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## Cost Offset Associated With Early Start Denver Model for Children With Autism

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### Abstract

**Objective:** To determine the effect of the Early Start Denver Model (ESDM) for treatment of young children with autism on health care service use and costs.

**Method:** We used data from a randomized trial that tested the efficacy of the ESDM, which is based on developmental and applied behavioral analytic principles and delivered by trained therapists and parents, for 2 years. Parents were interviewed about their children's service use every 6 months from the onset of the intervention to follow-up (age 6 years). The sample for this

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Disclosure: Dr. Cidav conceptualized the study and its design, conducted the analyses, and drafted the initial manuscript. Dr. Munson assisted with the analyses, especially the coding of different service types, and assisted with interpretation of data. Dr. Estes oversaw original data collection for the follow-up study, and assisted with acquisition and interpretation of data. Dr. Dawson designed and oversaw the original randomized controlled trial and its data collection, and assisted with the acquisition of data and interpretation of data. Dr. Rogers provided training and consultation on the original randomized controlled trial and assisted with interpretation of data. Dr. Mandell helped draft the initial manuscript, and assisted with analysis and interpretation of data. All authors assisted with revising the manuscript for important intellectual content, gave final approval of the version to be published, and agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Drs. Cidav and Munson served as the statistical experts for this research.

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study consisted of 39 children with autism who participated in the original randomized trial at age 18 to 30 months, and were also assessed at age 6 years. Of this sample, 21 children were in the ESDM group, and 18 children were in the community care (COM) group. Reported services were categorized and costed by applying unit hourly costs. Annualized service use and costs during the intervention and post intervention for the two study arms were compared.

**Results:** During the intervention, children who received the ESDM had average annualized total health-related costs that were higher by about \$14,000 than those of children who received community-based treatment. The higher cost of ESDM was partially offset during the intervention period because children in the ESDM group used less applied behavior analysis (ABA)/early intensive behavioral intervention (EIBI) and speech therapy services than children in the comparison group. In the postintervention period, compared with children who had earlier received treatment as usual in community settings, children in the ESDM group used less ABA/EIBI, occupational/physical therapy, and speech therapy services, resulting in significant cost savings in the amount of about \$19,000 per year per child.

**Conclusion:** Costs associated with ESDM treatment were fully offset within a few years after the intervention because of reductions in other service use and associated costs.

**Clinical trial registration information:** Early Characteristics of Autism; <http://clinicaltrials.gov/>; .

## Keywords

autism; cost; use; Early Start Denver Model; early intervention

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Autism spectrum disorder (ASD) is a neuro-developmental condition that emerges in early childhood and often is associated with lifelong disability and substantial economic cost to families, health service delivery systems, and society.<sup>1,2</sup> Early intervention services have been promoted as an important strategy to reduce future disability and associated costs.<sup>3–5</sup> Several reviews and meta-analyses have concluded, based on a large body of research, that early intensive intervention is associated with significant improvements in behavioral outcomes, including improvements in the domains of cognitive, language, social/emotional development, perceptual/fine motor development, and gross motor development.<sup>5–8</sup> Expert consensus and a smaller body of literature suggest that the earlier treatment begins, the better the clinical outcome.<sup>6</sup>

Although early intervention programs vary in their approach, most are characterized by a low child–staff ratio and involvement and coaching of parents and caregivers.<sup>9</sup> Usually, treatment is implemented intensively (i.e., 15–25 hours per week) for 2 years or more.<sup>10–12</sup> Due to treatment intensity and duration, early intervention is expensive, with estimated annual costs of \$40,000 to \$80,000.<sup>13</sup>

Some payers, including health care insurers and state and local early intervention systems, have balked at these costs.<sup>14</sup> In response, a number of studies have attempted to estimate the long-term clinical and economic gains associated with successful early intervention, finding a positive economic return on investment over the course of the individual's life.<sup>13,15,16</sup> These studies are based primarily on simulated data, however, and make many untested

assumptions regarding the extent to which clinical gains observed immediately after treatment will translate into reduced service use and increased community participation in ways that mitigate costs.<sup>17</sup> To our knowledge, only one published study comprises an individual-level, empirical economic evaluation of an ASD intervention, based on a randomized trial from the United Kingdom.<sup>18</sup>

Researchers, interventionists, advocates, and policy makers increasingly seek economic data alongside clinical data to show the value of adopting autism treatments.<sup>18,19</sup> Although evaluations of cost should by no means be the only or even the primary metric by which treatment usefulness is measured, considerations of treatment affordability are important for setting priorities and for ensuring that adequate resources are in place to support standard treatment models. This metric also is useful as new treatments are developed and compared with existing treatments.

