease and present to healthcare professionals for further investigation. One concern is whether such education increases participants' anxiety about cancer, leading to increased inappropriate use of healthcare resources. However the identification of benign abnormalities of the genitalia, which may be uncomfortable and distressing and affect sexual and reproductive health, should in itself be beneficial.

The objective of this study was to evaluate and compare, quantitatively and qualitatively, the impact of two different educational interventions about testicular health on knowledge, skills and health-related behaviours in regards to self-reported TSE at 1 week and 6 months post-intervention in male adults with IDD. One

intervention involved direct teaching, the other provision of a leaflet to take away and read. As both interventions utilized educational material specifically adapted for adults with IDD, it was hypothesized that both groups post-intervention would demonstrate increased knowledge of testicular cancer symptoms and signs, and increased skills in TSE (primary outcome). It was also hypothesized that the groups would be similar in secondary outcomes consequent to educational interventions about testicular health, including anxiety, health-related anxiety, health locus of control and self-efficacy. It was further hypothesized that both interventions would inform future approaches to involving young adult males with IDD meaningfully in their own health surveillance.

## Methods

### Research Design

This was a participatory randomized parallel study of two educational interventions. From the beginning research advisors with IDD were employed in assisting in the design of the study, in creating information, teaching materials and outcome measures accessible to individuals with IDD, in playing an active role in the research and through participating in steering and advisory group meetings. One research assistant with IDD was specifically recruited for the study, with responsibility for co-leading the teaching group with a male community IDD nurse (Butler, Cresswell, Giatras, & Tuffrey-Wijne, 2012).

The education methods and research evaluation tools were piloted for both teaching ( $n = 6$ ) and leaflet ( $n = 6$ ) interventions and alterations made as necessary. During the pilot study outcome measures were carried out immediately following each intervention; thus the Leaflet group did not have the opportunity to take the leaflet home and discuss the content with a family member, friend or helper prior to the initial post intervention evaluation. This protocol was altered for the main study so that first evaluation post intervention took place one week later for both Teaching and Leaflet groups.

## Participants

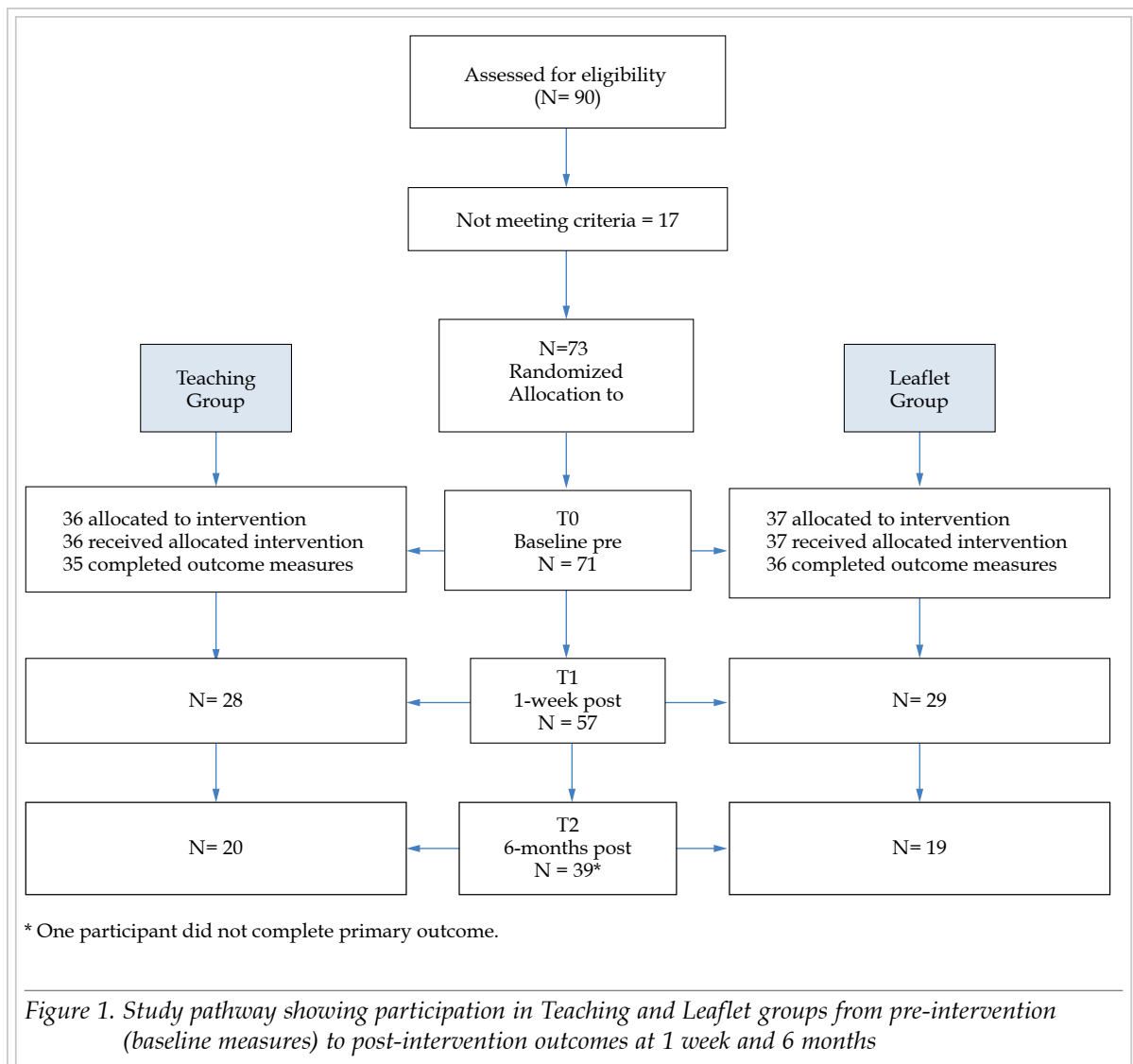
Ninety men with IDD aged 16 years or older were invited to take part in the research, having previously been selected as suitable by teachers and carers as likely to benefit from an educative intervention. Each participant received an information sheet about the study and informed consent was obtained. Seventy-three participants were assessed as meeting the study criteria (Figure 1). Researchers were careful to avoid coercing individuals to commence participation, or to continue as participants if they changed their minds during the study.

The inclusion criteria included: (1) being male aged 16 years or older; (2) registered with a GP,

and /or known to IDD services or on the social services IDD register or known to have Down syndrome; and (3) having the capacity to give informed consent. Ethics approval did not permit inclusion of males with moderate/severe and profound IDD.

## Procedure

Initially participants from several U.K. London boroughs were recruited through primary care services and IDD registers, but most successful recruitment occurred through colleges, day cen-



tres, voluntary organizations and youth clubs. As previous studies, such as those involving cervical screening for women with IDD, have described difficulties with recruitment for such research, the geographical areas chosen covered what was deemed to be a suitably large population to allow adequate numbers. Each participant was assessed as having capacity to consent to take part in the study by a specialist in IDD psychiatry.

## Randomization

Participants were randomly allocated into "Teaching" or "Leaflet" groups. This randomization was coordinated centrally by a statistician such that at each community or clinical site subjects were approximately evenly distributed between Teaching and Leaflet groups. Whilst this had the disadvantage of allowing some potential contamination between the groups, it had the advantage of ensuring that the two arms of the study were similar in terms of age and ethnicity of subjects. This was confirmed on subsequent statistical analysis.

## Interventions

**Teaching group.** An educational programme on testicular awareness was designed, using existing visual aids, video, a computer programme and an anatomically correct model with which to illustrate self-examination. The programme aimed to: (a) promote understanding of cancer risk and prognosis; (b) enhance the recognition of symptoms and signs, including those of more benign abnormalities; (c) instruct on how to seek help and self-refer to the GP; and (d) inform as to what further investigations and treatment might be expected if cancer was suspected. The aim was to design a programme which was understandable and accessible to people with IDD and which motivated the participants to alter their behaviour without provoking excessive levels of anxiety. The teaching methods were developed by a multi-disciplinary steering group incorporating representatives from consumer groups (young men with IDD), speech and language therapy, psychology, psychiatry, general practice, community intellectual disability nursing,

with advice from a urology specialist as needed. The teaching was delivered by a male community IDD nurse and a male co-researcher with IDD through two thirty-minute sessions. The teaching programmes were carried out in small groups no larger than six participants.

**Leaflet group.** Participants in the leaflet group received an educative leaflet on testicular awareness. The leaflet was designed by members of the steering group and an artist with expertise in publishing materials in accessible format, as part of a development project to create a picture book (on testicular examination) in the Books Beyond Words series (Hollins & Wilson, 2004a; Hollins & Wilson, 2004b). The leaflets were distributed and participants were encouraged to take them away and discuss them with family, friends or carers if they wished. This part of the study was not manualized, and carers/supporters received no training on how to use the leaflets with the men in the study.

## Outcome Measures

Members of the steering group were involved in adapting available evaluative tools and designing new tools where necessary.

**1: Primary measures.** The primary measure was a 10-item questionnaire available in verbal, pictorial and written format (so as to accommodate the communicative preference of the participant) to assess *knowledge and skills* (S&K) and was administered face to face by one of two medically qualified researchers. This S&K questionnaire included demonstration of testicular self-examination skills using an anatomically correct model.

**2: Secondary measures.** Twelve items were selected from existing inventories of measures of general anxiety, health anxiety, health locus of control and self-efficacy and adapted to the communication needs of participants (to create a 12-item adapted questionnaire – see below). Each item was represented by a statement and a 0 to 10-cm line. The latter was anchored at its ends by descriptive phrases, one end of the line indicating strong disagreement with the statement, and other end indicating strong agree-

ment. The line was then measured in millimetres providing a measure (analogue scale) of the individual's agreement/disagreement with the statement; higher scores indicated greater anxiety, greater self-efficacy and the extent of control (internal, external or chance).

*Outline of the 12-item adapted questionnaire (12-item Adpt-Q).* Four items measuring *General Anxiety* in the State-Trait Anxiety Inventory (Spielberger, 1989) were adapted and made accessible by using visual analogue scales (e.g., "I am tense" with strongly agree = 10 and strongly disagree = 0). Three items from the Wells Health Anxiety Scale (Wells, 1997), were used to assess *Health Anxiety*, with a higher score on the analogue scale indicating that someone feels in control of their health/not worried about their health. *Locus of control and health care related behaviours* were measured using three items from the Health Locus of Control scale (Wallston, Wallston, Kaplan, & Maides, 1976); this scale describes health related behaviours worded from internal ("if I take care of myself I can avoid illness"), external ("doctors and nurses control my health") and chance ("staying healthy is a matter of luck") perspectives. *Self-Efficacy* with regard to testicular self-examination and seeking help was measured by two questions ("I know how to check my balls/testicles" and "I feel able to tell someone if I find something wrong with my balls/testicles").

Also recorded were:

### 3: Participant reported self-examination and

**4: Self-referral activities and GP visits.** Self-recording diaries (adapted to the communication needs of the participant) were distributed to individuals in each group so that they could monitor their behaviour in terms of self-examination and GP visits over a six-month period.

**5: Qualitative evaluation.** This was through feedback from participants to the medical researchers using a 5-question semi structured interview. The self-advocate co-researcher participated in this feedback providing opportunity to explore and compare participant personal experiences to each intervention (Tuffrey-Wijne,

Bernal, & Hollins, 2008; Tuffrey-Wijne, Bernal, Butler, Hollins, & Curfs, 2007).

Both the 10-item S&K-Q and 12-item Adpt-Q were completed by the participant with guidance from the medical researchers who read each question out loud, repeating and clarifying the meaning as necessary. The same researchers were used throughout and were blind to the allocation to Teaching and Leaflet group. Each participant completed both questionnaires on three occasions: at baseline (T0); one-week (T1) and six-months (T2) following the interventions (Figure 1).

## Analyses of Data

Analyses were undertaken using IBM SPSS v24. Data were analyzed on an 'intention to teach' basis. Differences between the groups with respect to the primary outcome measure (10-item S&K-Q) was analyzed using analysis of covariance including the T0 level of the dependent variable to adjust for regression to the norm. This was done twice, for the one-week (T1) and six-months (T2) follow-ups. Secondary outcome measures, which were normally distributed, were analyzed in the same way. The Mann Whitney U test was used to analyze the data on frequency of self-examination and GP self-referral rates. Categorical variables were analyzed using chi squared analysis. Paired t-tests were used to make within treatment group comparisons to explore change from baseline in all outcomes.

## Results

The age range of participants was from 16 to 35 years (mean age = 20.9, *SD* = 4.8). Thirty-six were randomized to the teaching group and 37 to the Leaflet group (Figure 1). The two groups were similar for age (mean teaching = 21.3, *SD* = 5.5; leaflet = 20.4, *SD* = 4.0). At 1-week follow-up (T1), there were 28 participants in the Teaching group and 29 participants in the Leaflet group; at 6-month follow-up (T2) there were 20 participants in the Teaching group and 19 participants in the Leaflet group. Participants who dropped out showed no significant differences in age or scores on baseline measures.

## Primary Outcomes: Knowledge and Skills

There was a significant difference between the groups 1 week following the intervention (Teaching group scoring significantly higher ( $p = .002$ ) but this difference between the groups was no longer significant at the 5% level at 6 months ( $p = .055$ ; Table 1). Statistically significant change from baseline in knowledge and skills scores at 1 week and 6 months were seen in both treatment groups ( $p \leq .001$ ) for all hypotheses tested.

## Secondary Outcomes

**Health anxiety.** There was a significant difference between the groups at T1 with respect to health anxiety ( $p = .043$ ) with the Teaching group exhibiting less anxiety, but not at T2 ( $p = .283$ ). There was a significant reduction in health anxiety for the Teaching group at T1 which was maintained at T2 ( $p = .044$  and  $p = .012$  respectively). Changes in the Leaflet group were similar although not statistically significant.

Table 1. Primary and Secondary Outcomes at Baseline (Pre-Intervention), 1 Week and 6 Months Post-Intervention in Teaching and Leaflet Groups

T0 = baseline (pre-intervention); T1 = 1 week post-intervention; T2 = 6 months post-intervention

	Teaching Group		Leaflet Group		Significance
Outcome measure (0-max possible)	Mean (SD)	<i>n</i>	Mean (SD)	<i>n</i>	<i>F</i> ( <i>p</i> -value)
Knowledge and Skills (0-20)					
T0	5.8 (2.7)	35	5.7 (2.8)	36	
T1	11.9 (4.1)	28	8.9 (4.5)	29	10.2 (.002)
T2	12.6 (2.8)	19	9.6 (4.4)	19	4.0 (.055)
T1-T0 <sup>1</sup>	3.8 (2.8, 4.8) < .001	26	1.7 (0.7, 2.6) .001	29	
T2-T0 <sup>1</sup>	4.4 (3.2, 5.7) < .001	19	3.3 (1.7, 4.8) < .001	19	
Health Anxiety (0-30)					
T0	9.8 (6.6)	35	11.9 (6.1)	36	
T1	8.2 (4.2)	26	11.4 (6.4)	29	4.3 (.043)
T2	8.0 (5.3)	19	10.1 (4.2)	19	1.2 (.283)
T1-T0 <sup>1</sup>	-2.5 (-4.9,-0.1) .044	26	-0.7 (-2.5, 1.0) .382	29	
T2-T0 <sup>1</sup>	-3.4 (-5.9, -0.9) .012	19	-2.8 (-5.7, 0.1) .057	19	
General Anxiety (0-40)					
T0	18.7 (6.7)	35	20.1 (7.0)	36	
T1	17.9 (6.3)	27	20.0 (5.4)	29	0.6 (.436)
T2	21.0 (2.3)	20	21.5 (3.2)	19	0.5 (.500)
T1-T0 <sup>1</sup>	0.4 (-1.1, 1.8) .606	27	0.3 (-2.3, 2.8) .825	29	
T2-T0 <sup>1</sup>	2.1 (-1.4, 5.7) .224	20	0.3 (-2.8, 3.4) .839	19	

continued on following page.

