

The Cost-Effectiveness of Expanding Intensive Behavioural Intervention to All Autistic Children in Ontario

Rentabilité de l'élargissement des services d'intervention comportementale intensive à tous les enfants autistes de l'Ontario

In the past year, several court cases have been brought against provincial governments to increase funding for Intensive Behavioural Intervention (IBI). This economic evaluation examines the costs and consequences of expanding an IBI program.



by SANOBER S. MOTIWALA, MSC

*Department of Health Policy, Management and Evaluation
University of Toronto, ON*

SHAMALI GUPTA, MSC

*Department of Health Policy, Management and Evaluation
University of Toronto, ON*

MEREDITH B. LILLY, HON BA

*Department of Health Policy, Management and Evaluation
Fellow, CIHR Program in Healthcare, Technology and Place
University of Toronto, ON*

WENDY J. UNGAR, PHD

Scientist, Population Health Sciences, Hospital for Sick Children

Adjunct Scientist, Institute for Clinical Evaluative Sciences

Assistant Professor, Department of Health Policy, Management and Evaluation

University of Toronto, ON

PETER C. COYTE, PHD

Health Services Chair

Canadian Health Services Research Foundation and Canadian Institutes of Health Research

Professor, Health Economics, Department of Health Policy, Management and Evaluation

University of Toronto, ON

Abstract

Intensive Behavioural Intervention (IBI) describes behavioural therapies provided to autistic children to overcome intellectual and functional disabilities. The high cost of IBI has caused concern regarding access, and recently, several court cases have been brought against provincial governments to increase funding for this intervention. This economic evaluation assessed the costs and consequences of expanding an IBI program from current coverage for one-third of children to all autistic children aged two to five in Ontario, Canada. Data on the hours and costs of IBI, and costs of educational and respite services, were obtained from the government. Data on program efficacy were obtained from the literature. These data were modelled to determine the incremental cost savings and gains in dependency-free life years. Total savings from expansion of the current program were \$45,133,011 in 2003 Canadian dollars. Under our model parameters, expansion of IBI to all eligible children represents a cost-saving policy whereby total costs of care for autistic individuals are lower and gains in dependency-free life years are higher. Sensitivity analyses carried out to address uncertainty and lack of good evidence for IBI efficacy and appropriate discount rates yielded mixed results: expansion was not cost saving with discount rates of 5% or higher and with lower IBI efficacy beyond a certain threshold. Further research on the efficacy of IBI is recommended.

Résumé

L'intervention comportementale intensive (ICI) décrit les thérapies comportementales fournies aux enfants autistes pour les aider à surmonter leurs déficiences intellectuelles et fonctionnelles. Les coûts élevés de cette intervention ont soulevé des préoccupations quant à l'accès et, récemment, plusieurs poursuites judiciaires ont été intentées contre les gouvernements provinciaux en vue d'amener ces derniers à augmenter le finance-

ment accordé à l'ICI. Cette analyse économique visait à évaluer les coûts et les conséquences de l'élargissement de la portée d'un programme d'ICI pour le rendre accessible à tous les enfants autistes âgés de deux à cinq ans en Ontario, au Canada – au lieu du tiers des enfants comme c'est le cas actuellement. Les données sur les heures et les coûts liés à l'ICI, ainsi que sur les coûts des services éducatifs et de relève, ont été obtenues auprès du gouvernement. Les données sur l'efficacité du programme ont été tirées de la littérature. Ces données ont été modélées afin de déterminer les économies supplémentaires et les années de vie autonome gagnées. L'élargissement de la portée du programme actuel a permis de réaliser des économies de 45 133 011 \$ CAN en 2003. Selon les paramètres de notre modèle, étendre l'ICI à tous les enfants admissibles constitue une mesure de réduction des dépenses en vertu de laquelle les coûts totaux des soins fournis aux enfants autistes sont moins élevés et les gains d'autonomie sont plus élevés. Les analyses de sensibilité effectuées pour aborder l'incertitude et le manque de données solides corroborant l'efficacité de l'ICI et les rabais appropriés pour cette dernière ont donné des résultats mixtes : l'élargissement de la portée de l'ICI ne permet pas de réaliser des économies avec des rabais de 5 p. cent ou plus ou avec un seuil d'efficacité en deçà d'un certain niveau. Nous recommandons d'effectuer des travaux de recherche plus poussés sur l'ICI.

AUTISM IS AN EARLY-ONSET DEVELOPMENTAL DISABILITY CHARACTERIZED by impairments in social interaction, abnormal verbal and non-verbal communication, repetitive, stereotyped behaviour and resistance to change (Howlin 1998; American Psychiatric Association 1994). Most cases are diagnosed by three years of age, with a male–female ratio of 3:1 (Ontario Ministry of Community, Family and Children's Services [MCFCS] 2000). The reported prevalence of autism in Ontario almost doubled between 1996 and 1998, with the 1998 prevalence being 2.09 per 1,000 children aged five and younger (Ontario Health Insurance Program [OHIP] 2000). The etiological cause of autism is believed to be dysfunction of the right hemisphere of the brain, which is responsible for appropriate visual–spatial and emotional interactions (Gillberg and Coleman 2000).