The present study estimated the potential cost offset of one such early intervention program for children with ASD, the Early Start Denver Model (ESDM). Similar to many of the most recent generation of manualized, tested early interventions with demonstrated efficacy, ESDM is a comprehensive, naturalistic, developmental behavioral intervention for children with ASD who are 12 to 60 months of age. ESDM uses teaching strategies based on applied behavior analysis (ABA) that are delivered in a naturalistic, relationship-based context.<sup>20</sup> A randomized trial demonstrated that children beginning ESDM by 30 months of age who were offered 20 hours per week for 2 years along with parent coaching had substantial gains in cognitive, language, social abilities, and adaptive behavior relative to children receiving treatment as usual in the community.<sup>21–23</sup> Data concerning service use were collected from parents of trial participants during the trial and for up to 2.5 years after treatment. Although these data were not collected for economic evaluation and therefore do not contain information about the many indirect costs associated with raising a child with autism such as the impact on parental time, they provide a unique resource with which to evaluate the potential cost offset of the intervention—that is, the extent to which the intervention reduces subsequent use of other services. These data allow evaluation of the relative service use and associated costs in two groups: a group that received a manualized early intervention, including parent coaching, which was provided by trained paraprofessionals and supervised by skilled professionals, and a group that received treatment as usual in the community. We analyzed data from the ESDM randomized controlled trial evaluating this intervention versus usual care to determine whether ESDM treatment resulted in reduced health care service use and costs within the study period.

## METHOD

### Setting and Sample

ESDM is a comprehensive, naturalistic developmental behavioral intervention for infants through preschool-aged children with ASD that integrates naturalistic ABA principles with developmental and relationship-based approaches.<sup>24</sup> In this 2-year clinical trial, post-baccalaureate paraprofessionals who were trained to provide therapy and supervised by doctorate-level clinicians delivered the intervention. In addition, parents were offered

weekly or biweekly coaching sessions on the use of naturalistic strategies during daily activities.

The study procedures and participants are described in Dawson *et al.*<sup>21,22</sup> In brief, 48 children between 18 and 30 months of age who were diagnosed with ASD or pervasive developmental disorder—not otherwise specified (PDD-NOS) using gold-standard diagnostic methods were randomly assigned to either ESDM or usual community care (COM) available in the greater Seattle area. The ESDM group received yearly assessments, an average of 15 hours per week of therapist-delivered ESDM intervention and parent coaching. Parents were offered 20 hours per week of intervention but received 15 because of illnesses, vacations, and other logistical constraints of the family's schedule. Parents documented use of naturalistic behavioral strategies at home for 5 or more hours per week, in addition to whatever community services the parents chose. The COM group received yearly assessments, intervention recommendations, and referrals to available community intervention providers.

Participants were recruited through pediatric practices, birth-to-three centers, preschools, hospitals, and state and local autism organizations. Children with a history of issues such as significant vision, hearing, or motor impairment, serious traumatic brain injury, major physical anomalies, genetic disorders associated with ASD (e.g., Fragile X syndrome), seizure disorder, or prenatal drug exposure were excluded. Inclusion criteria included age <30 months at entry, meeting criteria for ASD on the Toddler Autism Diagnostic Interview,<sup>25</sup> meeting criteria for autism or ASD on the Autism Diagnostic Observation Schedule,<sup>26</sup> a clinical diagnosis based on *DSM-IV* criteria using all available information,<sup>27</sup> residing within 30 minutes of the University of Washington, and willingness to participate in a 2-year intervention. This sample was assessed at baseline and at 1 and 2 years after randomization, coinciding with the end of ESDM intervention. Long-term follow-up was conducted when participants were 6 years of age. These time points will be referred to as baseline, 1-year, 2-year, and age-6 assessments, respectively. The present study uses data on the 39 children who participated in the original randomized control trial<sup>20</sup> and also were assessed at age 6 years.<sup>21</sup> Of this sample with longitudinal data, 21 children were in the ESDM group and 18 were in the COM group.