Table 1. Primary and Secondary Outcomes at Baseline (Pre-Intervention), 1 Week and 6 Months Post-Intervention in Teaching and Leaflet Groups (continued)

All Anxiety (0–70)					
T0	28.6 (10.8)	35	32.1 (11.6)	36	
T1	25.8 (8.2)	27	31.4 (9.9)	29	3.6 (.062)
T2	28.8 (5.7)	20	31.6 (5.6)	19	1.1 (.311)
T1–T0 <sup>1</sup>	-2.0 (-5.0, 0.9) .112	27	-0.5 (-3.9, 3.0) .787	29	
T2–T0 <sup>1</sup>	-1.1 (-4.9, 2.7) .550	20	-2.5 (-6.8, 1.9) .248	19	
LOC Internal (0–10)					
T0	7.1 (2.4)	35	7.4 (2.3)	36	
T1	8.0 (1.6)	26	7.7 (2.1)	29	0.2 (.883)
T2	7.4 (2.3)	19	8.2 (1.0)	19	1.5 (.235)
T1–T0 <sup>1</sup>	0.2 (-0.4, 0.9) .465	26	0.4 (-0.3, 1.1) .270	29	
T2–T0 <sup>1</sup>	0.7 (-0.9, 2.4) .376	19	1.5 (0.1, 3.0) .042	19	
LOC External (0–10)					
T0	5.7 (3.0)	35	5.4 (2.5)	36	
T1	6.4 (2.8)	26	5.7 (2.4)	29	0.5 (.502)
T2	4.3 (2.8)	19	6.4 (1.6)	19	7.1 (.012)
T1–T0 <sup>1</sup>	0.1 (-0.9, 1.0) .847	26	0.0 (-1.0, 0.9) .935	29	
T2–T0 <sup>1</sup>	-1.5 (-3.1, 0.1) .061	19	0.0 (-1.1, 1.2) .940	19	
LOC Chance (0–10)					
T0	5.6 (2.7)	35	5.4 (2.6)	35	
T1	5.7 (2.6)	26	5.2 (2.7)	28	0.1 (.730)
T2	5.1 (2.8)	19	6.0 (2.4)	18	1.1 (.298)
T1–T0 <sup>1</sup>	-0.3 (-1.1, 0.6) .546	26	0.2 (-1.5, 1.1) .774	28	
T2–T0 <sup>1</sup>	-0.8 (-2.2, 0.7) .272	19	0.1 (-1.5, 1.7) .906	18	
Self-efficacy (0–20)					
T0	12.2 (3.9)	35	13.2 (4.6)	36	
T1	14.5 (4.1)	27	14.3 (3.6)	29	0.1 (.734)
T2	14.6 (4.7)	20	14.1 (3.5)	19	0.1 (.797)
T1–T0 <sup>1</sup>	2.3 (0.6, 4.0) .011	27	1.6 (-0.3, 3.4) .101	29	
T2–T0 <sup>1</sup>	2.3 (-0.2, 4.8) .066	20	2.3 (0.5, 4.0) .014	19	
	Median (min, max)	<i>n</i>	Median (min, max)	<i>n</i>	MWU ( <i>p</i> -value)
GP visits (over 6 months)					
T2	0.0 (0, 20)	33	0.0 (0, 20)	33	455.0 (.191)
Frequency of Self-Reported Examination					
T2	1.0 (0, 100)	22	6.0 (0, 100)	19	182.5 (.478)

<sup>1</sup> Values are mean difference (95% CI around mean difference) *p*-value resulting from paired *t*-test, testing for change in outcome between respective time points.

**General anxiety.** There was no significant change in generalized anxiety within either group at both stages. There was also no significant difference between groups at T1 and T2. Qualitative feedback identified several participants who confirmed the importance of discussing the subject matter with a parent, career or other supporter. This helped to clarify the information given and reduce subjective anxiety.

**Locus of control.** Between T0 and T2 there was a significant increase in "internal" locus of control for the Leaflet group ( $p = .042$ ) suggesting that pictorial information empowered people to feel that they had more control over their own health. There was no significant change in the Teaching group. While there was a significant difference between the groups at T2 ( $p = .012$ ) (the Leaflet group scoring higher), changes in locus of control relating to external factors and chance were not statistically significant in either group.

**Self-efficacy.** There was a statistically significant increase in mean self-efficacy scores from T0 in the Teaching group to T1 ( $p = .011$ ) and in the Leaflet group to T2 ( $p = .014$ ). Subjects felt more confident about their ability to check their testicles and to seek help if they found something wrong. There was no significant difference between the groups.

## GP Visits

There was no significant difference between groups in terms of GP visits over the 6-month period ( $p = .191$ ). Qualitative feedback indicated at least two individuals in the Leaflet group successfully sought reassurance and further information from their GP.

## Frequency of Testicular Examination

There was no significant difference overall between the groups in terms of frequency of testicular self-examination ( $p = .478$ ; Table 1); seventy-three percent of the Teaching group and 63% of the Leaflet group reported performing testicular self-examination at 6 months ( $\chi^2 = 0.43, p = .511$ ).

## Qualitative Outcome

**Subjective experience of the interventions.** Thirty-six per cent of participants gave suggestions on how education on the topic could be improved. Some suggested that photographs could be used in the leaflets. One participant said that the video used in the Teaching group was particularly helpful. Several participants felt they would benefit from more teaching sessions and that more detail should be included. Two participants wanted to know more about what would happen if you did get testicular cancer (i.e., prognosis and treatment). Some were keen to learn about other ways of keeping healthy. Several participants suggested that the subject should be taught to younger people and be included in sex education at school. Two said a doctor should be involved in the teaching. Others said that discussing the subject with a parent or carer after receiving either the leaflet or teaching was the best way to learn more without getting too worried. Most participants in both groups commented positively ("good") on the intervention they had participated in and could not think of any suggestions for improvement.

Subjective feedback on both interventions did indicate that many found the content at least "a bit" worrying (28% in the Teaching group and 33% in the Leaflet group). However neither intervention led to a statistically significant increase in measured health-related or general anxiety and indeed the trend was for anxiety levels to decrease between T1 to T3.

Significantly improved confidence in ability to check and seek help was gained initially by Teaching group participants and after six months by those in the Leaflet group, indicating that both interventions had an effect on behaviour as well as on knowledge and skills.

## Discussion

This randomized group intervention study involving male adults with IDD, but without prior clinical evidence of testicular cancer, demonstrates that both a formal teaching programme and provision only of a pictorial leaflet in accessible format are effective in improving knowledge and skills and this improvement was maintained 6 months after each intervention. Initially the formal teaching had a significantly greater impact on knowledge and skills

than leaflet provision only but by six months these were similar in each group (Table 1).

Qualitative data suggests that individuals in the Leaflet group tended to seek further information on the subject from family, friends or helpers during the six months after receiving the leaflet to a greater extent than those who received the teaching; it is possible that the leaflet assisted carers/supporters discuss the subject more easily (Jones, Tuffrey-Wijne, Bernal, Butler, & Hollins 2007). In both groups the majority of participants reported performing testicular self-examination 6 months post intervention.

As no testicular pathology was reported and none found at clinic follow-up (not unexpected given the short length of the study, the incidence of testicular pathology and small number of participants), there was not opportunity to evaluate correct identification of pathology by participants. However, the assessment of testicular awareness included the use of hands-on anatomical models which simulated pathology in terms of palpable testicular lumps, and the ability of participants to identify these and explain what they would do if they found such an abnormality was included in the assessment outcomes.

The study findings confirm that young men with IDD can benefit from health education to improve their levels of knowledge, and skills and change behaviour. The subject of testicular self-examination and cancer is potentially embarrassing and anxiety provoking but the participants in both educative approaches, responded well to the content and appeared to benefit without marked detrimental effects. The evidence suggests that when participants receive information in an accessible form they can feel more confident about having influence over their health and seeking help, leading to reduced anxiety levels. This has implications for other areas of health promotion and information provision for people with IDD where information may be withheld with the intent of protecting an individual from anxiety.

## Conclusion

Both pictorial leaflets and teaching sessions have a place in providing information to men with IDD about testicular awareness. If a leaflet or book is used the researchers recommend

that the man is encouraged to go through the information with a supporter. The use of pictorial leaflets is more efficient in terms of health service resources; additionally, there is no evidence from this study that teaching men with IDD increases demand on health care resources, other than that related to the teaching itself.

The combination of teaching sessions and an accessible information leaflet might be expected to combine the benefits of both approaches although such a conclusion cannot be drawn without further research. There is no evidence that providing accessible information for the carers/supporters to introduce to the person they support, would be an effective alternative. Indeed a previous randomized control trial (RCT) comparing the effectiveness of a bereavement intervention found that carers/supporters were reluctant to use the pictorial material provided on that sensitive subject, whereas bereavement counsellors used the materials effectively (Dowling, Hubert, White, & Hollins, 2006).

## Implications for Further Research and for Practice Development

The results of this study suggest that young men with mild IDD can benefit from health education even when the subject is potentially embarrassing and anxiety provoking and that the benefits can outweigh any harmful effects and not lead to inappropriate excessive use of primary care services. This has implications for other areas of health education, including sexual health and cancer information and screening; although similar studies in specific areas e.g. breast awareness for women as well as wider "men's health" education would be beneficial.

It was the intention of the researchers to carry out the study in a way that was as ethically acceptable as possible. To this end we ensured that information on the research was provided in an accessible format and that the process was explained fully using understandable language so that participants were as informed as optimally as possible before considering whether they wished to take part. This effectively maximized the capacity of individuals to give or withhold their consent. Researchers endeavoured to ensure that there was no coer-

cion of the subjects taking part and that their consent and willingness to participate was maintained throughout their participation. The design of the study was such that all subjects had the opportunity to benefit from participation whether in the teaching or leaflet group. Through such measures the study demonstrates that people with IDD, albeit mild in severity in this case, can be ethically included in such research as subjects as well as researchers (Tuffrey-Wijne et al., 2008).

However, it is likely that both the teaching and the leaflet can be used effectively with people with more severe intellectual disabilities, with the involvement of supporters as recommended above. The latter has the added benefit that supporters/carers also learn about the subject together. The wider Books Beyond Words series are designed to be used in this way. While assessing the benefits for people with more severe disabilities was beyond the scope of this study, the pictorial tools involved do help to increase each individual's capacity to understand the issues addressed. The authors believe that the findings of the study are likely to apply to those with more severe disabilities and there is no known reason to exclude them from similar educational interventions.

During the study, the participants scored higher on anxiety measures prior to any intervention. One explanation may be that raising the subject of testicular cancer caused anxiety, which was reduced as more information and education was provided. Alternatively, involvement in the research process itself and being "tested" may have provoked anxiety which reduced as the subjects became more accustomed to the process. Clearly when involving people with intellectual disabilities in research, measures should be taken to explain the process carefully and clearly and to provide full information to minimize anxiety.

The seven-page leaflet used in this project comprised coloured drawn pictures, usually one picture on a page supplemented with a simple phrase or sentence as a descriptor of the picture. The pictures were of the type found in the "Books Beyond Words" series ([www.books-beyondwords.co.uk](http://www.books-beyondwords.co.uk)) and were subsequently included in the title in the series "Looking After My Balls." The results indicated that leaflets given directly to a person with IDD by a health

professional, and taken home to read and share with a carer or friend, produced comparable results to face to face group teaching by an IDD nurse and peer. Whilst wordless leaflets are becoming an established method for providing people with IDD with information on difficult and emotive subjects (Hollins, Carpenter, Bradley, & Egerton, 2017), this has not been adopted as the optimal format for health promotional literature for people with IDD. Further research in this area could seek to compare the success of different formats, such as symbols, wordless books, easy read formats, audio and video presentation, for providing such information for people with IDD. Research could also follow participants for longer to determine whether improvements in knowledge and skills from similar health educative interventions can be sustained and whether peer support groups would assist with this objective.

While the focus of this study was on educating adult males with IDD about their testicular health, the study draws attention to the need also to educate health care providers (such as pediatricians, nurses, family doctors) about the increased prevalence of testicular cancer and cryptorchidism in this population. The latter, the most significant known risk factor for testicular cancer, is a condition that should be diagnosed and treated in childhood. This not only allows for easier detection of testicular cancer but also lowers the risk of developing it (Pettersson et al., 2007).

## Key Messages From This Article

**People with disabilities.** Hooray! "Nothing about us without us."

**Professionals.** Adults with intellectual disabilities are able, and want, to contribute to research about their health.

**Policymakers.** When adults with intellectual disabilities receive information in pictures, they feel more confident about having influence over their health and in seeking help.

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## Tell Us More – People With Developmental Disability Share Experiences to Enhance Health Care Education: An Exploratory Study

### Abstract

*The prime objective of this qualitative study was to understand the health care experiences of people with developmental disability (DD) in order to inform initial and continuous training of health care professionals. Previous studies of health care experiences have not exclusively involved individuals with DD. Similar investigations involved caregivers' perceptions and have focused on specific health care settings. This study highlights the benefits of including people with developmental disability in research and as health care advisors. The study involved 22 individuals, who participated in one of seven focus groups held between February and March, 2014. Focus group participants represented a diversity of ages, as well as physical and mental health conditions. Focus group participants provided examples of both good and not so good health care experiences. They described challenges, impacts on their health, and thoughts for improving interactions. Eight dominant themes were identified: Person-Centred Health Care, Barriers to Access, Attitudes, Communication, Professionalism, Outcomes, Continuity of Care, and Inter-Professional Care. Findings have been incorporated into health care education curriculum and ongoing training activities of health professionals.*

Studies in developed countries have shown that people with developmental disabilities (DD) experience poorer health, increased morbidity, earlier mortality and yet are poorly supported by health care systems (Sullivan et al., 2011). An unpublished 2004 American Academy of Developmental Medicine and Dentistry report on surveys of deans and students of American medical schools identified the need for more medical education related to people with DD (Holder, 2004). Of the deans who responded (N = 40), 53% did not feel that their graduates were competent to treat patients with neurodevelopmental disorders or intellectual disabilities. Similarly, of the graduating students who responded (N = 427), 56% felt that they were not competent to treat these patients. A 2009 publication by Holder, Waldman and Hood extended this preliminary report (Holder, Waldman, & Hood, 2009). The latter surveyed deans of American medical and dental schools and found that only 61% of deans of medical schools and 47% of deans of dental schools felt that their graduates were competent to treat patients with disabilities. Interestingly enough, they had a 55% response rate from deans of dental schools but only a 22% response rate

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from medical schools. Further, they found that deans were not responding because their programs were not offering any training regarding this population. There is a clear need to address not only systemic barriers to effective health care, but also the unsatisfactory experiences of people with DD as they interface with health professionals. It is well supported that compassion, empathy and person-centred approaches by health care providers are associated with improved clinical outcomes for patients (Lown, Rosen, & Martilla, 2011; Neumann et al., 2011). Person-centred care emphasizes understanding and respecting the patients' preferences, values, comfort, communication abilities and providing health care based on this understanding (Delaney, 2018). This is seen to be especially important for people whose cognitive ability and life experiences may have been quite different than peers who do not have DD. It can be assumed that significantly higher rates of abuse, exploitation and neglect for this population (Sullivan et al., 2011) would impact on this populations comfort level during health care interactions. Cognitive ability and high rates of co-occurring physical limitations (vision, hearing etc...) suggest the need for a more customized communication strategy to provide health care supports and soliciting consent to treatment (Chew et al., 2009).