Intensive Behavioural Intervention (IBI) is the general term for behavioural therapies provided to autistic children to overcome their intellectual and functional disabilities. Several variants of IBI and non-IBI therapies have been reported, but strong evidence is lacking regarding the effectiveness of many of these approaches. No single form of behavioural intervention is appropriate for all individuals with autism (Dawson and Osterling 1997). IBI typically involves one-on-one training provided by a therapist, in which children are trained to respond to environmental changes, understand and use language and interact appropriately with others in social settings

(Dawson and Osterling 1997). Positive reinforcement is used to internalize appropriate behaviours. Success of IBI is believed to correspond to the intensity and duration of the treatment – between 20 and 40 hours per week of one-on-one therapy, for a minimum of two years, is generally believed to yield optimal results (MCFCS 2000; Lovaas 1987). Beyond a minimum threshold of 20 hours per week, there is little agreement in the peer-reviewed literature as to the exact number of hours required to achieve the most favourable results (MCFCS 2000; Dawson and Osterling 1997; Bassett et al. 2000; Sheinkopf and Sigel 1998; Smith 1999; Ludwig and Harstall 2001). Annual IBI costs range from \$40,000 to \$75,000 per child in 2003 Canadian dollars, depending on the number of treatment hours provided and other factors, including administrative costs and training (Ontario Ministry of Children’s Services [MCS] and Ontario Ministry of Community and Social Services [MCSS] 2003; Jacobson and Mulick 2000; Jacobson et al. 1998; Hildebrand 1999; Freeman 1997).

IBI outcomes are generally categorized by level of functioning, assessed at the end of the intervention period. “Normal-functioning” individuals integrate into the community, receive schooling in mainstream classrooms and live independently as adults. “Semi-dependent” and “very dependent” individuals make partial and minimal gains, respectively, and continue to rely on social assistance throughout their lifetime (Lovaas 1987; Freeman 1997; Rutter 1996; Howlin 1997; Howlin et al. 2004). The most optimistic estimates available in the literature suggest that without receiving any form of intervention, as many as 25% of autistic individuals live normal lives, 25% are moderately disabled and 50% are severely compromised (Freeman 1997). However, other studies have reported lower rates of normalization without intervention (Rutter 1996; Howlin 1997). Success rates of IBI and similar interventions vary.

A highly publicized and controversial study, conducted by Lovaas (1987), reported a large proportion of children (up to 47%) achieving normal intellectual and educational functioning at the end of the intervention. However, Lovaas’s primary study and its follow-up (McEachin et al. 1993) have been criticized for their methodological limitations, particularly, exclusion of the poorest-functioning 15% of referred subjects, the non-random assignment of children to treatment groups and the statistically significant difference in sex ratios between the treatment and control groups. These limitations have led to concerns regarding the validity of Lovaas’s findings (Bassett et al. 2000).

In Canada, funding for IBI varies across provinces, but most provincial governments offer some support for IBI to children diagnosed with autism up to a certain age. As a result of high costs of treatment, several lawsuits have been launched by families of autistic children, rallying for increased government funding for IBI. In most cases, rulings have been favourable for the families, requiring governments to increase funding for IBI. In contrast, the Supreme Court of Canada recently ruled favourably in an appeal from the British Columbia government, denying increased funding for

IBI on the grounds that the therapy did not constitute “medically necessary” care as defined by the *Canada Health Act*.

In Ontario, the government currently funds up to three years of IBI for approximately a third of autistic children younger than six years of age (OHIP 2000; MCSS 2002). The Ontario government does not promote any particular form of IBI. It has contracted with a private organization (Behaviour Institute, Hamilton) that delivers training to regional service providers, who in turn are contracted through a competitive tendering process. In its provincial program guidelines for IBI, the government lists principles and teaching methods that regional providers are expected to follow, which include, where appropriate, one-on-one training, task analysis, positive reinforcement and small-group instruction (MCFCS 2000). Eligibility for IBI, duration and intensity of treatment are determined through formal assessment, with allocation of services geared towards children with more severe forms of autism (MCFCS 2000). Earlier this year, the Superior Court of Ontario ruled in favour of the plaintiffs in a class-action lawsuit against the Ontario government, challenging the termination of public funding for IBI at the age of six. The decision is currently being appealed.

The purpose of this study was to conduct a cost-effectiveness analysis to evaluate the expansion of the IBI program to all autistic children in Ontario from two to five years of age, commencing in 2003. We included costs incurred only by the government and excluded all other costs, for example, those incurred by autistic individuals, their families and employers. The government’s perspective was employed for the analysis because it is highly relevant to ongoing legal and policy debates across the country. The provision of IBI in this model was limited to children aged two to five because (1) IBI is believed by many to be most effective when provided at an early age (MCFCS 2000); (2) currently, the Ontario government funds IBI only for children under the age of six (MCS and MCSS 2003); and (3) previous economic analyses carried out in other jurisdictions have limited IBI provision to children of similar ages (Jacobson et al. 1998; Hildebrand 1999). Thus, the present model would facilitate comparisons.