### Service Use and Costs

Parents were interviewed about their children's service use every 6 months from enrollment to final follow-up (age 6 years). At each interview, parents were asked to characterize the child's use of all medical and behavioral health treatments, therapy provided by allied health professionals, and special education and typical/general education services that were used since the last interview. Records include the week that each service was received, average hours of that service in that particular week, and service type.

Services were grouped into the following categories: ESDM, ABA/EIBI (early intensive behavioral intervention), general education (for children <5 years of age, daycare-type typical class; for children >5 years of age, school-based typical class), OT/PT (occupational/physical therapy), social skills training, special education, speech therapy, and other miscellaneous (biomedical therapy, music therapy, parent-focused therapy, nutritional

therapy, counseling/psychological services). We define “total health-related service use” to be the sum of all use of these services except general education. Because general education services also are used by typically developing children and could be considered a desirable outcome, we report their use and cost separately from other services.

Services were then costed by applying hourly unit costs. Service prices in the United States vary greatly both across and within geographic regions<sup>28</sup>; a uniform unit price for each service is not available. A number of sources were used to obtain service unit costs. For ESDM costs, we worked with the intervention developers to identify and categorize costs. We identified personnel needs (paraprofessional, team leader, training professional, clinician, and administrative coordinator) for delivering ESDM treatment to 24 children for 20 hours per week for 2 years and estimated the cost of intervention sessions on the basis of the national average salary of the personnel, and employer costs (employee benefits, 40% of the salary). We assumed that all personnel were employed full time for 2 years (intervention) and 2 months (training), except for the training professional (PhD psychologist) who provided training for the team leaders for 2 months before the intervention. The hourly cost of ESDM was calculated as \$80/h (in 2015 dollars), and included cost of personnel, transportation, overhead (10%), materials, and supplies.

Other unit costs were obtained from published sources,<sup>29–31</sup> national surveys (Medical Expenditure Panel Survey),<sup>32</sup> local service providers, and government departments for school costs. Calculated unit costs for other services are: ABA/EIBI: \$70/h, general education: for age <5 years \$10/h; for age 5 years \$35/h, OT/PT: \$70/h, social skills training: \$85/h, special education: \$30/h, speech therapy: \$85/h, other miscellaneous: \$50/h (an average cost of services that constitute other miscellaneous services). All these unit costs are in 2015 dollars. Since the study spanned several years (2003 through 2011), all costs were adjusted for inflation using consumer price index conversion factors<sup>33</sup> and expressed in 2015 US dollars, and discounted back to the first year of the study using a discount rate of 3%.

Service use and costs were compared within two observation periods. The intervention period included all service use that occurred from the onset of the intervention to the 2-year assessment, including the ESDM use. The postintervention period included all service use that occurred from the 2-year assessment to the age-6 assessment. Our data confined us to adopting a payer perspective (as opposed to a societal perspective), in which we focused on direct health care and education costs.

### Statistical Analysis

We constructed annualized service use and costs within each period (intervention, postintervention) for each child. Annualized use for a specific service type was constructed by summing all hours for that service type that occurred during the study period (intervention, postintervention); dividing this sum by the number of months that the child was observed during this study period; and then multiplying the result by 12. Annualized costs were obtained similarly for each service type.

We compared average annualized service use and costs during the intervention and postintervention periods across study arms. Because both hours and cost data were highly skewed, the statistical significance of differences in mean annual hours and costs were estimated via bootstrapping, which is recommended in the cases of skewed data distributions<sup>34,35</sup> and for small sample sizes.<sup>36</sup> We report mean annualized service use and costs in each period for each study arm, and 95% bias-corrected confidence intervals for the use and cost differences and their *p* values (reported in the text), based on the bootstrap estimation.

## RESULTS

Sample characteristics are reported in previous studies<sup>21,22</sup> and were replicated in this study: The ESDM and COM groups did not differ at baseline in severity of autism symptoms, chronological age, IQ, sex, or adaptive behaviors, nor were there baseline group differences for the subgroup of children who completed the 2-year assessment.<sup>21,22</sup> In addition, 9 children lost to age-6 assessment and 39 children included in the sample of this study did not differ in demographic characteristics of age, sex, race, and ethnicity at baseline (all *p* values >.05). Furthermore, those lost to follow-up did not differ at baseline or the 2-year assessment in IQ, severity of autism symptoms, or adaptive functioning (behaviors, all *p* values >.05; Fisher exact test for categorical variables; Kruskal-Wallis test for continuous variables).