Multiple studies have found that compassion, empathy and person-centred approaches to care appear to decline as medical students move through their training (Lim et al., 2013; Neumann et al., 2011; Rosenthal et al., 2011). It is recommended that DD curricula for health care learners emphasize respectful, person-centred attitudes in addition to relevant medical knowledge and skills (Ouellette-Kuntz et al., 2012).

McMaster University's Michael G DeGroote School of Medicine Niagara Regional Campus (NRC), Bethesda Services, Southern Networks Of Specialized Care (SNSC), Brock University Centre for Applied Disability Studies and Nursing, have partnered in a collaborative initiative whereby medical and nursing students experience early interactive encounters with people who have DD. A Curriculum of Caring for people with DD has been developed with experiential learning opportunities for students to learn from people with DD and their caregivers. Components of this curricu-

lum have been used for continuing medical education training online and in person across the province of Ontario by Health Care Facilitators through the Community Networks of Specialized Care.

Focus group research on patients has become more prevalent in health care research. They have been especially helpful in exploring needs assessments, curricula development, program improvement, organization development and outcome evaluation (Leung & Savithri, 2009). Focus group research is viewed as especially advantageous to solicit information from disempowered patient groups who may feel that they have little of value to share or that any problems they experiences are a result of their own inadequacies (Kitzinger, 1995). Interestingly enough, care provider focus groups are significantly more prevalent in the research than focus groups of people with DD. A review of the literature identified a focus group study of experiences of people with DD and their care providers in U.K. hospitals (Gibbs, Brown, & Muir, 2008). While this study involved 11 individuals with DD, it also involved soliciting opinions of 14 care providers during the focus groups. This study focused on experiences within hospital settings. These tend to involve interactions with more specialized health care professionals who are involved for a shorter duration than community based providers. They tend to be more stressful for patients and focused on more acute health care issues. This study highlighted feelings of anxiety and fear, communication and behaviour problems; the practicalities of being in or attending hospitals, the role played by care providers; issues around negative comments and feelings of being treated differently. While the findings are helpful in supporting health care curricula, it was felt that additional information on lived experiences with a wider range of health care professionals (including those in longer term relationships) and different health care settings was required to enhance initial and ongoing health care education. Increasingly, medical schools have recognized the need for and benefits of involving patients with disability as teachers (Eddey, Robey, & McConnell, 1998; Minihan et al., 2004). Tracy and Iacono (2009) found a significant positive change in attitude and perception of people with DD by health care students when directly involved with them as educators.

The present focus group study advances the concept of involving people with DD as mentors and educators by soliciting perceptions of their health care experiences as well as advice to students and professionals. The focus group input enhances the Curriculum of Caring curricula for health care students and informs developmental disability health care education provided by the Southern Network of Specialized Care. The goal of this study was to inform curricula that would increase the capacity of professionals to provide competent and compassionate health care for people with DD. This article reports on findings specific to health care experiences and interactions with professionals. A separate article describing experiences with, and advice to, health care students has been published in a medical education journal (Moore, Lidster, Boyd, Kates, & Stobbe, 2015).

## Methods

As described below, information to address the objective was collected from participants using a focus group format.

### Ethics

This study was approved by the Hamilton Integrated Research Ethics Board and ensured that participants were informed, involved, supported and comfortable during both the recruitment phase and focus group discussions.

### Study Design and Analysis

The study was qualitative in nature; it focused on responses of people with DD during semi-structured, facilitated small group interviews and written answers to questions (Appendix 1) that were distributed prior to the focus group sessions.

A pilot focus group of seven participants was conducted to test and enhance study design. It was determined that smaller group sizes were preferable for discussion purposes. Responses from the pilot focus group were not included in study findings. All focus group participants were informed that they could bring someone with them to provide personal, emotional and communication support. At the beginning of each focus group, supports were reminded of

the limitations of their role. Focus groups were facilitated by two health care students and a Health Care Facilitator from the Southern Network of Specialized Care. All focus group facilitators received training from the Health Care Facilitator and the Principal Investigator on soliciting information from the participants in a way that promoted unbiased, broad-based discussion in a safe environment.

An ice breaker question (unrelated to health care) was introduced to stimulate and encourage discussion from all participants. Study participants were then asked to report on both "good" and "bad" health care experiences. They were prompted to consider interactions with all health professionals (medical doctors, nurses, dentists, paramedics, other therapists) in their responses. Focus group facilitators probed for additional context (e.g., "What made that a good/bad experience for you?"). Focus group discussions were audio recorded and then transcribed by a research team member. Audio transcripts and written responses were independently analyzed by two investigators for emerging themes and collated. Investigators translated information from the study into achievable recommendations for developmental disability medical education curriculum enhancement.

### Recruitment of Participants

Study Investigators contacted all 29 government funded developmental service providers in the Hamilton Niagara area to solicit their assistance in recruiting study participants from people they support. These providers distributed a plain language recruiting package (cover letter, description of research, consents, focus group questions) to individuals involved in their residential, day, and employment programs. Interested participants submitted completed packages to a developmental service provider in their local geographic area (Haldimand, Norfolk, Niagara, Brant, and Hamilton). Focus group participants were consecutively selected and assigned to focus groups of no more than four participants. Participants were contacted by a study investigator to provide additional information about the study, confirm consent, and encourage participants to review the focus group questions in preparation for the meeting. The investigator suggested that par-

ticipants write out their responses to reference during the discussion and to submit separately if they felt uncomfortable sharing with others. Participants were informed that they could invite supports to help them prepare for and communicate during the discussion.

## Study Sample

The study involved seven separate focus groups of three to four participants ( $N = 22$ ; female 17, male 5) during February and March, 2014. Study participants ranged in age from 20 to 70 years, with an average age of 51 years. Participants from both urban (13 from centres over 100,000 population) and rural centres (nine from centres under 100,000 population) participated in the study. Study participants represented a wide variety of both physical and mental health diagnoses. Five study participants (23%) used mobility aids. All study participants communicated verbally in discussions. Six (27%) of the study participants were accompanied by support persons (one parent, one spouse, four direct support professionals) to provide emotional or communicative support.

## Results

Study investigators identified salient themes from the analysis of participant responses. Themes were grouped under headings of (1) general health care experiences, (2) experiences with health care students, and (3) advice to health care students. This article focuses on participant's general health care experiences. Eight dominant themes were identified: Person-Centred Health Care, Barriers to Access, Attitudes, Communication, Professionalism, Outcomes, Continuity of Care, and Inter-Professional Care. These themes were universal among identified demographic characteristics (gender, age, rural/urban, physical or mental health co-morbidity).

### Person-Centred Health Care

Participants clearly expressed the importance of being consulted about their health care needs and preferences. Participants described positive experiences where health professionals spent time to understand their unique health care issues and anxieties (e.g., around proced-

ures) then jointly determined how to adapt/modify assessment and treatment.

One participant was frustrated during a hospitalization because health care providers would not give him the support he needed to manage ongoing health issues because they were not the reason he was hospitalized:

When I was in the hospital last ... they did not roll me. I'm a quadriplegic. I cannot roll myself. I get bedsores if I am not rolled. They didn't do my bowel routine either... They just didn't give a ----. Even with rolling me, I was on the one side 6-7 hours without being rolled. They just didn't care. One nurse even said 'we don't do that here.' I was in the hospital for one reason and they did not care about the other health care support I needed in addition to that one reason. My daily care was ignored. It was left behind. This happens every time. Normal people should not have to go through this.

This participant also thought the hospital staff should have considered his inability to use the call system and placed him in a hospital room close enough for him to get attention when needed:

If I am put in the very back of the hospital floor, far from the nursing station they can't hear me and they can't see me. I have no way of alerting them. I cannot ring the bell or push down the buzzer they leave for me ... If I have a dizzy spell I have no idea where I am, let alone know how to get the buzzer or bell working.

Others described unsatisfactory experiences where modifications could have made a great deal of difference. One participant indicated that, if consulted, she would have informed hospital staff that she was left handed, and an IV on the right arm would have allowed her greater independence during her hospital stay.

### Barriers to Access

Participants identified a number of significant barriers: reliance on caregivers, convincing others of illness, transportation, cost, clinic hours, emergency department busyness and wait times. Of particular interest for developmental service providers, participants identified barriers that occur prior to encounters with

health care workers. One participant described a necessary medical appointment being delayed by the need to convince caregivers that her concerns were valid followed by subsequent difficulties arranging staff coverage and transportation. Several participants described the need to have someone without a disability accompany them to appointments before health care providers believed their reports of illness.

## Attitudes

Participants valued friendliness, respect and honesty from health care providers. Participants commented on the importance these traits play in their level of trust with health professionals and the care they provide. One participant described her frustration with a physician not believing her:

He tried to tell me I didn't have a seizure and I did. He kept saying he knew a lot, but he didn't ... It is important that the doctor listens to your opinion on what is going on in your body ... The person that had the seizure knows, you (doctor) don't ... Don't tell me that I don't know my own body when I do!

Positively reinforcing the person's efforts towards good health/dental maintenance was also appreciated by participants.

## Communication

Listening, non-verbal communication (body language), manner of speech, and choice of words (plain language) are important:

Instead of using a 30-letter word, use a five-letter word...It is easy to explain things to her. She tries to explain things in a way I will understand. If I don't understand I can say that I don't understand and she will find another way to explain it to me.

All participants commented on the importance of effective communication. The willingness of health care providers to take the time and adapt their communication methods was identified by many as having a direct impact on health care outcomes. Participants identified less than ideal experiences where health care providers did not look at them during appointments and checked notes on the computer during interactions. "One

time the doctor was talking on his headset to the computer saying 'erase that, erase that.'" Several participants described positive interactions where health professionals asked if they understood and then took the time to modify their communication (plain language) so that they could be more involved in health care decisions.

## Professionalism

Respectful communication, explaining/obtaining permission, punctuality, soliciting informed choice/consent, and maintaining privacy were central to successful health care experiences. Participants consistently associated these elements of professionalism with positive health care experiences. Conversely, they frequently mentioned the absence of these traits in their negative health care experiences. Many participants described their frustration with health care professionals who spoke to their supports (like they were not present), prompting one participant to wave her arm and say "Hello, I'm right here!" Participants frequently described less than ideal experiences in hospitals where other patients, visitors and professionals not directly involved in their care heard their confidential and personal information. Some reported experiences where assessment and treatments were performed without their consultation or consent. Participants also described positive scenarios where health care providers explained procedures, sought input and obtained consent. "Our doctor is awesome! Whenever we see her she will actually sit and talk to us about it... she will say that this way is the better way to go and why. She will talk about side-effects and everything."

One participant described her frustrations with health care appointments that are not kept and how it affected her health:

[The person] was going to come and take my sugar for my diabetes. Well I waited and waited. I couldn't have my breakfast or medication. Then it came to 10:30 am and she didn't show up ... Then came Friday and she still didn't show up ... I was so, so mad at her for not coming those two days ... It is the waiting and no one telling me when they are coming or why they are not coming that gets me angry. They can tell me when they are coming by phone.

## Outcomes

Clear diagnoses, treatment success, relief from discomfort, pain management, and improved health were identified as important outcomes of health care interactions. Interestingly, several participants described positive experiences when treatments alleviated pain and discomfort; however, the attitude and behaviour of the health care professional lacked care and compassion.

## Continuity of Care

Consistency and communication among health care practitioners are valued. "We are very happy with our doctor. We are never leaving her. She doesn't go into the hospital, but she does have someone to look after us there. There is good communication between the doctor and hospital." Several participants described frustration with receiving contradictory advice from different health care providers:

The insulin was not helping. They took me off insulin and put me on metformin. Now my glucose is really, really high. They have me on another pill and I am a lot better ... They blamed it on one doctor, because he put me on the insulin. I didn't like that they disagreed.

## Inter-Professional Care

Involvement and collaboration from multiple health care disciplines was noted repeatedly. Participants, especially those describing multiple physical health and mental health comorbidity, identified the importance of good communication and collaboration among their health care providers for achieving satisfactory health care outcomes:

My doctor is always doing what he can to help me. He phones around to get services for me. He gets a physiotherapist for me. He also gets nurses for me and these nurses call me. So I like my doctor.

Based on these focus group findings, enhancements to the Curriculum of Caring have been implemented.

## Limitations of the Study

Investigators identified a number of potential problems related to using focus groups to solicit perceptions. There was a risk of individuals being

unwilling to share experiences with supports or a group of peers (some known and some new). Facilitators provided instructions at the start of each focus group around the confidentiality of information among participants. Facilitators provided an option of providing written responses (supported if needed) for experiences that would be uncomfortable to share within a group.

There was an identified risk that coaching or comments from supports could influence participant discussion. Supports were informed prior to and at the beginning of the focus group discussion that their role was to support and not to share their own perspectives of the participants' health care experiences.

There was a potential risk that dominant opinions would stifle divergent opinions. Facilitators encouraged and validated differing perspectives. To determine whether this was a factor, investigators analyzed the verbal comments made by the participants who also submitted written comments. There was a high correlation between written and spoken comments. Investigators also analyzed transcripts within each of the focus groups and found that each group reported varied and divergent opinions about health care experiences.

There was a risk that the sample size might have been insufficient to identify a diversity of experiences. Investigators analyzed the transcripts and found that no new themes emerged (saturation) after the fourth focus group, indicating reliable results.

Obtaining a full representation of opinions of such a diverse group of people as those with DD is a challenge. Our research and findings excluded a number of important sub groups of the population including: people who have extreme difficulty communicating, people not actively being supported by a government funded developmental service provider, people faced with transportation barriers, people who could not participate due to lack of supports and people with significant emotional or behavioural issues.

## Discussion

This research reinforces the invaluable benefits of directly involving people with developmental disabilities in research related to the sup-

ports and services they receive to live full and healthy lives. Focus group participants identified important attitudes and behaviours for health care providers to develop during their training and practice when interacting with patients. While these attitudes and behaviours are important in the provision of good health care to all citizens, they are essential when providing support to this population who tend to have more complex physical, medical, and mental health needs. People with lived experience can tell us more about what they need to improve the health care experience. They highlight the need for better access, caring attitudes, respectful professionalism, adapted communication, person-centred practices, good outcomes, coordinated and continuity of care. Attending to the voices of experience brings us closer to the ideals of bridging gaps in service and reducing health service inequity.