Methods

Including costs incurred only by the government, we developed a model that reflects the current public provision of autism services in Ontario. The prevalence of autism in Ontario, or the cohort size for this study ($n = 1,309$), was calculated as the sum of the number of children receiving IBI ($n = 485$), the number of children eligible but wait-listed for IBI ($n = 91$) and the number of children waiting for an assessment, multiplied by the proportion of assessed children who have historically been deemed eligible for IBI ($n = 952 \times 0.77$). The three comparison groups were (1) *Status Quo* provision, (2) *Expansion* of IBI services and (3) *No Intervention*. *Status Quo* was based on the current provision of autism services by the provincial government, whereby 37% of

children with autism aged two to five ($n = 485$) receive up to three years of IBI for 23 hours per week on average, while the remainder ($n = 824$) do not receive IBI. While the majority of children currently eligible for IBI in Ontario receive it for less than three years because of diagnostic delays and waiting lists, our study was based on the assumption that all children eligible for these services would receive them for a fixed three-year duration. Under *Expansion*, IBI was provided to all autistic children ($n = 1,309$) for three years at 23 hours per week. Under the third scenario, *No Intervention*, IBI was not provided to any of the 1,309 children in the cohort. Although this scenario represents an unlikely regression from the current situation in Ontario, it makes our findings relevant for jurisdictions where IBI may not be currently publicly funded.

Efficacy rates

Under all three scenarios, children were categorized according to their levels of functioning – normal, semi-dependent and very dependent – upon completion of IBI until the age of 65 (Table 1) (Jacobson et al. 1998; Hildebrand 1999). Efficacy rates for *No Intervention*, the cohort that received no IBI, were based on published literature (Freeman 1997; Howlin et al. 2004; Green et al. 2002). It was assumed that 25% attain normal functioning, 25% are semi-dependent and 50% are very dependent without receiving IBI (Freeman 1997). The figures from Freeman (1997) are the most optimistic reported in the literature; they match closely more recent estimates of adult functioning by Howlin et al. (2004), which are slightly lower. Although many studies report even lower rates of normalization (Rutter 1996; Howlin 1997), we selected the highest published rates to investigate the cost-effectiveness of IBI from a best-case scenario, thereby increasing the robustness of our model.

Because of ongoing controversy regarding the reported efficacy of Lovaas's treatment and other forms of behavioural intervention (Dawson and Osterling 1997; Bassett et al. 2000; Sheinkopf and Sigel 1998; Smith 1999; Ludwig and Harstall 2001; Green et al. 2002), we assigned IBI efficacy rates that were more conservative than those reported for Lovaas's intervention (1987) and its replications (McEachin 1993; Sallows and Graupner 2001). The efficacy rates for *Expansion* were assumed to be 30% normal, 50% semi-dependent and 20% very dependent. *Status Quo* efficacy was based on a weighted average of 824 children receiving no IBI (efficacy equivalent to *No Intervention*) and 485 children receiving IBI (efficacy equivalent to *Expansion*) for three years. The resultant efficacy rates for *Status Quo* were 26.9% normal, 34.3% semi-dependent and 38.9% very dependent.

Cost Data Sources

All costs in the model were converted to 2003 Canadian dollars using growth in the

consumer price index from the period when the underlying data were available, and were estimated for individuals from age two to 65.

The Ontario Ministry of Children's Services and Ontario Ministry of Community and Social Services (2003) reported the annual cost of IBI as \$75,670 per child aged two to five, based on 23 hours per week of therapy. This figure represents the aggregate cost of the IBI program incurred by the Ontario government and includes the training costs of IBI therapists, contractual payments to service providers, and salaries, benefits and overhead costs incurred by provincial civil servants. Average wage rates from Statistics Canada's Ontario Wage Survey (1999) were used to estimate costs for government-funded respite services and speech and language therapy (BBB Autism Support Network 2002). In all cases, costs were converted to 2003 dollars.

No autism-related costs were assumed for normal-functioning individuals after the age of five; families of semi-dependent and very dependent individuals in both the *Status Quo* and *Expansion* groups continued to receive respite services until 18 years of age. All education costs were derived from Ontario Ministry of Education documents (2000; 2001a,b,c). This ministry incurs two levels of special-education costs, Intensive Support Amount 2 (ISA 2) and Intensive Support Amount 3 (ISA 3) for semi- and very dependent individuals from five to 18 years of age.