At age-6 assessment, participating children were on average 6 years of age (mean = 72.9 months, SD = 2.6 months), and 72% were of white race/ethnicity. The sample consisted of 9 girls and 30 boys. Mothers were on average highly educated, with only 13% reporting no college, 23% reporting some college, and 64% reporting college completion. The ESDM and COM groups did not differ with regard to maternal education (ESDM: 62% college graduate, 28% some college, 10% no college; COM: 70% college graduate, 18% some college, 12% no college;  $\chi^2 = 0.63$ , *p* > .50). Median annual household incomes between the groups also did not differ (ESDM \$90,000, COM \$85,000; Wilcoxon rank-sum test, *W* = 152.5, *p* > .50). These findings suggest successful randomization; hence, the estimated models do not include sample characteristics as covariates.

Average age was 23 months at intervention initiation, 52 months at the year-2 assessment, and 73 months at the age-6 assessment. The intervention period analysis characterizes use and costs that span ages 2 to 4 years; the postintervention period analysis characterizes use and costs that span ages 4 to 6 years on average. Table 1 reports the analysis results.

### Intervention Period Service Use and Costs

During the intervention period, total health-related average annual service hours were not significantly different between the ESDM and COM groups (913 versus 755, 95% CI for difference = -101,417). The ESDM group had on average 661 hours of ESDM treatment hours per year. The ESDM group used on average 107 hours of ABA/EIBI per year, and this was significantly lower than the COM group's ABA/EIBI use (490 hours per year, 95% CI for difference = -593, -172). The ESDM group used fewer speech therapy (64 versus 121,

95% CI for difference = -113, -1) and general education hours (319 versus 481, 95% CI for difference = -301, -22) per year than the COM group.

During the intervention period, total health-related average annual service costs were not significantly different between the ESDM and COM groups (\$60,369 versus \$45,975, 95% CI for difference = -1525, 30314). Average annual ESDM treatment cost was \$45,580. Average annual ABA/EIBI costs for the ESDM group, from community sources outside of the ESDM treatment, were \$6,306, and these were significantly lower than the COM group's ABA/EIBI costs (\$29,554 per year, 95% CI for difference = -35798, -1069). The ESDM group incurred lower speech therapy costs (\$4,636 versus \$8,758, 95% CI for difference = -8188, -56) and general education costs (\$2,863 versus \$4,271, 95% CI for difference = -2662, -155) per year than the COM group.

### Postintervention Period Service Use and Costs

During the postintervention period, total health-related average annual service hours were less for the ESDM group than for the COM group, but the difference was not statistically significant (676 versus 961, 95% CI for difference = -606, 36). Compared with the COM group, the ESDM group had significantly fewer ABA/EIBI hours (187 versus 327, 95% CI for difference = -276, -3), fewer OT/PT hours (47 versus 90, 95% CI for difference = -84, -1), and fewer speech therapy hours (52 versus 94, 95% CI for difference = -75, -9).

During the postintervention period, total health-related annual service costs were significantly lower for the ESDM group (\$31,962 versus \$50,969, 95% CI for difference = -\$36,154, -\$1,860). Compared with the COM group, the ESDM group incurred significantly lower ABA/EIBI costs (\$11,798 versus \$20,589, 95% CI = -\$17,388, -\$192), lower OT/PT costs (\$2,959 versus \$5,653, 95% CI for difference = -\$5,337, -\$52), and lower speech therapy costs (\$4,010 versus \$7,199, 95% CI for difference = -\$5,745, -\$633).

## DISCUSSION

In this study of the cost offsets associated with early intensive delivery of ESDM, we found that during the intervention period, children who received the ESDM had average annualized total health-related costs that were higher by about \$14,000 than those of children who received community-based treatment, although this difference did not reach statistical significance. The higher cost of ESDM was partially offset during the intervention period because children in the ESDM group used fewer hours of other types of ABA/EIBI and speech therapy services than children in the comparison group. In the postintervention period, compared with children who had earlier received treatment as usual in community settings, children in the ESDM group used fewer hours of other types of ABA/EIBI, OT/PT, and speech therapy services, resulting in significant cost savings in the amount of about \$19,000 per year per child. This finding suggests that costs associated with early intensive ESDM treatment were fully offset within a few years after the intervention because of the reductions in other service use and associated costs.