This research provided valuable personal perspectives with patient-centred recommendations to guide the enhancement of the Curriculum of Caring for People with Developmental Disability. This curriculum has been developed through a partnership of McMaster University, Michael G DeGroote School of Medicine (Niagara Region Campus) and Brock University Centre for Applied Disability Studies, Department of Nursing and Bethesda Services. Input from this focus group study has been used for curriculum refinements for broader health care training. Components of Curriculum of Caring can be accessed at [CommunicateCARE.machealth.ca](http://CommunicateCARE.machealth.ca). They have also been incorporated into medical/health care education at the University of Toronto, Centre for Addiction and Mental Health (<https://www.camh.ca/en/education/continuing-education/continuing-education-programs-and-courses/developmental-disabilities-and-mental-health-online-course>) and toolkits developed by Ontario's Health Care Access Research and Developmental Disabilities (HCARD). The HCARD tools can be accessed at <https://www.porticonetwork.ca/web/hcardd>.

## Key Messages From This Article

**People with disabilities.** You deserve good health care. Good health care is provided by professionals who listen to you and involve you in health care decisions. It is important

that your voice is heard by health care providers. This study is a way for health care students and professionals to hear your voice.

**Professionals.** People with DD are an important source of information for evaluating and improving health care services. People with DD can contribute greatly to improving communication and clinical practices that are mutually beneficial. This study has identified key provisions, attitudes and behaviours for effective health care provision to people with DD.

**Policymakers.** Attitudes and behaviours of health care providers have a direct impact on health care experiences and outcomes. People with DD are at greater risk for poor health (complex medical and mental health concerns) and require extra consideration and provision to overcome barriers associated with communication and cognitive limitations. Attitudes and behaviours can be developed and enhanced during health care education and are an important component of ensuring not only good health care experiences but also outcomes for this population. People with DD should be involved in advising about provision of healthcare as well as training for health care professionals.

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## Appendix 1: Focus Group Questions

We have eight main questions. We might ask you for extra information to better understand your answers.

1. Think about being outside. What is your favourite season (Fall, Winter, Spring, Summer) and why is it your favorite?
2. Think about a good health care experience. This could be with a doctor or a nurse. This could be in an office or a hospital. What made it a good experience?
3. Think about a bad health care experience. This could be with a doctor or a nurse. This could be in an office or a hospital. What made it a bad experience?
4. What would you tell students to help them be better health care workers?
5. What do you feel about health care students in the room during appointments?
6. Is there anything else you would like to tell us about your health care experiences?
7. Do you have any questions for us?
8. Is there anything else you would like to add?

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## Family Quality of Life When There is More Than One Child With a Developmental Disability

### Abstract

*This study explored whether Family Quality of Life (FQOL) differs in families with two or more children with developmental disability (DD) compared to families with only one child with DD and examined predictors of FQOL including child, family, and context variables. The current study used convenience sample information from a nationwide survey completed by 209 parents of children with severe DD across Canada. The two subgroups were compared using independent t-tests and, contrary to our original hypothesis, families with more than one child with DD had significantly higher FQOL ( $t(177) = -2.35, p = .02$ ) with a medium effect size. However, a hierarchical regression revealed that no additional variance was accounted for by whether there were one or multiple children with DD in the family after accounting for other child, family and parent, and context variables. These findings suggest that having two or more children with DD has no major negative impact on families' QOL compared to having only one child with DD. In conclusion, these caregivers seem to be resilient despite the extra time and effort they devote to their multiple care giving roles. Furthermore, previous care giving experiences may have enhanced their resiliency to cope positively with their current circumstances.*

This study aimed to include participants with many different developmental disabilities (DD). The term DD will therefore be used as an umbrella term to encompass children or adolescents with an intellectual disability (ID), autism spectrum disorder (ASD), physical disability (e.g., cerebral palsy, seizures) and all other forms of disabilities, such as Down syndrome, fragile X syndrome, etc. Indeed, many individuals with one disability, for instance ID, often have a co-morbid diagnosis of another, such as ASD, including the individuals in the present study.

Previous research has suggested that families of children with ASD or other DDs are at risk for higher levels of distress or less optimal Family Quality of Life (FQOL) compared to families of typically developing children (Baker-Ericzén, Brookman-Frazee, & Stahmer, 2005; Cuzzocrea, Larcán, Baiocco, & Costa, 2011; Hastings, Kovshoff, Ward, Espinosa, Brown, & Remington, 2005). Care of a child with a DD places additional financial, psychological, and interpersonal demands upon the members of the family (Goudie, Narcisse, Hall, & Kuo, 2014; Majnemer, Shevell, Law, Poulin, & Rosenbaum, 2012). In particular, parents of children with ASD not only report higher levels of stress but are also at a higher risk for mental illnesses such as major depressive disorder. Additionally, parents of children with ASD commonly report feeling incompetent as caregivers, habitually frus-

trated and tend to isolate themselves (Dunn, Burbine, Bowers, & Tantleff-Dunn, 2001; Lach et al., 2009). Likewise, families of children with a DD have been found to experience lower FQOL compared to families of children without a disability (Brown, MacAdam-Crisp, Mian Wang, & Iarocci, 2006; Gupta, 2007).

It is well known that there is increased risk of having more than one child in the family with ASD as the result of various genetic, epigenetic, and non-genetic factors (e.g., Perry, Koudys, Dunlap, & Black, 2017) and that families can have more than one child with other forms of DD (Percy, Brown, & Fung, 2017). However, the impact of having more than one child with a DD on a family's QOL, has yet to be been investigated.

There are several reasons that the presence of more than one child with a DD in a family might exacerbate the stress already experienced by families of individuals with a DD. The additional stress could be derived from the increased level of dependence and time demands of the children with the DD, or the additional emotional and financial burden (Kuo, Cohen, Agrawal, Berry, & Casey, 2011; McCann, Bull, Winzenberg, 2012). Thus, it is imperative to investigate how the presence of multiple children with a DD affects overall FQOL.

## Objectives

There are two main objectives in the current study; first, we investigated how family quality of life differs between families with two or more children with DD compared to families with only one child with a DD. We hypothesized that families with multiple children with a DD would have lower family quality of life compared to families with only one child with a DD. Second, we examined predictors of family quality of life including child, family and parent, and context factors. Child factors include adaptive and maladaptive skills. Family and parent factors include marital status, parent socialization, mental health, positive experiences related to parenting a child with a DD (positive gain), and marital support and satisfaction. Context factors include median income of residential area, school satisfaction, child socialization, and support from neighbours. We hypothesized that having more than one child with a DD would be a significant predictor of family quality of life, above and beyond these other predictors.

## Methods

This study is part of a larger study, namely the Great Outcomes for Kids Impacted by Severe Developmental Disabilities project (GO4KIDDS). This study was an Emerging Team grant funded by the Canadian Institutes of Health Research (PI: Perry). GO4KIDDS included a number of studies using different methodologies, including several different caregiver surveys. The objective of the overall project was to provide a better understanding about the health, well-being, and social inclusion of school-aged children and youth with severe DDs and the experiences of their families. Recruitment materials stated the sample being invited to participate was parents of children with intellectual disabilities in the moderate, severe, or profound range, with or without autism, with or without physical disabilities, and with or without mental health or behavioural challenges. That is, the sample was intended to include children with significant and multiple needs (see below for specific child characteristics of the sample in the present study). Researchers gained ethics approval from the Human Participants Review Committee at York University and caregivers provided informed consent.

Participants were recruited through approximately 500 agencies across Canada, and postings on websites and social media sites. Over 400 caregivers of children and adolescents with DDs completed the Basic Survey (Perry & Weiss, 2008a) online or by paper and pencil. About half of these families then went on to complete an Extended Survey (Perry & Weiss, 2008b) with additional questions to provide a more in depth understanding of their family dynamics. The present study utilizes data collected from both the GO4KIDDS Basic and Extended Surveys.

Please contact authors if further detail regarding the study methodology is required.

## Participants

The participants in this study were part of the GO4KIDDS Extended Survey that made use of a convenience subsample of 209 caregivers of children and adolescents with severe DDs. Of the participants who completed the survey, 66% reported having one child with a severe DD, while 34% reported having two or more (up to four) children with a DD. If parents reported having more than one child with a DD, they

were asked to complete the survey based on their child with the most severe DD. The following participant details are based on the child with the most severe DD in these families; information regarding the other child(ren) with DD (or additional siblings without a DD) in these families was not collected. The primary individuals with DD ranged in age from 3 to 18 years with a mean of 10.8 ( $SD = 3.5$ ). The 122 individuals who did not have a sibling with a DD had an average age of 10.7 ( $SD = 3.44$ ) and the average age of the 87 individuals who had one or more siblings with a DD was 10.4 ( $SD = 3.36$ ). The majority of the individuals with a DD were male (71.4%). Of the families who had only one child with a DD, 67.5% reported their child being male. Similarly, of the families who had more than one child with a DD, 77.4% reported their child with the most severe DD to be male.

The 209 families included in our analysis inhabited both rural and urban areas from 11 differ-

ent provinces and territories ranging all across Canada. Of these families, the majority of the caregivers completing the survey were the biological parent (91.3%), with the remaining 8.7% being comprised of adoptive parents, stepparents, and grandparents. The caregivers in these families were predominately female (90.7%) and had an average age of 42 years (range = 24 to 57). Over half (57.4%) were employed, and 81.1% reported their highest level of education as beyond high school. Three quarters (75.6%) of the caregivers were married and the median income of the family's neighbourhood was \$65,516.66 (derived from the forward sortation index of their postal code) based on the 2006 census from Statistics Canada (Statistics Canada, 2006).

## Measures

Table 1 provides details about all measures included in the study.

*Table 1. Constructs and Measures Included in Study*

<i>Construct</i>	<i>Name of Measure</i>	<i>Type of Measure</i>	<i># of Items</i>	<i>Scale</i>	<i><math>\alpha</math></i>
<b>Child</b>					
Age	GO4KIDDS Extended Survey (Perry & Weiss, 2008)	Continuous	1	N/A	N/A
Gender	GO4KIDDS Extended Survey (Perry & Weiss, 2008)	Dichotomous	1	1 = Male 2 = Female	N/A
Diagnosis	GO4KIDDS Extended Survey (Perry & Weiss, 2008)	Nominal	1	1 = ASD 2 = ID 3 = other	N/A
* Adaptive Skills	SIB-R-Age Equivalent (Bruininks, Woodcock, Weatherman, & Hill, 1996)	Ordinal	35	4-point	0.95
* Maladaptive Skills	SIB-R (Bruininks, et al., 1996)	Ordinal	8	Standardized score, based on 6 point frequency and 5 point severity ratings for each item, standardized for age. Scores range from most severe (-70) to average range (-10 to +10)	0.80
* construct used in regression			continued on following page		

Table 1. Constructs and Measures Included in Study (continued)

Construct	Name of Measure	Type of Measure	# of Items	Scale	$\alpha$
<b>Parent/Family</b>					
Gender	GO4KIDDS Extended Survey (Perry & Weiss, 2008)	Dichotomous	1	1 = Male 2 = Female	N/A
Relation to child	GO4KIDDS Extended Survey (Perry & Weiss, 2008)	Nominal	1	1 = Biological 2 = Adoptive 3 = Step 4 = Grandparent 5 = Other	N/A
* # of children in family w/ DD	GO4KIDDS Extended Survey (Perry & Weiss, 2008)	Dichotomous	1	1 = 1 child with a DD 2 = 2 or more children with a DD	N/A
* Marital Status	GO4KIDDS Basic Survey (Perry & Weiss, 2008)	Nominal	1	1 = Married or common law 2 = Separated, divorced, single or widowed 3 = Other	N/A
Maternal Employment	GO4KIDDS Extended Survey (Perry & Weiss, 2008)	Nominal	1	1 = Not currently 2 = Full or part time	N/A
* Socialization	GO4KIDDS Basic Survey (Perry & Weiss, 2008)	Ordinal	7	5-point ranging from -2 (much less than others) to +2 (much more than others)	0.81
* Mental Health	Kessler-6 (Kessler et al., 2003)	Ordinal	6	5-point	0.89
* Positive Experience	(Positive Gain Scale [PGS]: Pit-ten Cate, 2003).	Ordinal	7	5-point	0.85
Family Stress	Excerpt from Family Stress and Coping Interview (Nachshen, Woodford, & Minnes, 2003)	Ordinal	11	4-point	0.87
* Marital Satisfaction	GO4KIDDS Extended Survey (Perry & Weiss, 2008)	Ordinal	2	5-point	0.71
* construct used in regression				continued on following page	

Table 1. Constructs and Measures Included in Study (continued)

Construct	Name of Measure	Type of Measure	# of Items	Scale	$\alpha$
<b>Context</b>					
Median Income of neighbourhood (postal code)	Stats Canada (Statistics Canada, 2006).	Continuous	1	N/A	N/A
* SES	Barratt Simplified Measure of Social Status (Barratt, 2012)	Ordinal	2	Based on combination of Education (7 levels) and Occupation (9 categories)	N/A
# of Services	GO4KIDDS Extended Survey (Perry & Weiss, 2008)	Continuous	20	# out of 20	N/A
* School Satisfaction	GO4KIDDS Extended Survey (Perry & Weiss, 2008)	Ordinal	9	5-point	0.92
* Child Socialization	GO4KIDDS Extended Survey (Perry & Weiss, 2008)	Continuous	7	# out of 7 social and recreational activities	0.70
* Neighbour Support	GO4KIDDS Extended Survey (Perry & Weiss, 2008)	Ordinal	1	5-point	N/A
<b>Dependent Variable</b>					
* FQOL	Beach Center FQOL Scale (Hoffman et al., 2006)	Ordinal	25	5-point	.94
* construct used in regression					

## Results

Prior to comparing the FQOL scores in the two subgroups, we examined several potential factors that, if different in the two groups, might confound the comparison. This included child characteristics (age, gender, diagnosis, adaptive and maladaptive behaviour) and SES variables (median income of their neighbourhood, and Barratt (2012) – proxies for SES based on parents' education and occupation). Independent *t*-tests showed no subgroup differences for children's age, adaptive skills, maladaptive behaviour, or SES variables (see Table 2). Also, two chi-squared tests showed there was no difference in the percentage of the two subgroups of children who had ASD (57.4% of families with

one child with DD; 56.5% of families with multiple children with a DD;  $\chi^2 = 0.014$ ; Fisher exact  $p = .51$ ) and no difference in the percentage of the sample who were boys (67.5% versus 77.4%,  $\chi^2 = 1.95$ , Fisher exact  $p = .11$ ). Thus, there seemed to be no differences between the groups on any of the examined child or SES factors that might confound the comparison of the two subgroups.

These two subgroups were then compared on the remaining parent/family (parent mental health, socialization, positive gain, family stress, marital support and satisfaction) and context factors (median income, Barratt SES, number of services, school satisfaction, child socialization and neighbour support). In general, the two groups did not differ with the exception of their amount of posi-

tive experiences as a result of having a child with DD (Positive Gain Scale [PGS]: Pit-ten Cate, 2003), which was significantly higher in families who had multiple children with a DD. There was also

a trend for stress to be lower and neighbour support to be higher in families with two or more children with a DD. These *t*-test comparisons are also provided in Table 2.