Adult care costs for semi- and very dependent individuals were based on reports prepared by the Auditor of Ontario (MCSS 2001). Costs for adult day programs were obtained from Ontario Agencies Supporting Individuals with Special Needs (OASIS 2000). Due to limited availability of data on housing and care of autistic adults, 50% of semi-dependent individuals were assumed to live independently and 50% in public residential facilities, while all very dependent individuals were assumed to live in public residential facilities. Autistic adults are eligible for compensation through the Ontario Disability Support Program (ODSP) (Canadian Legal Information Institute 2004). ODSP benefits represent transfer payments rather than costs related directly to autism; therefore, these monthly ODSP entitlements were excluded from the model. The cost to government and other employers of administering assisted-employment programs for developmentally disabled adults was based on current programs of Human Resources Development Canada (HRDC 1999, 2001).

While healthcare utilization might be related to the level of functioning (Jarbrink and Knapp 2001), we did not have access to such data; hence, the cost-effectiveness analysis does not capture these healthcare costs. However, since utilization may increase with the level of dependence, the potential cost savings identified in this study would increase if healthcare utilization were included.

In projecting costs over the productive lifetime, a discount rate of 3.0% per annum was applied to calculate present values (Drummond et al. 1997). In sensitivity analyses, discount rates from 1.0% to 5.0% were used.

TABLE 1. Levels of functioning, efficacy rates, and dependency-free years gained for *No Intervention*, *Status Quo* and *Expansion*

LEVEL OF FUNCTIONING	DESCRIPTION	EFFICACY RATES		
		No Intervention	Status Quo*	Expansion
Normal	Mainstream classroom education; independent functioning; earn average Canadian high school graduate income as adults	25%	26.9%	30%
Semi-Dependent	Special education; respite services; 50% live independently as adults; 50% live in residential facilities; participate in day programs; earn assisted employment income as adults	25%	34.3%	50%
Very Dependent	Intensive special education; respite services; 100% live in residential facilities as adults; participate in day programs; earn assisted employment income as adults	50%	38.9%	20%
Discounted Dependency-free years gained until 65 years of age†		9.6 years	11.2 years	14.0 years

*Weighted average based on 485 children receiving IBI (efficacy: 30% normal, 50% semi-dependent, 20% very dependent) and 824 children receiving no IBI (efficacy: 25% normal, 25% semi-dependent, 50% very dependent)

†Calculated as a weighted average based on efficacy rates for each scenario, discounted at 3% per annum

Outcomes

IBI outcomes were measured by the number of dependency-free years gained to age 65, where dependency was defined as the need for special education and other special services comprising adult day programs, disability supports and assisted employment. Normal-functioning individuals were not dependent after age five and, as a result, gained 60 dependency-free years. Very dependent individuals made minimal gains

from IBI, remained dependent throughout life and gained zero dependency-free years. Semi-dependent individuals continued to be partially dependent. Their outcome was assumed to be the midpoint between normal and very dependent functioning outcomes; they gained 30 dependency-free years. Estimated dependency-free years for the study time horizon were discounted at 3.0% per annum. The discounted number of dependency-free years gained under *No Intervention*, *Status Quo* and *Expansion* were calculated as the weighted average of dependency-free years for normal, semi- and very dependent individuals under each scenario (Table 1). The number of discounted dependency-free years per person to age 65 was 9.6 years for *No Intervention*, 11.2 years for *Status Quo* and 14.0 years for *Expansion*.

Results of the analysis were expressed in terms of incremental cost savings in present values (PVs) and gains in dependency-free years (also measured in PVs). The incremental cost analyses compared *Status Quo* to *No Intervention*, *Expansion* to *No Intervention* and *Expansion* to *Status Quo*.

Productivity costs incurred by semi- and very dependent individuals were included in a sensitivity analysis to examine costs and benefits from a partial societal perspective. Lost wages to age 65 were derived from sex-adjusted income estimates from the 1996 and 2001 Canadian censuses (Statistics Canada 1996; 2001a,b,c) and federal assisted-employment initiatives data (HRDC 1999, 2001). Potential earnings for the normal-functioning group were assumed to be equivalent to the sex-adjusted annual income of high school graduates. Semi-dependent incomes are derived from the average earnings of workers in a supported employment initiative in Newfoundland, adjusted for Ontario (HRDC 2001). Owing to lack of data, income for very dependent individuals was assumed to be 60% of the semi-dependent income. All earnings were converted to 2003 dollars. Sensitivity analyses performed also varied IBI efficacy rates and discount rates to compensate for potential estimation uncertainties and methodological controversies (Drummond et al. 1997). Additional sensitivity analyses varied the cost of IBI, adult care costs and number of dependency-years, but did not significantly affect the results presented.