Postintervention decrease in use and associated costs for other types of ABA/EIBI, as well as OT/PT and speech therapy services for the ESDM group, may be attributed to the positive

effects of the ESDM treatment on child outcomes. Research on the efficacy and effectiveness of the ESDM showed substantial gains in cognitive, language, and social abilities, and adaptive behavior relative to children receiving treatment as usual in the community, which are sustained in all areas 2 years posttreatment.<sup>21–23,37</sup> Reduced use of other forms of ABA/EIBI, OT/PT, and speech therapy hours for the ESDM group likely reflects the reduced need for these therapies due to better child outcomes.

Several factors could underlie the decreased use and associated costs for other types of ABA/EIBI, speech therapy, and general education services for the ESDM group during the intervention period. First, ESDM may have ameliorated some of the impairments that may lead to the need for these services. Second, the ESDM may have addressed the needs these therapies would otherwise address; hence, the parents substituted the ESDM treatment for these therapies. This is likely to be the case for the ABA/EIBI and speech therapy services. Third, the time costs associated with the ESDM (ESDM may have largely consumed free, waking time of the young toddlers) may have precluded the feasibility of using other services. This is likely to be the case for the general education services category, comprising mostly daycare services, in this age group.

We estimated per year per child cost of ESDM as \$45,580. The only other estimate of the ESDM treatment cost, to our knowledge, comes from a Canadian study, which estimated the cost to be \$50,487 in 2013 Canadian dollars per year per child (\$49,835 in 2015 US dollars).<sup>38</sup> This similarity provides confidence in our estimate, as do estimates of service costs (\$40,000–\$80,000) from other studies of the costs of intensive 1:1 early intervention.<sup>13,15,16</sup> Although more detailed costs analysis for different settings and populations are warranted, this initial analysis of the ESDM treatment suggests that it is possible to provide high-quality, evidence-based intervention for young children with ASD within the cost parameters described in previous estimates for such services.

To our knowledge, this is the first study to report individual-level economic data from a clinical trial of an autism treatment in the United States, and it is unique in its analysis of the effects of early intervention based on parental recollection of use and best estimates of spending as opposed to simulated estimates. The study results can serve as a benchmark for further studies related to service needs for preschool-aged children. A strength of our study is the rarely available data from the 2-year follow-up period. In addition, being able to analyze service use measures in conjunction with cost enabled us to obtain a clearer picture of the changes that drove the observed cost offset.

A number of study limitations should be mentioned. First, the statistical power to identify significant differences between study groups with respect to some cost categories was low, given the sample size in this study. Second, we estimated only service use, and did not have measures of the other costs that have been shown to drive total costs among children with ASD, especially the indirect costs associated with caregiving.<sup>39</sup> The ESDM demands a higher level of parental involvement due to parent coaching sessions, which implies significantly higher caregiver time costs. Productivity losses due to such caregiver time costs have been shown to be an important component of societal costs of ASD<sup>31,39</sup>; these costs should be considered when evaluating benefits of the ESDM. Third, service use was based



on parent/caregiver report (rather than record review) and was collected every 6 months, raising concerns about recall bias, although we have no reason to believe that recall would differ across the two study arms. Fourth, the cost of intervention was retrospectively assessed via inputs from the intervention developers, rather than concurrent with its implementation, which could affect the reliability of our estimates. The similarity of our estimates of others from other groups increases our confidence, however. Fifth, generalizability of the service use and cost estimates are likely directly related to the generalizability of estimates of intervention efficacy. More specifically, participating families from both the ESDM and community care groups may have received higher-quality intervention than they would otherwise. For the children in the ESDM group, the expertise of supervising clinicians and the additional resources (e.g., early gold-standard diagnosis and referral) available because of grant funding likely provided a higher standard of care than would be received in community services. For the community children, being randomized to a comparison group may well have motivated the families (who enrolled seeking the experimental treatment) to find all the services possible for their children in the community. In addition, the early diagnosis provided by the study gave them recommendations for needed interventions that may have guided them to receive more services, earlier, than they otherwise would have received. In addition, this sample was recruited from a geographically constrained area, 30 minutes from an urban university, which meant that families from both groups lived close to a number of excellent community intervention providers. Finally, participating families may have further differed from families who did not participate in regard to their ability and motivation to obtain intensive early behavioral intervention for their children. These caveats should be carefully considered when applying our findings to the potential benefits of ESDM and similar treatments delivered in a less controlled community (as opposed to university-based) setting.