Table 2. Comparison of Subgroups With One Child With DD vs. Two or more Children With DD

Group	1 or 2+ children with DD	N	M	SD	t (p)
<b>Child Factors</b>					
Age	One	122	10.7	3.45	0.52
	Two or more	62	10.4	3.36	(p = .61)
Adaptive Behaviour	One	122	42.4	29.64	0.63
	Two or more	62	39.5	31.52	(p = .53)
Maladaptive Behaviour	One	122	-15.6	12.84	0.52
	Two or more	62	-16.7	13.23	(p = .61)
<b>Parent/Family Factors</b>					
Mental Health Problems	One	120	1.2	0.90	0.39
	Two or more	60	1.1	0.85	(p = .70)
Parent Socialization	One	121	-1.13	0.69	-0.25
	Two or more	61	-1.09	0.73	(p = .80)
Positive Gain	One	120	4.04	0.62	-2.03
	Two or more	61	4.25	0.72	(p = .04)
Family Stress	One	121	2.75	0.63	1.89
	Two or more	60	2.55	0.69	(p = .06)
Marital Support & Satisfaction	One	98	7.52	1.64	-0.11
	Two or more	54	7.56	2.14	(p = .91)
<b>Context Factors</b>					
Median Income	One	121	\$63,853.00	\$16,813.73	-1.67
	Two or more	58	\$68,364.95	\$17,239.82	(p = .10)
Barratt SES	One	122	39.6	14.04	1.80
	Two or more	59	35.4	15.65	(p = .07)
Number of Services	One	122	10.60	3.55	0.22
	Two or more	61	10.48	3.47	(p = .82)
School Satisfaction	One	118	3.53	1.02	0.25
	Two or more	60	3.49	1.09	(p = .80)
Child Socialization	One	93	19.43	4.49	-0.08
	Two or more	47	19.49	4.23	(p = .94)
Neighbour Support	One	98	2.41	0.74	-1.80
	Two or more	50	2.66	0.92	(p = .07)
<b>Dependent Variable</b>					
Family Quality of Life	One	119	3.58	0.63	-2.35
	Two or more	60	3.82	0.71	(p = .02)

Finally, to test hypothesis 1, we compared the Family Quality of Life score (Hoffman et al., 2006), between groups. Contrary to our original hypothesis, families in the group with two or more children with a DD reported a significantly *higher* family quality of life score. See last line of Table 2.

In order to address the second research question, which was to explore factors related to FQOL, and choose variables for the regression analysis, including child factors, parent/family, and context variables, we began by examining relevant correlations. Child factors were examined first to see whether they were related to FQOL. Child's age was not correlated with FQOL ( $r = -.057$ , ns) and a scatter plot revealed no non-linear relationship with age. Child's diagnosis did not seem to be strongly related to FQOL either as the FQOL scores for the group with ASD, though slightly lower, did not differ significantly ( $M = 3.59$ ,  $SD = .63$ ) from the group without ASD ( $M = 3.77$ ,  $SD = .70$ ;  $t(190) = 1.90$ ,  $p = .06$ ). However, children's level of adaptive skills was significantly, though weakly, correlated with FQOL ( $r = .20$ ,  $p = .005$ ) as were maladaptive behaviour problem scores ( $r = .35$ ,  $p < .001$ ). Thus, in terms of child characteristics, child age and diagnosis were unrelated to FQOL, but higher FQOL was associated with higher adaptive skills and lower levels of problem behaviour.

Parent and family variables (parent mental health, socialization, positive gain, family stress, marital support and satisfaction) as well as contextual factors (median income, Barratt SES, number of services, school satisfaction, child socialization and neighbour support) were also examined in relation to FQOL scores using either Pearson correlations for continuous variables or independent  $t$ -tests as appropriate (see Table 2). Most of the parent/family variables were significantly related to FQOL. Parent mental health problems and family stress were negatively correlated with FQOL ( $r = -.47$ ,  $p < .001$  and  $r = -.44$ ,  $p < .001$  respectively). Marital satisfaction, parent socialization, and positive gain were all positively correlated with FQOL ( $r = .53$ ,  $p < .001$ ,  $r = .31$ ,  $p < .001$ , and  $r = .48$ ,  $p < .001$  respectively). Married respondents ( $n = 143$ ,  $M = 3.72$ ,  $SD = .62$ ) reported significantly higher FQOL than single parents ( $n = 45$ ,  $M = 3.47$ ,  $SD = .78$ ,

$t(186) = 2.30$ ,  $p = .023$ ). However, there was no difference in FQOL scores between respondents who were employed or not currently employed (3.62 vs. 3.70,  $t(190) = -.81$ ,  $p = .42$ ). Contextual factors were somewhat related as well. School satisfaction was correlated with FQOL ( $r = .37$ ,  $p < .001$ ) as was children's activity participation ( $r = .33$ ,  $p < .001$ ). The two SES measures were only weakly related to FQOL ( $r = -.17$ ,  $p = .02$  for Barratt SES, and  $r = .08$ ,  $p = .296$  for median neighbourhood income).

Next, a hierarchical multiple regression analysis was performed in order to determine how child, parent/family, as well as context variables contributed to the overall FQOL score, and whether families having one child with a DD versus having more than one child with a DD accounted for any unique variance in FQOL above and beyond predictors in the other steps of the model. Only pertinent variables that were shown to be associated with FQOL were included in the regression analysis. Results are shown in Table 3. Child factors, specifically adaptive and maladaptive behaviours, were entered as the first predictors (step 1), followed by parent and family factors (parent socialization, mental health, positive gain, marital status, satisfaction and support) (step 2). Next, contextual factors were entered into the model (child social participation, school satisfaction, total Barratt score and neighbour support) (step 3) and finally having one child with DD or multiple children with DD in the family was entered as the final step (step 4).

The results from step 1 indicate that child factors significantly contributed to the regression model and accounted for 16% of the variance. Introducing the parent and family variables at step 2 accounted for an additional 34% of the variance and this change was significant. At this step, all child variables remained significant. Furthermore, of the family variables, positive gain, marital support, and parental mental health were significant. Next, the addition of context variables at step 3 accounted for another 5% of the variance and this change was significant. At this step, parent mental health was no longer significant but neighbour support, child variables, positive gain, as well as marital support remained significant. Finally, the addition of the one child with DD or multiple children with DD in the family variable at step

Table 3. Predictors of Family Quality of Life

Step	Predictors	$\Delta R^2$	$\beta$	<i>p</i>
1	<b><u>Child Factors</u></b>			
	Adaptive Behaviour	$R^2 = .162$	.132	.062
	Maladaptive Behaviour		.151	.046
2	<b><u>Parent &amp; Family Factors</u></b>	<b>.338</b>		
	Parent Socialization		-.068	ns
	Mental Health	<i>F</i> for change	-.125	ns
	Positive Gain	$p < .001$	.194	.011
	Marital Status		.020	ns
	Marital Support & Satisfaction		.363	< .001
3	<b><u>Context Factors</u></b>	<b>.054</b>		
	Child Socialization		.142	ns
	School Satisfaction	<i>F</i> for change	.126	ns
	Total Barratt Score	$p = .017$	-.094	ns
	Neighbour Support		.134	.050
4	1 or 2+ Children with DD	<b>.014</b>	<b>.014</b>	ns
		<i>F</i> for change		
		$p = .073$		
Final Model		<b>Final <math>R^2 = .567</math></b>		
$F(12,104) = 11.37, p < .001$				

4 accounted for only 1% of additional variance and therefore was not significant. The final model (see table 3) accounted for a substantial 57% of the variance in FQOL ( $F_{(12,104)} = 11.37, p < .001$ ) and included the following significant predictors: maladaptive behaviour, positive gain, marital support, and neighbour support.

## Discussion

The purpose of this study was to report on the FQOL experienced in families with more than one child with DD. We compared families who had multiple children with DD to families who had only one child with a DD. With regard to our first objective, our hypothesis that families who have multiple children with DD will have a lower FQOL than families with only one child with a DD, was rejected. Surprisingly, families with two or more children with a DD reported experiencing a slightly higher level of FQOL than families with only one child with a DD. Furthermore, with regard to our second objective, our hypothesis that having more than one child with a DD would be a significant

predictor of FQOL, after taking other variables into account, was also rejected. Moreover, the regression indicated that 16% of the variance was accounted for by child factors, an additional 34% by parent factors, and a further 5% by context factors. However, there was no additional variance accounted for by whether there were one or multiple children with DD in the family.

These findings may suggest that, after having the initial child with a DD, additional caregiving roles may not add significant additional strain on the family. Rather, additional caregiving roles may be a straightforward adaptation, as these families have already experienced caring for an individual with a DD. Furthermore, these families may now have a better understanding of their additional child's needs and have possibly become experts at their caregiving roles. For instance, these caregivers may have become better prepared and equipped with knowledge about the service system, advocacy skills, and empathy required to care for an individual with a DD. Thus, pertinent coping skills and caregiving skills may have already been developed after caring for an initial child with a DD.

Similar to previous reports of having one child with a DD (McMillen & Fisher, 1998), the Positive Gain Scale was found to be a significant predictor of FQOL in families with more than one child with a DD. Furthermore, scores on the Positive Gain Scale, which assesses positive experiences associated with raising a child with DD, were higher in families with more than one child with DD. This may be a result of families who have more than one child with DD gaining an even stronger sense of purpose and fulfillment in their lives. Additionally, an optimistic perspective may be an integral part of a parent's ability to cope with the presence of a DD in family members.

### Limitations and Future Research

Although this study is statistically sophisticated and examines information that very little research has attempted to investigate before, there are limitations present. For instance, the current study relied on self-report measures and convenience sampling methodology, which can lead to biases in the results. In addition, the current study is cross sectional and therefore does not reflect dynamic factors. FQOL may change over time and researchers may find cumulative effects of variables. Therefore, longitudinal research is needed to examine the long-term impact of caring for multiple individuals with DD. Likewise, in order to gain a more comprehensive understanding of families who care for multiple children with DD, further in-depth, and perhaps qualitative research may prove beneficial. Other factors that might be advantageous to examine in order to gain a more comprehensive understanding of FQOL in families with one or more children with DD include caregivers' total time spent caring for their children, use of specific coping strategies, specific diagnoses, sibling diagnosis and dependence level, as well as the dependence level and number of siblings without DD in each family.

Managing multiple caregiving roles undoubtedly presents considerable challenges for parents. However, the caregivers in the present study seem to be resilient despite the extra time and effort they presumably have to devote to their multiple caregiving roles. Furthermore, previous caregiving experiences may have enhanced their resiliency to cope positively with their current circumstances. This knowledge helps

us better understand the context of families caring for more than one child with DD and has important clinical relevance. Moreover, the identification of potential risk factors and protective factors associated with FQOL will allow professionals to provide services that are more catered to the needs of families who have multiple children with a DD. As well, this knowledge may help alleviate stress, and enlighten families on what they can expect with regard to having one or multiple children with DD.

### Key Messages From This Article

**People with disabilities.** It is important for people with disabilities and their families to have good quality of life. Different people might have different ideas about what good quality of life is, but usually it means things like being healthy, happy, having friends, having enough money, etc. In this study, families who have more than one child with a disability told us that their quality of life was just as good, or even better, than families who had only one child with a disability.

**Professionals.** A subset of parents we work with have more than one child with a disability. It is important to bear this in mind for goal setting and family work with these families. Parents who have multiple children with disabilities may be quite resilient, in spite of having many challenges, and do not necessarily experience less optimal Family Quality of Life than parents with only one child with a disability.

**Policymakers.** It is important to remember that a substantial proportion of families of children with disabilities have more than one affected child. This may require reconsideration of certain policies (e.g., waitlist management).

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## **"He's on the Streets, and Stealing, and Perpetuating the Cycle... and I'm Helpless": Families' Perspectives on Criminality in Adults Prenatally Exposed to Alcohol**

### **Abstract**

*Despite the high rate of criminality in adults with fetal alcohol spectrum disorder (FASD), little is known about the risk and protective factors associated with criminal behaviour in this population. Semi-structured interviews were conducted with eight families of adult children with prenatal alcohol exposure (PAE) to better understand the factors that increase or decrease criminality. Families identified four risk factors that contributed to their adult child's trouble with the law, including difficulty with self-regulation, negative influences from the peer group, substance use, and multiple transitions from one living environment to another (e.g., living with different caregivers, residing in group homes, incarceration, or participating in inpatient treatment programs). Protective factors that were reported to help mitigate the effects of the legal issues were structure and supervision, educational and occupational success, access to clinical and financial support, and having a strong network of positive influences. The findings from this study emphasize the need for more family-centred interventions to improve the outcome for adults with PAE.*

The teratogenic effects of alcohol cause lifelong physical, cognitive, and behavioural impairments that are collectively called fetal alcohol spectrum disorder (FASD). As many as 60% of individuals with FASD will experience trouble with the law (Streissguth, Barr, Kogan, & Bookstein, 1997) and a systematic literature review has established that individuals with FASD are 19 times more likely to be incarcerated than members of the general population (Popova, Lange, Bekmuradov, Mihic, & Rehm, 2011). Although current models of criminality in FASD emphasize the poor fit between an individual's capabilities (e.g., neurocognitive impairments) and the environmental demands (e.g., lack of structure, multiple transitions such as moves to foster homes or group homes, kinship placements, not having a fixed address, or temporary placement in residential programs or correctional facilities; Brown, Connor, & Adler, 2012; Malbin, Boulding, & Brooks, 2010), few studies have examined the contributing role of the family system and psychosocial milieu of the individual. The aim of this qualitative study is to explore the risk factors and protective factors associated with criminality identified by families of adults with prenatal alcohol exposure (PAE).

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## Fetal Alcohol Spectrum Disorder

Fetal alcohol spectrum disorder (FASD) is an umbrella term that describes the range of effects of prenatal exposure to alcohol (Cook et al., 2015). Estimates vary, but the prevalence of FASD in Canada is estimated to be 9.1 in 1,000 live births (Health Canada, 2017), while the prevalence of FASD in the United States and Western Europe has been estimated to range from 2% to 5% of the population (May et al., 2009). In 2018, the estimated prevalence of FASD among first-graders in four U.S. communities was found to range from 1.1% to 5.0% using a conservative approach, but the findings may not be generalizable to all communities (May et al., 2018). FASD is considered to be the leading preventable cause of intellectual disability in North America (O'Leary et al., 2012).

Although the direct mechanism whereby alcohol affects the developing fetus remains unclear, the permanent teratogenic effects of alcohol have been well documented (Riley & McGee, 2005). Factors such as quantity of alcohol consumed, number of exposures to alcohol and timing of exposure, individual differences in the tolerance of alcohol, poor nutrition during pregnancy, and exposure to other intoxicants can affect fetal development and lead to congenital impairments that are attributed to organic brain damage (Astley & Clarren, 2000; Streissguth et al., 1997). There is extensive evidence that PAE leads to cognitive and behavioural impairments, termed "primary disabilities" or adverse life outcomes, even in the absence of physical characteristics (Connor, Sampson, Bookstein, Barr, & Streissguth, 2000; Rasmussen, 2005). There is a wealth of research that suggests that individuals with FASD have significant impairments in executive functioning (Pei, Job, Kully-Martens, & Rasmussen, 2011; Vaurio, Riley, & Mattson, 2011), adaptive behaviour (Brown et al., 2012; Carr, Agnihotri, & Keightley, 2010), academic abilities (Rose-Jacobs et al., 2012), and social skills (Kully-Martens, Denys, Treit, Tamana, & Rasmussen, 2012).