Results

The annual cost during the intervention period (age two to five) for each autistic child was \$5,378 for *No Intervention*, \$33,414 for *Status Quo* and \$81,048 for *Expansion* (Table 2). The annual cost during schooling (age five to 18) was \$6,616 for normal, \$21,422 for semi-dependent and \$38,672 for very dependent individuals. No costs were incurred during adulthood for normal-functioning individuals. The annual cost during adulthood (age 18 to 65) was \$37,380 for semi-dependent adults and \$75,648 for very dependent adults. The average total discounted cost per individual, based on a weighted average of normal, semi-dependent and very dependent costs

over the study time horizon, was \$1,014,315 for *No Intervention*, \$995,074 for *Status Quo* and \$960,595 for *Expansion*. The cost of *Status Quo* was lower than the cost of *No Intervention*, indicating that the present provision of IBI was preferable to providing no IBI at all. While significant costs were incurred under all scenarios, the cost of *Expansion* was lowest, resulting in savings of \$34,479 per individual over his or her lifetime compared to *Status Quo*. Expansion of the current program to fund IBI for all autistic children (n = 1,309) in Ontario younger than six years of age results in net cost savings of \$45,133,011 for the government. The greatest number of dependency-free life years was gained under *Expansion*: 4.5 years per person compared to *No Intervention* and 2.8 years per person compared to *Status Quo*. *Expansion* is the dominant strategy, as it yields both a decrease in cost as well as gains in dependency-free years.

TABLE 2. Average costs per person of *No Intervention*, *Status Quo* and *Expansion* and cost savings from pair-wise comparisons

AGE RANGE	COST ITEM	NORMAL		SEMI-DEPENDENT		VERY DEPENDENT	
		ANNUAL COST (\$)	PV OF TOTAL COST (2003 \$)	ANNUAL COST (\$)	PV OF TOTAL COST (2003 \$)	ANNUAL COST (\$)	PV OF TOTAL COST (2003 \$)
Intervention Age (2-5)	IBI and other costs: <i>No Intervention</i>	5,378	15,211	5,378	15,211	5,378	15,211
	OR IBI and other costs: <i>Status quo</i> *	33,414	94,516	33,414	94,516	33,414	94,516
	OR IBI and other costs: <i>Expansion</i>	81,048	229,252	81,048	229,252	81,048	229,252
Schooling Age (5-18)	Education and Respite Services	6,616	64,393	21,422	208,490	38,672	376,372

continued

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Adulthood (18-65)	Day programs, residential costs, and assisted employment program costs	0	0	37,380	588,568	75,648	1,191,110
Total cost	No Intervention		\$ 79,604		\$ 812,269		\$ 1,582,693
	Status quo		\$ 158,909		\$ 891,574		\$ 1,661,998
	Expansion		\$ 293,645		\$ 1,026,310		\$ 1,796,734
Average cost per individual (PV): No Intervention †			\$ 1,014,315				
Average cost per individual (PV): Status quo ‡			\$ 995,074				
Average cost per individual (PV): Expansion **			\$ 960,595				
Incremental cost savings per individual: No Intervention → Status quo			\$ 19,241				
Incremental cost savings per individual: No Intervention → Expansion			\$ 53,720				
Incremental cost savings per individual: Status quo → Expansion			\$ 34,479				
Cost savings for cohort (n = 1,309): No Intervention → Status quo			\$ 25,186,469				
Cost savings for cohort (n = 1,309): No Intervention → Expansion			\$ 70,319,480				
Cost savings for cohort (n = 1,309): Status quo → Expansion			\$ 45,133,011				
<small>* Based on 485 individuals out of 1,309 receiving IBI and all 1,309 receiving respite services and speech and language therapy † Based on a weighted average: 25% normal, 25% semi-dependent, 50% very dependent ‡ Based on a weighted average: 26.9% normal, 34.3% semi-dependent, 38.9% very dependent ** Based on a weighted average: 30% normal, 50% semi-dependent, 20% very dependent</small>							

Sensitivity analyses

The cost-effectiveness model was run with productivity costs to examine the economic impact of IBI from a partial societal perspective. Inclusion of productivity costs incurred by semi- and very dependent adults resulted in increased cost savings from *Expansion* of \$54,757 per person and \$71,676,776 for the entire cohort compared to *Status Quo*.

TABLE 3. Results of sensitivity analysis varying the efficacy of IBI

		DECREASED EFFICACY	BASELINE CASE	INCREASED EFFICACY	
Efficacy rates	Expansion	Normal	25.0%	30.0%	40.0%
		Semi-dependent	50.0%	50.0%	50.0%
		Very dependent	25.0%	20.0%	10.0%
	Status quo	Normal	25.0%	26.9%	30.6%
		Semi-dependent	34.3%	34.3%	34.3%
		Very dependent	40.7%	38.9%	35.2%
Results	Incremental savings (cost) per individual: Status quo → Expansion		\$ (13,493)	\$ 84,031	\$ 128,433
	Discounted dependency-free years gained: Status quo → Expansion		2.0 years	2.8 years	4.4 years

IBI efficacy was modified to accommodate controversy in the research literature (Table 3). When the efficacy of IBI was increased to 40% of subjects who achieve normal functioning, 50% achieving semi-dependent functioning and 10% achieving very dependent functioning, the cost savings from *Expansion* compared with *Status Quo* increased to \$128,433 per person. In contrast, under the assumption that IBI yielded efficacy rates of 25% for normal functioning, 50% for semi-dependent functioning and 25% for very dependent functioning, *Expansion* cost \$13,493 more per person compared to *Status Quo* and yielded gains of 2.0 dependency-free years per person. Results of the sensitivity analyses suggest that a significant drop in treatment efficacy from the base case scenario would be required in order to yield a net cost for achieving dependency-free years in this population.