Despite these limitations, our results have important implications. Although there is a critical need to evaluate the effectiveness of programs such as ESDM among culturally diverse populations, less-educated families, and lower resource and rural communities,<sup>22</sup> our results, combined with findings from other publications from this same study, suggest that high-quality, professional-delivered, intensive early intervention results in both positive clinical outcomes reflected in gains in cognitive, language, social, and adaptive behavior function<sup>21,22</sup> and in significant cost offsets after treatment. Although it remains to be seen whether these short-term cost savings prevail in the long term, our results have logical policy implications regarding the benefit of identifying young children with autism and providing early high-quality, intensive, and comprehensive developmental behavioral treatment. &

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## REFERENCES

1. Buescher A, Cidav Z, Knapp M, Mandell D. Costs of autism spectrum disorders in the United Kingdom and the United States. *JAMA Pediatrics*. 2014.
2. Barrett B, Byford S, Sharac J, Hudry K, Leadbitter K, Temple K. Service and wider societal costs of very young children with autism in the UK. *J Autism Devel Disord*. 2012;42:797–804. [PubMed: 22089166]
3. Rogers S, Vismara L. Evidence-based comprehensive treatments for early autism. *J Clin Child Adolesc Psychol*. 2008;37:8–38. [PubMed: 18444052]
4. Eldevik S, Hastings RP, Hughes JC, Jahr E, Eikeseth S, Cross S. Meta-analysis of early intensive behavioral intervention for children with autism. *J Clin Child Adolesc Psychol*. 2009;38:439–450. [PubMed: 19437303]
5. Dawson G, Bernier R. A quarter century of progress on the early detection and treatment of autism spectrum disorder. *Dev Psychopathol*. 2013; 25:1455–1472. [PubMed: 24342850]
6. Warren Z, McPheeters M, Sathe N, Foss-Feig J, Glasser A, Veenstra-Vanderweele J. A systematic review of early intensive intervention for autism spectrum disorders. *Pediatrics*. 2011;127:e1303–e1311. [PubMed: 21464190]
7. Reichow B, Barton E, Boyd B, Hume K. Early intensive behavioral intervention (EIBI) for young children with autism spectrum disorders (ASD). *Cochrane Database Syst Rev*. 2012;10:CD009260.
8. Lai M, Lombardo M, Baron-Cohen S. Autism. *Lancet*. 2014;383:896–910. [PubMed: 24074734]
9. Virues-Ortega J. Applied behavior analytic intervention for autism in early childhood: meta-analysis, meta-regression and dose-response meta-analysis of multiple outcomes. *Clin Psychol Rev*. 2010;30:387–399. [PubMed: 20223569]
10. Eikeseth S. Outcome of comprehensive psycho-educational interventions for young children with autism. *Res Dev Disabil*. 2009;30:158–178. [PubMed: 18385012]
11. Green G, Brennan LC, Fein D. Intensive behavioral treatment for a toddler at high risk for autism. *Behav Modif*. 2002;26:69–102. [PubMed: 11799655]
12. Lovaas O. Teaching individuals with developmental delays: basic intervention techniques. Austin, TX: Pro-Ed; 2003.
13. Chasson G, Harris G. Cost comparison of early intensive behavioral intervention and special education for children with autism. *J Child Fam Studies*. 2007;16:401–413.
14. Boudier J, Spielman S, Mandell D. Brief report: quantifying the impact of autism coverage on private insurance premiums. *J Autism Devel Disord*. 2009;39:953–957. [PubMed: 19214727]
15. Motiwala S, Gupta S, Lilly M, Ungar W, Coyte P. The cost-effectiveness of expanding intensive behavioural intervention to all autistic children in Ontario. *Healthcare Policy/Politiques de Sante*. 2006;1:135–151. [PubMed: 19305662]
16. Peters-Scheffer N, Didden R, Korzilius H, Matson J. Cost comparison of early intensive behavioral intervention and treatment as usual for children with autism spectrum disorder in the Netherlands. *Res Dev Disabil*. 2012;33:1763–1772. [PubMed: 22705454]
17. Marcus L, Rubin J, Rubin M. Benefit-cost analysis and autism services: a response to Jacobson and Mulick. *J Autism Dev Disord*. 2000;30:595–598. [PubMed: 11261470]
18. Byford S, Cary M, Barrett B, et al. Cost-effectiveness analysis of a communication-focused therapy for pre-school children with autism: results from a randomised controlled trial. *BMC Psychiatry*. 2015;15:316. [PubMed: 26691535]
19. Romeo R, Byford S, Knapp M. Annotation: Economic evaluations of child and adolescent mental health interventions: a systematic review. *J Child Psychol Psychiatry*. 2005;46:919–930. [PubMed: 16108995]