In addition to the adverse life outcomes associated with CNS dysfunction, many individuals with FASD are exposed to adverse environmental factors postnatally such as trauma, instability in the home environment, living in an impoverished environment, and inconsistent

parenting styles (Henry, Sloane, & Black-Pond, 2007; Olson, O'Connor, & Fitzgerald, 2001). Furthermore, one publication has reported that only 20% of individuals with FASD reside with their biological mothers (Streissguth et al., 2004). When cognitive and behavioural impairments are accompanied by an adverse environment, the interaction of these vulnerabilities may lead to the development of additional problems, labelled "secondary disabilities" or secondary impacts, such as mental health disorders (90%), disrupted school experience (61%), legal problems (60%), and confinement (50%; Clark, Lutke, Minnes, & Ouellette-Kuntz, 2004; Streissguth et al., 2004).

Historically, research has sought to delineate the risk factors associated with secondary impacts in FASD, yet few studies have examined the protective factors that contribute to positive life outcomes. One longitudinal study identified an array of protective environmental factors for individuals with FASD, including having a stable and nurturing home environment, having basic needs met, never experiencing violence, receiving a diagnosis of FASD before the age of six, receiving a diagnosis of FAS, and receiving supports and services (Streissguth et al., 1997; 2004). Furthermore, Canadian researchers have noted that factors such as having stable employment, having a strong social network, having access to supports and services, and living independently may help reduce the effects of secondary impacts for adults with FASD (Clark, Minnes, Lutke, & Ouellette-Kuntz, 2008). Taken together, the findings from previous research suggest that although individuals with FASD have biological predispositions that cannot be altered (i.e., adverse life outcomes), the presence of protective factors can help prevent the emergence of adverse life outcomes such as criminality and incarceration (i.e., secondary impacts).

## Criminality in Individuals With FASD

Individuals with FASD are overrepresented in the criminal justice system (Burd, Selfridge, Klug, & Juelson, 2003; Popova et al., 2011). In a study conducted in British Columbia, Canada, a cohort of 287 incarcerated youth between the ages of 12 and 18 participated in a foren-

sic psychiatric or psychological assessment. The researchers found that of the individuals screened, 23.3% met the criteria for a diagnosis of FASD; however, only 1% had been diagnosed prior to the mandated assessment (Fast, Conry, & Looock, 1999). The findings from this study suggest that many individuals with FASD are undiagnosed prior to their involvement with the criminal justice system and are therefore precluded from accessing the supports and interventions that could help reduce the likelihood of criminality.

The adverse life outcomes experienced by individuals with FASD often lead to higher rates of problematic behaviour, such as lying, cheating, disobedience, and stealing, which could put them at increased risk of engaging in criminal acts (Nash et al., 2006). Furthermore, impulsivity (Schonfeld, Paley, Frankel, & O'Connor, 2006), aggression (Brown et al., 2012), difficulty understanding social cues (Kully-Martens et al., 2012), and limited understanding of cause and effect (Fast & Conry, 2004) create a susceptibility to peer pressure and victimization, violation of rules, and understanding the consequences of their actions (Brown et al., 2012; Chudley, Kilgour, Cranston, & Edwards, 2007). Whereas behaviours that violate social norms are typically ascribed to antisocial attitudes and defiance, in the case of FASD there is strong consensus among researchers, clinicians, and policy makers that the challenging behaviours exhibited by individuals with FASD are not willful but rather a consequence of neuropsychological deficits (Brown et al., 2012; Malbin et al., 2010). Indeed, it appears that the majority of criminal acts perpetrated by individuals with FASD are reactive and opportunistic rather than premeditated (Brown et al., 2012).

Malbin and colleagues (2010) proposed a neurobehavioural model of criminality in FASD in which neurobehavioural deficits are regarded as important contributors to criminal activity as a result of a poor fit between the individual's abilities, their environment, and the expectations placed upon them. Rather than focusing on changes that involve a person's deficits, proponents of the neurobehavioural model argue that effective treatment for offenders with FASD necessitates accommodations geared towards the individual's unique needs as well as their strengths (Burd, Fast, Conry, & Williams, 2010;

Malbin et al., 2010). By increasing the congruence between the individual's neurobehavioural profile and the environmental demands, a preventative approach based on skill building and enhancing protective factors can be employed to mitigate the impact of secondary impacts such as trouble with the law (Henry et al., 2007).

## Familial and Environmental Factors

Streissguth and colleagues (2004) found that the caregiving environment had a substantial impact on the outcome of individuals with FASD. Nevertheless, relatively little is known about the specific challenges and stressors that may affect the home environment (Olson, Rudo-Stern, & Gendler, 2011; Watson, Coons, & Hayes, 2013). The bulk of research has examined the impact of raising a child with FASD through the lens of parenting stress. A study examining parenting stress in 42 biological mothers of children with FASD found that higher levels of child externalizing behaviours (e.g., aggressive, oppositional, or defiant behaviour) and lower levels of parental support predicted greater maternal stress (Paley, O'Connor, Kogan, & Findlay, 2005). A subsequent study conducted with both biological and adoptive parents found that increased levels of parenting stress were associated with greater child externalizing behaviours, internalizing behaviours (e.g., anxious or depressed behaviour), and larger impairments in executive functioning and adaptive behaviours (Paley, O'Connor, Frankel, & Marquardt, 2006).

Similarly, qualitative studies have identified stressors reported by parents, including difficulty managing challenging behaviours and providing safety for their child (Caley, Winkelman, & Mariano, 2009), fear for the child's future (Watson, Coons, Hayes, & Radford-Paz, 2013), and insufficient formal and informal supports (Brown & Bednar, 2004). Collectively, studies on parenting stress suggest that caregiving stress is associated with dysregulated behaviour in children and a lack of support and resources for families (Jirikowic, Olson, & Astley, 2012). While research has focused on the stressors faced by caregivers of children with FASD, little is known about the factors that contribute to resiliency and positive life outcomes. There is a need for further qualitative research in order to include the voice of families in research on FASD, with the goal of gaining an understanding of their

experiences and identifying factors that may decrease the likelihood of criminality (Olson et al., 2011; Watson et al., 2013).

## Method

A qualitative study consisting of semi-structured interviews was undertaken to explore the factors associated with criminality in adults with PAE. This study was approved by the Laurentian University Ethics Board, in accordance with the Canadian Tri-Council Recommendations for Research with Human Participants. Pseudonyms were used in reporting.

## Participants

Participants were recruited from various FASD groups across Ontario, Canada employing convenience sampling techniques. Thirty-three online FASD groups were contacted via e-mail inviting families who were interested in participating in the study to contact the researcher by telephone or e-mail. Individuals known to the researcher were also contacted and invited to participate in the study. This latter sampling technique, called respondent-driven sampling, is commonly utilized with hard-to-reach populations (Benoit, Jansson, Millar, & Phillips, 2005). Eight families from Ontario, Canada, participated in this study. In order to partici-

pate, caregivers had to have at least one adult child, 18 years of age or older who had been exposed to alcohol prenatally. One biological mother, five adoptive mothers, and two custodial grandmothers were included in the study (herein referred to as parents or caregivers). The age of the adult children ranged from 19 to 42 years, with an average age of 29.8 years. Six interviews were conducted with only the caregiver present, while two interviews consisted of parent-child dyads. See Table 1 for demographic information of the participants.

## Qualitative Interviews

The semi-structured interviews employed a basic interpretive qualitative approach, which seeks to understand how individuals interpret and make sense of their lived experiences (Merriam, 2002). This methodology is grounded in the principles of phenomenology and symbolic interactionism and provides a richly descriptive account of the individual's perspective of a situation. The interviews consisted of a blend of less and more structured questions, such as "How would you characterize your child's relationship with the law?" and "Can you describe any strategies or supports you used to help you deal with your son/daughter's challenging behaviours?" (Appendix A). The questions for the interviews were based on the

*Table 1. Demographic Information for Participants*

<i>Participant and Pseudonym</i>		<i>Family Type</i>	<i>Age of Adult Child</i>
<i>Parent</i>	<i>Adult child</i>		
Lauren	Isabelle	Biological	38
Sadie	Maria	Adoptive	33
	Dawson	Adoptive	35
Emma	Jordan	Custodial grandmother	22
Caroline	Noah	Adoptive	22
Kate	Cody	Adoptive	32
Tara	Gavin	Adoptive	27
Leah	Logan	(Biological)	42
	Chloe	Custodial grandmother*	19
Audrey	Serena	Adoptive	38

\* This participant's primary caregiving role was as a custodial grandmother and considered this to be her family type.

universal risk and protective factors identified in previous research (Streissguth et al., 1997; 2004). The length of the interviews ranged from 45 minutes to 2 hours. With permission of the participants, interviews were digitally recorded to allow for subsequent transcription. To protect participant confidentiality, all names and identifying information have been changed.

## Data Analysis

Interviews were analyzed using interpretive phenomenological analysis (Storey, 2007), which aims to capture the meaning participants ascribe to their experiences. The systematic process of analysis involves multiple reviews of verbatim transcripts by recording general comments in the margins and gradually identifying commonalities in participants' experiences and overarching themes (Smith & Eatough, 2007). Families were given the opportunity to ensure that their words were not misrepresented through the use of member check, whereby a summary of themes is provided to participants and the content of the interview is synopsisized to ensure that the information was accurately understood by the interviewer.

## Results

The qualitative analysis of the interviews yielded four superordinate themes related to risk factors for criminality and four superordinate themes related to protective factors. Each theme and the corresponding subthemes are discussed in detail to provide an overall picture of families' perspectives of the risk and protective factors associated with criminality.

### Risk Factors for Criminality

Contributing factors fell under four superordinate themes: difficulty with emotional and behavioural self-regulation, having a negative peer group, substance use, and multiple transitions.

**"He had uncontrollable rages": Emotional and behavioural self-regulation.** Families attributed their adult children's involvement with the law to underlying difficulties with self-regulation and inhibitory control. Seven of the eight families reported that regardless of their child's age, there was a lifelong pattern of poor

frustration tolerance and emotional dysregulation. These seven families described violent outbursts that involved death threats, physical assaults, the use of weapons, and destruction of property. In many cases, the intensity of the aggressive episodes warranted police involvement to keep the individual and family members safe from harm. As Caroline, adoptive mother to Noah, explains, "He would threaten to kill our daughter. That was when he first became involved with the police. Threatened to kill us... we had to call the police sometimes because he wouldn't calm down."

Parents also described how poor inhibitory control, coupled with difficulty understanding the consequences of actions, led to impulsive criminal acts. In fact, every account of offending behaviour reported by the families in this study was opportunistic and reactive, not having been planned beforehand. Gavin, a young man with FASD, explained how impulsivity and difficulty in understanding abstract concepts such as ownership contributed to his criminal behaviour:

When I was younger I didn't realize the consequence of ownership type of thing so I tended to sometimes go into people's house and be like 'oh that's shiny, that's nice' and I would take it and I wouldn't really feel... bad about it and then I would go in the grocery store or something and say "I want that but I don't have any money, yoink!"

**"Needed to belong, needed to be accepted": Peer group.** Families also reported that social difficulties contributed to their child's criminal behaviour and that their children often experienced rejection by their peers. Caroline, an adoptive mother to a 22-year-old son with PAE, stated:

[H]e seems to have friends for a while, and all of a sudden they disappear and we don't know why. He said to me the other day 'I decided all my friends are not really my friend except for two.' One was a girl that we've met once. And the other was a guy that we haven't seen for maybe two years now.

Previous research has hypothesized that impairments in social skills may interfere with the ability to establish and maintain positive relationships (Kully-Martens et al., 2012). In par-

ticular, impulsivity, disregard for boundaries, hyperactivity, and disruptive behaviour may lead to rejection by well-adjusted peers. Without a positive peer group to turn to, individuals with PAE may be vulnerable to negative peer influences. Parents observed that peers who typically befriended individuals with PAE do not value appropriate social behaviour; rather, they tend to engage in behaviour such as substance use, truancy, and criminality. Six parents noted that their children's criminal behaviour occurred in the presence of peers and families expressed concern that their children's criminality was influenced by their group of friends. Leah, a biological mother to Logan, worried that involvement in the criminal justice system exposed her son to a group of peers who have a history of criminality, thereby compounding the effect of the negative peer group.

**"It's a battle every day": Substance use.** The contribution of substance use to offending behaviour was noted by seven families. Parents explained that their child's involvement with the law often occurred as a result of intoxication related to alcohol, drugs, or a combination of the two. In the case of Dawson, a 35 year-old man with PAE, criminality was a way to support his addiction. Sadie, his adoptive mother, explained how substance use led to her son's involvement with the law: "He was a mess. He was doing drugs, prostituting himself to get drugs. He just trashed his apartment ... he was on drugs, really trashed it, supposedly about \$4,000 damage so his roommate had to call the police."

Three parents expressed concern that substance use also served as a gateway to other dangerous behaviour, such as stripping, prostitution, and stealing. Furthermore, Lauren stated that substance use interfered with her daughter Isabelle's ability to maintain steady employment and avoid trouble with the law. Six families in this study reported that their child's substance use began between the ages of 9 and 14 years and of these six parents, five reported that they are still concerned about their adult child's drug or alcohol consumption. Consequently, substance use appears to be a lifelong battle for many adults with PAE and an important risk factor for criminality.

**"It was just a continuous chaos": Multiple transitions.** A final contributing factor to crim-

inality discussed by parents was the lack of structure related to multiple transitions from one living milieu to another, including foster or kinship placements, friends' houses, residential treatment programs, juvenile detention centres, and jail. Five parents reported that their children often moved from one household to another, rendering it difficult to have a stable and consistent home environment. For some individuals with PAE, changes in the home environment may include a combination of living with biological, foster, and/or adoptive families. In addition, some parents reported that their children would reside with a multitude of friends for extended periods of times, not having a permanent household for a significant timeframe. For example, Audrey, an adoptive mother to a daughter with FASD, stated, "I don't think I'm exaggerating if I say she lived in 20 different places between the ages 13 and 20." The lack of structure ensuing from multiple transitions often led to unsupervised time, which four families identified as a contributing factor to criminality for individuals with PAE. Furthermore, Caroline indicated that she is unaware of the full extent of her adoptive son's legal problems because of the multiple transitions that he has experienced: "So we ended up kicking him out of the house... He couch surfed for a while. And he had all sorts of times when he was involved with the police that we don't know about."

## Protective Factors

Although families identified risk factors that contributed to their adult child's involvement with the law, caregivers also spoke at length about the protective factors that helped decrease the likelihood that their child's adverse life outcomes would result in criminality. The four superordinate themes identified were the importance of structure and supervision, the need for education and employment that is compatible with the individual's strengths and vulnerabilities, as well as having positive influences.