Varying the discount rate modified the present value of the cost savings. With a discount rate of 1%, cost savings from *Expansion* were even greater than those realized in the base case. Cost savings were not realized with a discount rate of 5%: it cost \$29,912 more per person to expand from *Status Quo* to *Expansion*, but gains of 1.8 dependency-free years per person were still realized under *Expansion*.

Discussion

The results demonstrate that expansion of the IBI program, which currently serves 485 children (*Status Quo*), to all 1,309 autistic children in Ontario (*Expansion*) would

yield savings of \$45,133,011 over the entire cohort's lifetime (from two to 65 years of age). Significant costs are incurred under both *Status Quo* and *Expansion*; however, under *Expansion*, the government would spend \$45 million less on autistic individuals when compared with *Status Quo*.

The cost of expanding IBI to all autistic individuals is small (less than 10% of total costs) compared to the significant cost of educating and supporting semi- and very dependent individuals over their lifetime. The present value of total costs incurred during intervention (ages two to five), including respite services and speech and language therapy, is higher for *Expansion* (\$229,252 per person) compared with *Status Quo* (\$94,516 per person). However, the larger intervention cost under *Expansion* yields lower support costs during schooling and adulthood (ages five to 65) compared to *Status Quo*. The primary reason for cost savings from expansion of IBI, from *No Intervention* to *Status Quo* and from *Status Quo* to *Expansion*, is the change in the distribution of functional dependence. Increased provision of IBI results in a shift of individuals from the very dependent to semi-dependent category and, to a lesser extent, from the semi-dependent to the normal-functioning group.

To guard against criticisms of previous economic evaluations (Marcus et al. 2000), IBI efficacy rates in this study were deliberately conservative. The proportion of children who attain normal functioning from IBI was set lower, and the proportion of children who function normally without IBI was set higher, than the proportions cited in the literature (Jacobson et al. 1998; Hildebrand 1999). As a result, cost savings realized under this model (\$34,479 per individual for *Expansion* vs. *Status Quo* and \$53,720 per individual for *Expansion* vs. *No Intervention*) are lower than those reported by previous studies (Jacobson et al. 1998; Hildebrand 1999). Lower normalization rates under *No Intervention* and higher normalization rates from IBI would yield more favourable results for expansion of the current IBI program in Ontario.

Although the costing data utilized in this study are specific to Ontario, our findings may be generalized to inform health policy decisions in other jurisdictions. The increased awareness of intensive behavioural intervention and its high program cost have made the financing of IBI and its cost-effectiveness relevant concerns for governments and other payer organizations. The grounding of our model parameters in peer-reviewed research evidence and the scope of the sensitivity analyses make our findings relevant for policy decision-makers.

Limitations

Several study limitations should be noted. First, only costs borne by the Ontario government were included in this economic evaluation; hence, costs borne by other payers, including autistic individuals, their families and employers, were not considered. Inclusion of such cost items as opportunity costs, quality of life of families and unpaid

caregiver expenses could potentially increase the savings realized under *Expansion* (Curran et al. 2001; Jarbrink and Knapp 2001; Jarbrink et al. 2003). Second, expansion of the IBI program may result in higher average costs per child in the short term due to shortage of qualified IBI therapists in the province and the resulting increase in their earnings. Third, this model assumed that all children initiated IBI at the age of two. However, children may be diagnosed with autism at later ages. Because of age restrictions currently enforced by the Ontario government, these children may not receive IBI for the full three-year period. This contingency may affect the efficacy of the treatment and the associated IBI costs incurred. Fourth, the 485 children currently receiving government-funded IBI in Ontario were assumed to be representative of the entire cohort of autistic children. Fifth, while healthcare utilization might be related to the level of functioning, we did not have access to such data and, hence, the cost-effectiveness analysis does not include these costs. However, since utilization may increase with the level of dependence, the cost savings identified in this study would increase if healthcare utilization were included. Sixth, the provincial government provided only aggregate costs for its entire IBI program, resulting in the very high annual IBI therapy cost of \$75,670 per child. This figure includes the operating costs associated with the launch of the IBI program in Ontario, including a large training component for new IBI therapists. As a result, costs per child are expected to decrease in coming years as start-up costs diminish. Finally, every attempt was made to obtain accurate costing information. However, in the absence of reliable estimates, costs from other jurisdictions within Canada, and costs for developmentally disabled people in general, were used to represent costs incurred for autistic individuals in Ontario.