20. Schreibman L, Dawson G, Stahmer A, et al. Naturalistic developmental behavioral interventions: empirically validated treatments for autism spectrum disorder. *J Autism Dev Disord*. 2015;45:2411–2428. [PubMed: 25737021]
21. Dawson G, Rogers S, Munson J, et al. Randomized, controlled trial of an intervention for toddlers with autism: the Early Start Denver Model. *Pediatrics*. 2010;125:e17–e23. [PubMed: 19948568]
22. Estes A, Munson J, Rogers SJ, Greenson J, Winter J, Dawson G. Long-term outcomes of early intervention in 6-year-old children with autism spectrum disorder. *J Am Acad Child Psy*. 2015;54:580–587.
23. Rogers S, Estes A, Lord C, et al. Effects of a Brief Early Start Denver Model (ESDM)–based parent intervention on toddlers at risk for autism spectrum disorders: a randomized controlled trial. *J Am Acad Child Adolesc Psychiatry*. 2012;51:1052–1065. [PubMed: 23021480]
24. Rogers S, Dawson G. *Early Start Denver Model for Young Children With Autism*. New York, NY: Guilford; 2010.
25. Lord C, Rutter M, Le Couteur A. Autism Diagnostic Interview-Revised: a revised version of a diagnostic interview for caregivers of individuals with possible pervasive developmental disorders. *J Autism Dev Disord*. 1994;24:659–685. [PubMed: 7814313]
26. Lord C, Rutter M, DiLavore P, Risi S. *Autism Diagnostic Observation Schedule*. Los Angeles, CA: Western Psychological Services; 1999.
27. American Psychiatric Association. *Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition* Washington, DC: American Psychiatric Association; 1994.
28. Sinaiko AD, Rosenthal MB. Increased price transparency in health care—challenges and potential effects. *N Engl J Med*. 2011;364:891–894. [PubMed: 21388306]
29. Curtis L. *Unit Cost of Health and Social Care 2011*. Canterbury, UK: Personal Social Services Research Unit, University of Kent; 2011.
30. Amendah D, Grosse S, Peacock G, Mandell D. The economic costs of autism: a review In: Amaral D, Dawson G, Geschwind D, eds. *Autism Spectrum Disorders*. New York, NY: Oxford University Press; 2011: 1347–1360.
31. Buescher A, Cidav Z, Knapp M, Mandell D. Costs of autism spectrum disorders in the United Kingdom and the United States of America. *JAMA Pediatrics*. 2014;168:721–728. [PubMed: 24911948]
32. Agency for Healthcare Research and Quality. *Medical Expenditure Panel Survey*. Available at: <https://meps.ahrq.gov/mepsweb/>. Accessed May 16, 2017.
33. Liberal Arts, Oregon State. *Consumer Price Index (CPI) Conversion Factor for Years 1774 to Estimated 2026 to Convert to Dollars of 2015*. Available at: <http://liberalarts.oregonstate.edu/sites/liberalarts.oregonstate.edu/files/polisci/faculty-research/sahr/inflationconversion/pdf/cv2015.pdf>. Accessed May 16, 2017.
34. Barber JA, Thompson SG. Analysis and interpretation of cost data in randomised controlled trials: review of published studies. *Br Med J*. 1998; 317:1195–1200. [PubMed: 9794854]
35. Thompson SG, Barber JA. How should cost data in pragmatic randomised trials be analysed? *Br Med J*. 2000;320:1197–1200. [PubMed: 10784550]
36. Barber JA TS. Analysis of cost data in randomised controlled trials: an application of the non-parametric bootstrap. *Stat Med*. 2000;19: 3219–3236. [PubMed: 11113956]
37. Vivanti G, Paynter J, Duncan E, et al. Effectiveness and feasibility of the Early Start Denver Model implemented in a group-based community childcare setting. *J Autism Dev Disord*. 2014;44:3140–3153. [PubMed: 24974255]
38. Penner M, Rayar M, Bashir N, Roberts SW, Hancock-Howard RL, Coyte PC. Cost-effectiveness analysis comparing pre-diagnosis autism spectrum disorder (ASD)-targeted intervention with Ontario’s autism intervention program. *J Autism Dev Disord*. 2015;45:2833–2847. [PubMed: 25936527]
39. Cidav Z, Marcus S, Mandell D. Family costs of childhood autism: implications for parental labor supply and earnings. *Pediatrics*. 2012;129: 617–623. [PubMed: 22430453]