**"His behaviour is not going to change until you change his environment": Structure and supervision.** According to parental accounts, many instances of criminal activity occurred when there was little to no structure and a lack of adult supervision. Six parents attrib-

uted their children's success to structured environments, stating that their children thrive in environments in which they do not have to engage in planning or organize their time. Along the same vein, three caregivers discussed the importance of supervision to protect their children from adverse life outcomes. Tara, an adoptive mother to a son with FASD explains:

It was always this fine line of saying what his needs are and people saying you're not letting him take enough risk ... you're enabling him and not empowering him ... we're sitting here today saying all the risks I didn't let him take. The fact that he hasn't had a criminal record is because we contained his environment.

**"If he had been able to get an education then I think he'd be able to be a functioning member of society": Education and employment.** Many individuals with PAE are unable to meet the demands in educational and vocational settings (Streissguth et al., 1997); consequently, they are often faced with a lot of unstructured time in adolescence and adulthood. Five parents spoke of the importance of having activities that provide social connections within a structured setting. As Kate, an adoptive mother to a son with FASD, stressed, "He's always looking for work because the biggest, biggest thing that we would notice is that filling his spare time is essential, that he cannot have spare time..." Although these five parents noted that there were challenges in finding vocations tailored to the needs and strengths of the individuals, families also emphasized the positive impact that education and employment had on the well-being and self-esteem of their children.

Parents reported that their children experienced more positive outcomes when the emphasis was placed on their strengths rather than their vulnerabilities. Although school was a challenge for most individuals with PAE, three parents stated that their children had greater success in environments that focused on practical skills instead of academic abilities. Furthermore, six families expressed the critical need to build a partnership with teachers and principals in order to support the specific needs and learning styles of individuals with PAE.

**"They really wanted him to succeed in this endeavour and not end up in jail": Supports.**

Beyond the strategies employed by parents to support their children, families also emphasized the importance of having access to clinical and financial resources in order to mitigate the effects of secondary impacts. Although there was overwhelming consensus among families that the services available were insufficient to adequately meet the needs of individuals with PAE and their family members, caregivers remained appreciative of the assistance they had received. In part, families spoke of the programs that were available to assist their children in gaining their independence in adolescence and adulthood. Parents also spoke of the importance of having supports for family members to counteract the feelings of hopelessness, frustration, and isolation experienced by parents of individuals with PAE.

**"That helps because you know you're not the only one": Supports for the family.**

All caregivers spoke of the importance of having supports for family members. Five parents expressed that formal supports such as parent support groups helped decrease the sense of isolation and hopelessness. Perceived benefits of formal supports for families were twofold; first, the supports provided an avenue where caregivers could learn strategies or techniques that helped them respond to their child's behaviour more effectively, and second, it also helped them share their experiences with other families who truly understood what it was like to raise a child with PAE. Parent support groups allowed parents to shift from a framework of accountability of behaviour and shame to one that takes the CNS dysfunction into account, thereby allowing parents to have more realistic expectations for their children. As Audrey, an adoptive mother to a daughter with FASD, stated:

[O]ne night [at the parent support group meeting] I shared something that Serena had done and one of the mothers whose daughter is living the exact same life that Serena did said to me jokingly, "when are you going to get it? That's what these kids do!" And we laughed. And I thought if I'd been home alone, I'd be bawling my eyes out for days. But because she really understood we laughed about it, like I couldn't believe we laughed about this terrible situation, but it broke the pressure.

**"They really wanted him to be safe. It was nice, it was a miracle": Supports for individuals with PAE.** One of the lifelong challenges described by families of children with PAE was striving for interdependence instead of independence, which entails greater emotional, social, and financial support for daily living (Clark et al., 2008). Three parents discussed the challenge of providing supervision once their adult children moved to a different city for post-secondary studies or employment. Given the dearth of FASD-specific supports, all parents were faced with the challenge of finding services suitable to their child's unique vulnerabilities, including individual counselling, residential programs, child and youth workers, and summer camps. The majority of parents reported having to look beyond community supports to find adequate services, often paying out of pocket. Regardless of the modality of the support provided, four parents reported that targeting adaptive behaviours such as social skills and daily living skills had the greatest impact in terms of preventing criminal behaviour. As Tara, an adoptive mother to a son with FASD, explained:

He's always had a worker to help him with either daily living skills or community involvement. He still has the special services home worker. She helps him with their dishes and laundry and cleaning the apartment. So Gavin's grown up learning to be interdependent. He never had somebody say to him oh you're 16, you should be able to do this. He never had those kinds of comments.

By striving for interdependence rather than independence, parents were able to balance their child's need for autonomy and their need for safety. Furthermore, improving social skills helped individuals with PAE integrate more easily into peer groups that provided more positive influences.

**"I just surrounded her by what I consider really positive forces": Positive influences.** Despite the existing vulnerability to negative pressures, positive peer influences can have an equally important impact on individuals with PAE. Six parents discussed their child's lifelong challenge of finding a positive peer group. Three parents noted that their children benefited greatly from summer camps designed specifically for individuals with disabilities because

it provided a safe and structured environment where they could meet individuals who were at similar developmental levels. Leah, a custodial grandmother to Chloe, spoke of the value of surrounding her granddaughter with role models who instilled hope that she could lead a happy, productive life. Noting the intergenerational patterns that often precede individuals with PAE, Leah emphasized the need to disseminate the message that individuals can overcome challenges and adversity related to PAE and experience positive life outcomes.

Furthermore, parents often acted as a consistent positive influence in their child's life through their role as advocates. Although families faced a multitude of barriers, caregivers exemplified the positive characteristics they hoped to cultivate in their children by remaining optimistic and positive in the face of challenges. Citing the paucity of supports and services, Leah, a custodial grandmother to Chloe, gave up her occupation and dedicated herself fully to seeking resources, educating professionals, and providing financial support for the few services available. Within the legal system, three parents reported having to find lawyers who were either familiar with the lifelong effects of PAE or amenable to learning about FASD, as well as parole officers who were willing to take their adult child's vulnerabilities into account. Kate, an adoptive mother, stressed the need to be tenacious: "And you can't take no for an answer. Like for someone to say, 'sorry we can't afford that' is absolutely an abrogation of every human being's rights." Through their unwavering dedication, parents hoped to protect their children from secondary impacts by increasing awareness of FASD and thereby changing the expectations placed upon individuals with PAE by the educational and judicial systems.

## Discussion

Difficulties with behavioural and emotional self-regulation, combined with environmental factors such as negative peer influences, substance use, and multiple transitions from one place of residence to another can create situations in which the demands placed on the individuals outweigh the capabilities to cope with the exigencies. The results from this study are consistent with previous research findings that

individuals with PAE fare better in situations that are predictable, structured, and do not require decision-making (Brown et al., 2012). While it is important to note that the risk and protective factors identified in this study are similar for all offenders irrespective of whether there was prenatal exposure to alcohol or not, the lack of consistency in routine is particularly problematic for individuals with PAE because they often have difficulty adapting to new environments as a result of adverse life outcomes, such as reduced ability to generalize skills from one setting to another (Malbin, 2004), deficits in executive functioning relating to planning, inhibitory control, and cognitive flexibility (Pei et al., 2011), as well as impairments in adaptive behaviours such as daily living skills and communication (Carr et al., 2012). The results from this study tend to support the neurobehavioural model, which suggests that cognitive deficits, particularly impairments in executive function, interfere with the ability to self-regulate, grasp social cues, control impulsive behaviour, anticipate consequences, and learn from mistakes or the mistakes of others (Coggins, Timler, & Olswang, 2007; Kodituwakku, Handmaker, Cutler, Weathersby, & Handmaker, 1995). Furthermore, environmental demands such as multiple transitions and lack of structure may exceed the individual's capabilities to adapt to situations and consequently, individuals with FASD are more likely to display maladaptive behaviour that may lead to delinquent behaviour (Fast & Conry, 2009; Malbin et al., 2010; Page, 2002).

Where offenders with FASD might differ from typically-developing offenders is in terms of antisocial cognitions, which implies that individuals hold beliefs and attitudes that are in favour of criminality and intentionally engage in illegal activities. There is a wealth of research suggesting that individuals with FASD may not have the cognitive ability to meet legal standards for criminal intent (Brown et al., 2010; Fast & Conry, 2009; Moore & Green, 2004; Roach & Bailey, 2009). From a psycholegal perspective, deliberate criminal behaviour involves decision-making in complex, social situations (McLachlan, Roesh, Viljoen, & Douglas, 2013; Kully-Martens, Treit, Pei, & Rasmussen, 2013). The self-regulation involved in criminality requires individuals to be aware of cues from the social context, rely on cognitive and emo-

tional processes to make decisions, and inhibit behaviour that is in violation of social norms (Brown et al., 2012; Kully-Martens et al., 2013). However, individuals with FASD show marked deficits in these self-regulatory abilities (Connor et al., 2000; Kodituwakku, Kalberg, & May, 2001; Kully-Martens et al., 2012). While criminal behaviour is often attributed to defiance, conduct problems, antisocial attitudes, and lack of motivation, it may be the case that deficits in executive functioning, adaptive behaviour, and social skills may be a more accurate explanation for criminality in FASD (Brown et al., 2012; Malbin et al., 2010).

Parental concerns with the impact of peer influences on their child's behaviour is reflective of existing research, which suggests that the majority of criminal acts perpetrated by individuals with PAE occur within a group setting and rarely occur unaccompanied (Brown et al., 2012). Currently, offenders with FASD are sentenced in the same manner as typically developing peers despite their unique needs. Insofar as it can be ascertained, current rehabilitation and treatment models for offenders that are predicated on a cognitive approach are less effective for individuals with FASD than structured behavioural treatments targeting maladaptive behaviours (Brown et al., 2012). Moreover, as parents in this study indicated, incarceration may have a detrimental effect on individuals with FASD because they are vulnerable to the influence of their peers as a result of adverse life outcomes (Kully-Martens et al., 2012). Without a positive group of friends to turn to, children with FASD have a tendency to seek social connections with negative peer groups. Thus, by placing them in a setting with other offenders, it is likely that individuals with FASD will seek a sense of belonging among other incarcerated criminals.

Families also discussed the protective factors that led to more positive outcomes for their children. In particular, caregivers reported that having a structured environment, finding educational programs or employment opportunities suited to their adult children's strengths and vulnerabilities, having access to supports and services for both the individual with PAE and the family members, and surrounding their children with positive influences were all factors that promoted well-being and decreased

the likelihood of experiencing trouble with the law. Although families differed in how they were able to achieve certain protective factors for their adult children, there was a resonating proclamation from parents that significant tenacity and advocacy were needed in the process.

Parents in this study reported that their adult children required lifelong supports for day-to-day living. Clarren (2000) coined the term "external brain" to describe the role of caregivers as providers of structure and consistency to help individuals with PAE regulate their emotions and behaviour. Indeed, many researchers and clinicians highlight the need to focus on strength-based approaches and environmental accommodations as a way to mediate the interaction between the individual's neurobehavioural profile and the environmental demands (Green, 2007; Malbin et al., 2010). Recognizing the importance of preventative strategies such as reducing the amount of transitions, identifying precipitating factors that contributed to distress, and maintaining a routine, treatment plans for individuals with FASD should therefore delineate strategies for both the individual and the family to cope with unexpected circumstances that may arise (Brown et al., 2012). This message was echoed by the parents who participated in this study, who spoke of the importance of family-based interventions in addition to individual services. Unfortunately, many individuals with PAE do not get diagnosed prior to their involvement with the criminal justice system (Fast et al., 1999) and therefore do not benefit from treatment or clinical services. Consequently, there is a striking need to shift the management of FASD from the judicial system to a more preventative approach that includes both mental health and educational sectors.

Although parents spoke of the importance of having supports and services, many families mentioned the financial burden associated with accessing and maintaining services for their children. Emma, a custodial grandmother, indicated that even when supports were available, many families were unable to access them because they lacked the financial resources. Many times, families had to make considerable sacrifices in order to pay for services for their children. As Kate, an adoptive mother, explained, "Eventually social servi-

ces refused to pay for [the caregiver] so three weeks before he started college I had to sell my house." Given that the caregiving environment has a significant impact on the well-being of individuals with PAE and that parental stress is negatively impacted by the lack of supports, it follows that facilitating access to services is an important part of ameliorating life outcomes for individuals with FASD.

According to the findings from this study, environmental accommodations should target the degree of structure and supervision in the home environment, facilitating access to educational programs and employment, improving emotional, social and financial supports for individuals with FASD and their families, as well as building a strong network of positive influences. In order to meet this demand, more services are needed within the health care and educational sectors in order to employ a preventative approach to criminality in FASD.

## Limitations

Although this study helps elucidate some of the risk and protective factors associated with criminality in individuals with PAE, there are some important limitations. Despite efforts to recruit both offenders and non-offenders for this study, all individuals with PAE had engaged in some type of criminal activity, ranging from shoplifting to physical assault. The difficulty in recruiting individuals with PAE that have had no involvement with the law speaks to the severity of the issue. Previous research has found that at least 60% of adults with FASD experience trouble with the law (Streissguth et al., 1997) and most offenders engage in a repeated pattern of criminality that leads to incarceration (Chudley et al., 2007). Thus, more research is needed to compare offenders and non-offenders with FASD with regards to protective factors from criminality.

This study did not employ quantitative measures to assess executive functioning, adaptive behaviour, or cognitive abilities. As a result, it is impossible to determine the extent to which deficits in these neurobehavioural domains contribute to criminal behaviour in individuals with FASD. Although it is speculated that the difficulties with self-regulation and social behaviour discussed by participants are related to adverse

life outcomes or primary disabilities commonly found in FASD, it is impossible to verify the relationship with the reported deficits employing a qualitative methodology. More research is needed to establish the unique contribution of cognitive and environmental risk factors to the experience of criminality in individuals with FASD.

It is also recognized that the sample in this study may not be fully representative of the heterogeneity of families of individuals with FASD. For instance, only one biological parent participated in the study. Moreover, participants consisted only of mothers or grandmothers. Although the term “parents” and “caregivers” were used to describe participants, it is important to note that the voice of male family members was not represented. While fathers and grand-fathers were invited to participate in the research project, the sample did not include any patriarch family members. It is possible that fathers may have a different perspective than mothers, and it is therefore essential to keep sample characteristics in mind when interpreting the results. Furthermore, it remains unclear how social factors such as socioeconomic status, level of education, gender, family type, race, social support, and parental characteristics such as background, age, and their level of education may also contribute to the unique challenges and opportunities faced by families. Some parents discussed this point, acknowledging that their own education and resources allowed them to access services that may otherwise not have been available to them. Other parents discussed the beneficial role of being part of a support group, whereas some families were not connected to such networks. When relevant, follow-up questions were asked to clarify how support groups may have helped families in enhancing protective factors. Future research with a more multifarious sample is therefore needed to better understand the contributory role of social characteristics on criminality in FASD.

Another limitation of the study was the small number of adults with PAE who participated in this study. While the focus of the research was on the family experience, only two adults with PAE participated in the semi-structured interview. As a result, the majority of the perspectives presented herein are from the caregiver's point of view. Furthermore, it was not feasible to confirm if any of the other adult

children with PAE actually had FASD. A possible avenue for future qualitative research is to explore the experience of adults diagnosed with FASD. Although attempts were made to include the voice of the individuals with PAE, it is important to consider that conducting interviews with parent-child dyads may affect the information disclosed during the interview because participants may not be comfortable disclosing information in the presence of their parent or child. Families were given the choice of interviewing together or separately, and in both cases the family requested to participate in the study together. In light of this, the use of unstructured focus groups in future research may be beneficial.