Conclusion

This economic evaluation demonstrates positive outcomes from expansion of the current IBI program offered by the Ontario government. In the absence of high-quality evidence on the efficacy of IBI, but under reasonable assumptions, estimated cost savings in present-value terms associated with this expansion were \$45 million for the government, with potential improvement in the quality of life of autistic individuals and their families because of increased dependency-free years gained under *Expansion*. These cost savings and improvements in outcomes were largely maintained in the sensitivity analyses. However, savings to government disappeared when the annual discount rate of 5% was used or when IBI was assumed to be less effective than in the base case scenario, with *Expansion* resulting in 25%, 50% and 25% of individuals in normal, semi-dependent and very dependent categories (compared to 30%, 50% and 20% in the base case), respectively. Owing to uncertainty surrounding the efficacy of IBI, further study in the area is recommended, perhaps in the form of a randomized, controlled trial, to allow more definitive economic evaluations in the future.

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Correspondence may be directed to: Dr. Peter C. Coyte, Department of Health Policy, Management and Evaluation, Faculty of Medicine, University of Toronto, 155 College Street, Suite 425, Toronto, ON M5T 3M6; tel.: 416-978-8369; fax 416-978-7350; email: peter.coyte@utoronto.ca.

REFERENCES

- American Psychiatric Association. 1994. *Diagnostic and Statistical Manual of Mental Disorders* (4th ed.). Washington, DC: Author.
- Bassett, K., C. Green and A. Kazanjian. 2000. *Autism and Lovaas Treatment: A Systematic Review of Effectiveness Evidence*. Vancouver: BC Office of Health Technology Assessment, University of British Columbia.
- BBB Autism Support Network. 1999. "Sample Requests for Services. Special Services at Home Program." Retrieved December 7, 2005. <http://www.bbbautism.com/ssah_acsd_guidelines.htm>.
- Canadian Legal Information Institute. 1997. *Ontario Disability Support Program Act, 1997*. Retrieved December 7, 2005. <<http://www.canlii.org/on/laws/sta/1997c.25sch.b/index.html>>.
- Curran, A.L., P.M. Sharples, C. White and M. Knapp. 2001. "Time Costs of Caring for Children with Severe Disabilities Compared with Caring for Children without Disabilities." *Developmental Medicine and Child Neurology* 43: 529–533.
- Dawson, G. and J. Osterling. 1997. "Early Intervention in Autism." In M.J. Guralnick (ed.), *The Effectiveness of Early Intervention* (pp. 307–326). Baltimore: Paul H. Brooks.
- Drummond, M.F., B. O'Brien, G.L. Stoddart and G.W. Torrance. 1997. *Methods for the Economic Evaluation of Health Care Programmes* (2nd ed.). Oxford: Oxford University Press.
- Freeman, B.J. 1997. "Guidelines for Evaluating Intervention Programs for Children with Autism." *Journal of Autism and Developmental Disorders* 27: 644–645.
- Gillberg, C. and M. Coleman. 2000. *The Biology of the Autistic Syndromes* (3rd ed.). London: MacKeith Press.
- Green, C., K. Bassett and A. Kazanjian. 2002. *Critical Appraisal of Submitted Cost–Benefit Models for Lovaas's Early Intensive Behavioural Intervention for Autism*. Victoria: BC Office of Health Technology Assessment, Centre for Health Services and Policy Research.

- Hildebrand, D. 1999. *Cost–Benefit Analysis of “Lovaas” Treatment for Autism and Autism Spectrum Disorder (ASD)*. Preliminary report prepared for Harper Grey Easton, Barristers and Solicitors. Vancouver: Columbia Pacific Consulting.
- Howlin, P. 1997. “Prognosis in Autism: Do Specialist Treatments Affect Long-Term Outcomes?” *European Children and Adolescents Psychiatry* 6: 56.
- Howlin, P. 1998. *Children with Autism and Asperger Syndrome – A Guide for Practitioners and Carers*. Chichester, UK: John Wiley & Sons.
- Howlin, P., S. Goode, J. Hutton and M. Rutter. 2004. “Adult Outcomes for Children with Autism.” *Journal of Child Psychology and Psychiatry* 45: 212–229.
- Human Resources Development Canada (HRDC). 1999. *Lessons Learned from Evaluations of Disability Policy and Programs*. Retrieved December 7, 2005. <http://www11.hrdc-drhc.gc.ca/pls/edd/DPPTR_81000.htm>.
- Human Resources Development Canada (HRDC). 2001. *Supported Employment Initiative – Summative Evaluation Final Report*. Retrieved December 7, 2005. <<http://www.releases.gov.nl.ca/releases/2002/hre/0509n15Report.pdf>>.
- Jacobson, W.J. and A.J. Mulick. 2000. “System and Cost Research Issues in Treatments for People with Autistic Disorders.” *Journal of Autism and Developmental Disorders* 30: 585–593.
- Jacobson, W.J., A.J. Mulick and G. Green. 1998. “Cost–Benefit Estimates for Early Intensive Behavioral Intervention for Young Children with Autism – General Model and Single State Case.” *Behavioral Interventions* 13: 201–226.
- Jarbrink, K., E. Fombonne and M. Knapp. 2003. “Measuring the Parental, Service and Cost Impacts of Children with Autistic Spectrum Disorder: A Pilot Study.” *Journal of Autism and Developmental Disorders* 33(4): 395–402.
- Jarbrink, K. and M. Knapp. 2001. “The Economic Impact of Autism in Britain.” *Autism* 5(1): 7–22.
- Lovaas, O.I. 1987. “Behavioral Treatment and Normal Educational and Intellectual Functioning in Young Autistic Children.” *Journal of Consulting Clinical Psychology* 55: 3–9.
- Ludwig, S. and C. Harstall. 2001. *Intensive Intervention Programs for Children with Autism. Health Technology Assessment Report*. Edmonton: Alberta Heritage Foundation for Medical Research.
- Marcus, L.M., J.S. Rubin and M.A. Rubin. 2000. “Benefit–Cost Analysis and Autism Services: A Response to Jacobson and Mulick.” *Journal of Autism and Developmental Disorders* 30(6): 595–598.
- McEachin, J.J., T. Smith and O.I. Lovaas. 1993. “Long-Term Outcome for Children with Autism Who Received Early Intensive Behavioural Treatment.” *American Journal of Mental Retardation* 97: 359–372.
- Ontario Agencies Supporting Individuals with Special Needs (OASIS). 2000. “How Agencies Use Their Resources for Administration and Direct Support.” Retrieved December 7, 2005. <<http://www.oasonline.ca/pages/pdf/admincoststudy.pdf>>.
- Ontario Health Insurance Program (OHIP). 2000. *Review of OHIP Databases with Respect to Autism: Ministry of Health Report, Assessment of Autistic Children*. Toronto: Ontario Ministry of Health.
- Ontario Ministry of Children’s Services (MCS) and Ontario Ministry of Community and Social Services (MCSS). 2003 (December). *Intensive Early Intervention Program (IEIP) Source Document*. Presented by the Ontario Government in the case of *Wynberg v. Ontario*.

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- Ontario Ministry of Community, Family and Children's Services (MCFCS). 2000. *Integrated Services for Children Division. Program Guidelines for Regional Intensive Early Intervention Programs for Children with Autism*. Toronto: Author.
- Ontario Ministry of Community and Social Services (MCSS). 2001. *Community Accommodation Program. Provincial Auditor's Report, Follow-up to VFM Section 3.04, 1999 Annual Report*. Retrieved December 7, 2005. <<http://www.auditor.on.ca:81/isysquery/irl1088/3/doc>>.
- Ontario Ministry of Community and Social Services (MCSS). 2002. "Intensive Early Intervention Services for Children with Autism – Questions and Answers." Letter dated January 25, 2002.
- Ontario Ministry of Education. 2000 (March). "Fact Sheet: Special Education Grant." Retrieved December 7, 2005. <<http://www.edu.gov.on.ca/eng/document/nr/00.03/fs2.html>>.
- Ontario Ministry of Education. 2001a. "Student-Focused Funding: School Board Funding Projections for the 2001–2002 School Year." Retrieved December 7, 2005. <<http://www.edu.gov.on.ca/eng/funding/fund0102.html>>.
- Ontario Ministry of Education. 2001b. "Intensive Support Amount (ISA) Guidelines for School Boards." Ministry of Education, Student-Focused Funding. Retrieved December 7, 2005. <<http://www.edu.gov.on.ca/eng/funding/t4.html>>.
- Ontario Ministry of Education. 2001c. "Parents Guide 2001–2002 – Student-Focused Funding." Retrieved November 18, 2002. <<http://www.edu.gov.on.ca/eng/funding/guide02.pdf>>.
- Rutter, M. 1996. "Autism Research: Prospects and Priorities." *Journal of Autism and Developmental Disorders* 26: 257–275.
- Alloways, G. and G. Graupner. 2001 (May). "Replicating Lovaas: Results After Three Years." *Proceedings of the 27th Annual Convention of the Association for Behavior Analysis*, New Orleans, LA.
- Sheinkopf, S. and B. Sigel. 1998. "Home-Based Behavioral Treatment of Young Children with Autism." *Journal of Autism and Developmental Disorders* 28: 15–23.
- Smith, T. 1999. "Outcome of Early Intervention for Children with Autism." *Clinical Psychology and Scientific Practice* 6: 33–49.
- Statistics Canada. 1996. "Average Earnings of the Population 15 Years and Over in 1995 by Highest Level of Schooling: Census 1996, Ontario and Newfoundland and Labrador Data." Ottawa. <<http://www.statscan.ca>>.
- Statistics Canada. 1999. "Occupational Summary Table (Ontario): Ontario Wage Survey." Retrieved December 7, 2005. <<http://www.statscan.ca>>.
- Statistics Canada. 2001a. "2000 Canadian Average Earnings by Sex and Work." Ottawa. <<http://www.statscan.ca>>.
- Statistics Canada. 2001b. "2000 Population Data in Ontario: Census 2000." Ottawa. <<http://www.statscan.ca>>.
- Statistics Canada. 2001c. "Labour Force Participation Rates in Canada." Ottawa. <<http://www.statscan.ca>>.