## Conclusions

The present study explored families' perspectives on the factors that either increased or decreased the likelihood of criminal behaviour in adults with PAE. Families reported that their children faced several risk factors for criminality, including difficulty with self-regulation, vulnerability to negative peer influences, susceptibility to substance abuse, and the experience of multiple transitions. Conversely, families found that structured environments, educational and occupational accommodations geared towards strengths, having access to supports, and increasing positive influences ameliorated life outcomes for individuals with PAE. The findings from this study highlight the importance of including the voice of families in research to inform policies aimed at preventing criminality and developing rehabilitative strategies that accommodate the developmental needs of individuals with PAE.

## Key Messages From This Article

**People with disabilities.** You deserve access to education, employment, and supports that are appropriate for your strengths and needs. These services should include your family and others who have a positive influence on your life.

**Professionals.** Services for individuals prenatally exposed to alcohol should consider the specific adverse life outcomes and secondary impacts that may increase risk of criminality, while enhancing the protective factors that promote more positive outcomes. Interventions should be family-centred.

**Policymakers.** Individuals exposed to alcohol prenatally are overrepresented in the correctional system. There is a need for a provincial strategy focused on prevention, early intervention, family-centered services, and appropriate rehabilitative strategies.

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## Appendix A: Interview Guide for Parents

1. Tell me about your son/daughter. What is he/she like?
2. What is it like being a parent to your son/daughter?
3. When did you first suspect that your son/daughter had FASD?
4. How old was your son/daughter when he/she received the diagnosis of FASD?
5. What kind of supports or services for FASD has your son/daughter received?
6. If you think back to your son/daughter's childhood and adolescence, how would you describe the home environment?
7. Please describe your child's current living arrangement.
8. Before the age of 18, in how many different households did your son/daughter live?
9. What were their living arrangements like in the past?
10. Was there a time in your son/daughter's life when you would say they did not have their basic needs met? (Follow-up: If so, at what ages?)
11. What were the challenges you faced during that time?
12. Can you tell me about the most positive time during your son/daughter's life?
13. At which ages would you describe your son/daughter's living situation as stable and nurturing?
14. Can you describe some of the challenging behaviours your son/daughter exhibited before the age of 18?
15. Can you describe any strategies or supports you used to help you deal with these behaviours?
16. What did you find helpful in dealing with the challenging behaviours?
17. How would you describe your son/daughter's relationship with his/her friends?
18. Has your son/daughter ever been the victim of physical abuse, sexual abuse, or domestic violence?
19. Can you tell me about your child's alcohol or drug use? (Follow-up: Can you describe what that experience was like for you and for your child? Can you tell me at which ages he/she used alcohol and/or drugs?)
20. How would you characterize your child's relationship with the law? (Prompt: problems with police, accused of a crime, convicted of a crime)  
If there is involvement with the law:
  - a. What was the most difficult part of that experience?
  - b. What was the most helpful during that experience?
  - c. What factors do you think contributed to your son/daughter's involvement with the law?
 If there is no involvement with the law, why do you think your son/daughter did not experience any trouble with the law?
21. If you could have any services or supports, which services would you have wanted?
22. How is having access to services meaningful to you? (Prompt: Why do you think having access to services is important?)
23. Do you think that there is something that could have changed the outcome for your son/daughter's life, either in a positive or negative way? (Prompt: Is there anything that could have improved or worsened the outcome of your son/daughter's life?)

## BRIEF REPORT: Staff Experience of an Initiative to Improve Emergency Care for Patients With Developmental Disabilities

### Abstract

*A Toronto emergency department (ED) implemented an initiative to improve emergency care for persons with developmental disabilities (DD). Feedback from ED staff was used to assess the process with respect to: (1) staff awareness of the initiative; (2) staff access to organizational support; (3) burden of the initiative on staff time; (4) fit of the initiative with current practices; and (5) what impact the initiative had on care provided to patients with DD. Overall, staff felt the initiative improved patient care through better awareness and a modified care approach, however some process gaps were identified.*

Adults with developmental disabilities (DD) in Ontario are almost twice as likely to visit the emergency department (ED) than those without a developmental disability (Lunsky et al., 2011). A study by Lunsky, Balogh and Cairney (2012) identified that individuals with DD who visited an ED in the previous year were seven times more likely to visit again. This is particularly challenging for EDs where time and resources are limited, as well as for individuals with DD when their needs are not adequately met. Studies of hospital staff have found that inexperience caring for this population, challenges communicating, and lack of knowledge or understanding of individuals with DD are some important barriers to care (Lunsky, Gracey, & Gelfand, 2008; Sowney & Barr, 2007).

To address the need for evidence-based practice change, a Toronto ED implemented an initiative that aimed to improve awareness and identification of patients with DD; modify care approach during ED assessments; and enhance communication and transition to the community at discharge (for more details on this initiative, see Lunsky et al., 2014). This initiative was called "DD CARES." The present study used feedback obtained via survey from ED staff to assess implementation of this initiative to improve emergency care for patients with DD along the following dimensions: (1) staff awareness of the initiative; (2) staff access to organizational support; (3) burden of the initiative on staff time; (4) fit of the initiative with current practices; and (5) what impact the initiative had on care provided to patients with DD. We opted to survey ED staff members because it enabled broad reach across different disciplines, allowed responses to remain anonymous, and could be completed relatively quickly after seeing a patient with DD when the interaction was fresh in their memory.

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## Methods

### Participants

Staff members who were involved in the care of an ED user with DD participated in this study. This included physicians (MD) and registered nurses (RN) directly involved in the care of individual patients, triage nurses who attend to individuals upon arrival to the ED, clinical care leaders (CCL; nurses who are responsible for patient flow), as well as patient administrative associates (PAA) who prepare medical charts, and social workers who become involved in patient care if extra support services are required. In total, surveys were completed by 13 physicians, 25 nurses (triage, CCL, RN), and five other ED staff (PAA and social workers).

### Procedure

Following involvement in the care of a patient with DD, ED staff members were approached by a researcher and asked to complete a brief, follow-up survey. This survey included eight items which asked about fit and burden of the initiative, access to ongoing training and support opportunities, and how staff felt the process impacted patient care. The items in the survey were modified from the expanded evidence-based practice scale by Aarons, Cafri, Lugo, Sawitzky, 2012. The survey included three discipline-specific items about use of DD-specific tools (discipline-specific Tip Sheets, a DD resource binder kept in the ED for staff reference, and electronic discharge letters (Dear Doctor/Dear Patient) to be completed by physicians. For detail on these tools, see the Emergency Care Toolkit at [www.hcardd.ca](http://www.hcardd.ca). This study received approval from the hospital research ethics board.

### Analysis

Survey responses were analyzed using descriptive statistics, and responses to an open-ended question about impact on patient care were qualitatively analyzed for emerging themes.

## Results

All staff who completed follow-up surveys were aware of the initiative and its purpose. Responses from staff regarding organizational support, and burden and fit of the process, are presented in Table 1. Most staff reported that training and

ongoing support were provided. Additionally, most of staff were receptive to learning new procedures, and most responded that the process fit with their administrative work. Very few staff reported that the process did not fit with their clinical approach, and all staff agreed that patients benefited from the process.

Two themes, improved staff approach and improved patient care, emerged from responses to "How did the DD CARES initiative impact patient care?" Staff approach to caring for patients with DD was improved because staff obtained supplementary information from caregivers, spent more time with the patient, had better awareness of the patient's disability, identified additional needs, improved communication with the patient, and had a better understanding of common presenting issues in this population. The initiative also impacted patient care since patients felt more comfortable and important, they received more personalized care, extra support was available, and patient communication of issues was improved.

Staff responses regarding the use of discipline specific tools are presented in Table 2. The majority of CCLs and PAAs (89%) reported printing Tip Sheets for RNs and physicians, and attached them to the patient's chart. However, fewer than 30% of RNs or physicians reported seeing a Tip Sheet. Just over half of physicians and nurses (53%) were aware of the DD Resource Binder and knew where it was located in the ED. Only one-quarter of physicians (25%) completed an electronic, autofill discharge letter designed to help patients understand what happened during their ED visit, as well as communicate information to their primary care physician.

## Discussion

The high level of awareness of the initiative among surveyed ED staff indicated communication about the practice-change initiative was effective. Findings also suggested that staff had the time and opportunity to learn about and implement the process. Staff did not view the initiative as a burden and felt it fit with their current role. The process appeared to serve as a reminder to staff to adjust their clinical approach when providing care for persons with DD, and as a result, staff noted better communication with these patients. Critically, staff felt the process improved emergency care for patients with DD. However, areas of improvement at the time

*Table 1. ED Staff Assessment of Organizational Support, Burden and Fit of the ED Practice-Change Initiative (N = 43)*

Degree of agreement with statement	Not at all	Frequency of Responses (%)			
		To a slight extent	To a moderate extent	To a great extent	To a very great extent
<b>Organizational Support</b>					
Training was provided to learn about DD CARES	14.6%	4.9%	22.0%	17.1%	41.5%
Ongoing support was provided to learn about/use DD CARES	4.9%	12.2%	14.6%	34.2%	34.2%
<b>Burden</b>					
I don't have time to learn anything new	74.4%	9.3%	9.3%	7.0%	0.0%
I don't know how to fit DD CARES into my administrative work	65.1%	18.6%	2.3%	11.6%	2.3%
<b>Fit</b>					
DD CARES fits with my clinical approach	2.7%	8.1%	13.5%	29.7%	46.0%
Patients benefit from DD CARES	0.0%	0.0%	8.1%	29.7%	62.2%

*Table 2. ED Staff Awareness and use of Discipline-Specific ED Tools*

<i>Survey Question</i>	<i>Respondents</i>	<i>Yes (%)</i>
"Did you print the MD and RN Tip Sheets and attach to the patient's chart?"	Clinical care leaders (CCL) & patient administrative associates (PAA)	88.9%
"Did you receive a Tip Sheet?"	Registered nurses (RN) & physicians (MD)	28.6%
"Are you aware of the resource binder?"	Physicians (MD) & nurses (triage, CCL, RN)	52.9%
"Did you complete a Dear Doctor/ Dear Patient letter at discharge?"	Physicians (MD)	25.0%

of evaluation were identified as clinical tools were not being used by all clinicians.

Feedback from staff surveys has allowed the ED team to evaluate its efforts thus far and identify areas for improvement. Process gaps, such as low uptake of DD-specific tools printed and attached to the chart and limited distribution of patient information at discharge, indicated that continued refinements to the process could help ensure information is effect-

ively relayed among ED staff. It is possible that some tools such as the "Dear Doctor" letter are perceived as taking too much extra time, but it is also quite likely that tools, although useful, are simply not remembered from visit to visit because the patient group is seen infrequently.

Limitations of the study include limited detail from a brief survey, not all staff provided feedback, and the results cannot speak to the maintenance of the initiative. Since the surveys were

completed, a more extensive evaluation of this initiative, along with two related initiatives at other hospitals was carried out. The more recent evaluation combined survey data with individual interviews and focus group information (see Selick et al., 2018; [www.hcardd.ca](http://www.hcardd.ca) for more information). Importantly, staff feedback was specific to implementation at one hospital and may not apply elsewhere. Other sites may also not have the same electronic capacity of this hospital, or the same staffing combination. Differences between hospitals can impact which tools are selected within the initiative, as well as its outcomes.

Efforts at this ED are continuing and lessons learned from implementation of the initiative are being applied. For example, e-mail blasts continue to remind staff of DD-specific tools in the ED, and some tools and process steps have been adapted to better fit with existing ED procedures. An implementation toolkit has been developed for emergency care providers at other hospitals interested in improving care for this population (visit [www.hcardd.ca](http://www.hcardd.ca) to download the Emergency Care Toolkit).

## Key Messages From This Article

**People with disabilities.** This is a project to help make care better for people with disabilities when they come to the emergency department. We found that giving doctors and nurses information about people with disabilities helped do this.

**Professionals.** The goal of this project is to improve care for people with developmental disabilities when they come to the emergency department. Hospital staff can use tools in the hospital to help them provide better care.

**Policymakers.** If hospitals are more prepared to support their patients with developmental disabilities, they can make the experience at the emergency department better. Staff are open to improving the care they provide and should be encouraged to do so.

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## Winners of the Brown & Percy *Journal on Developmental Disabilities* 2017 Student Publication Prizes

### About the Student Publication Prize

Since its inception in 1992, the *Journal on Developmental Disabilities* (JODD), published by the Ontario Association on Developmental Disabilities (OADD), has consistently encouraged students to contribute papers to the journal that are based on their individual and collaborative research. A prize initiated by Ivan Brown and Maire Percy in 2007 – the Annual Brown and Percy *Journal on Developmental Disabilities* Student Publication Prize – further encourages student contribution to the journal. Funding for this award comes from royalties generated by the textbook *“Developmental Disabilities in Ontario”* that is edited by Ivan and Maire. The prize is awarded to an author of what is judged to be the most outstanding student contribution in a calendar year. A student contribution is deemed to be a paper that has been successfully peer-reviewed and published in the JODD based on research or ideas of someone who is currently a student, or of someone reporting work completed while he or she was a student (e.g., a thesis). Normally, an eligible author for this award would be the first author of the paper, but intermediate authors also are eligible if they have contributed 50% or more to the content and writing. The prize consists of a letter of congratulations signed by Ivan and Maire, a cash award of \$500, and a plaque from the OADD commemorating the event. If there is a tie for first place, the cash award to each winner is \$250 and two plaques are awarded. Sometimes there are runners up for the prize. In this case, runners up receive a congratulatory letter in recognition of their achievement. Awards are presented at the Annual Meeting of Research Special Interest Group (RSIG) of the OADD. Award winners are announced in an upcoming issue of JoDD. This year’s award winners were selected from among papers published in JoDD issues 23(1) and 23(2). Judging was done by a small sub-committee of the RSIG. In this year’s competition, two papers tied for first place and two papers tied for second place. The awards were presented at the 2018 RSIG meeting held at the Ambassador Hotel and Conference Centre, in Kingston, Ontario.

### 2017 Winners

The two papers which tied for first place were:

- **Dustin Heinrichs, Shahin Shooshtari, Marni Brownell, Rosemary Mills, and Brenda Stoesz.** (2018). Health and health care utilization of Manitoba children in care with and without developmental disabilities: A population-based comparative study. *Journal on Developmental Disabilities*, 23(2), 5–16.
- **Kelly D. Coons, Alexandra L. Clement, and Shelley L. Watson.** (2017). Are rural and urban Ontario health care professionals aware of fetal alcohol spectrum disorder? A secondary data analysis of the fetal alcohol syndrome survey for health professionals. *Journal on Developmental Disabilities*, 23(1), 76–91.

The two runner up papers were:

- **Rebecca-Jane McAllister, Kelly Carr, Chad A. Sutherland, Nadia Azar, and Sean Horton.** (2018). Bystander perceptions of an exercise program for adults with autism spectrum disorder and an intellectual disability within a university setting. *Journal on Developmental Disabilities*, 23(2), 50–54.
- **Alshaba S. Billawala, Chloe A. Hamza, and Shannon L. Stewart.** (2018). Risk factors for complex special needs among male children seeking mental health services. *Journal on Developmental Disabilities*, 23(2), 17–26.