**TABLE 1**

Comparison of Mean Annualized Hours and Costs by Study Arm, Intervention, and Postintervention Periods

Service	Intervention Period					
	Annualized Hours			Annualized Costs		
	ESDM	COM	Bootstrapped 95% CI <sup>d</sup>	ESDM	COM	Bootstrapped 95% CI <sup>d</sup>
Total health-related	913 (270)	755 (535)	-101, 417	60,369 (16,015)	45,975 (33,089)	-1,525, 30,314
ESDM	661 (84)			45,580 (5,471)		
ABA/EIBI	107 (190)	490 (433)	<b>-593, -172</b>	6,306 (11,010)	29,554 (25,946)	<b>-35,798, -1,069</b>
OT/PT	45 (46)	69 (56)	-58, 9	2,683 (2,745)	4,200 (3,445)	-3,523, 489
Social skills training	3 (8)	29 (69)	58, 7	247 (614)	2,146 (5,179)	-4,323, 525
Special education	30 (76)	42 (123)	-70, 47	790 (2,022)	1,090 (3,218)	-1,831, 1,232
Speech therapy	64 (50)	121 (120)	<b>-113, -1</b>	4,636 (3,591)	8,758 (8,732)	<b>-8,188, -56</b>
Miscellaneous/other	3 (11)	5 (15)	-10, 6	126 (446)	227 (671)	-442, 241
General education	319 (194)	481 (198)	<b>-301, -22</b>	2,863 (1,743)	4,271 (1,793)	<b>-2,662, -155</b>
Service	Postintervention Period					
	Annualized Hours			Annualized Costs		
	ESDM	COM	Bootstrapped 95% CI <sup>d</sup>	ESDM	COM	Bootstrapped 95% CI <sup>d</sup>
Total health-related	676 (338)	961 (681)	-606, 36	31,962 (17,220)	50,969 (37,945)	<b>-36,154, -1,860</b>
ESDM						
ABA/EIBI	187 (233)	327 (251)	<b>-276, -3</b>	11,798 (14,742)	20,589 (15,856)	<b>-17,388, -192</b>
OT/PT	47 (58)	90 (77)	<b>-84, -1</b>	2,959 (3,632)	5,653 (4,907)	<b>-5,337, -52</b>
Social skills training	48 (66)	101 (203)	-149, 43	3,673 (5,063)	7,763 (15,676)	-11,507, 3,327
Special education	329 (296)	333 (301)	-198, 189	8,938 (8,116)	9,027 (8,168)	-5,348, 5,170
Speech therapy	52 (56)	94 (61)	<b>-75, -9</b>	4,010 (4,308)	7,199 (4,746)	<b>-5,745, -633</b>
Miscellaneous/other	13 (19)	16 (43)	-26, 20	584 (849)	737 (1,939)	-1,187, 880
General education	490 (335)	349 (287)	-42, 325	12,050 (11,059)	7,193 (8,643)	-1,112, 10,826

Note: Significant differences (5% confidence level or lower) are shown in bold. ABA/EIBI = applied behavior analysis/early intensive behavioral intervention; COM = community care; ESDM = Early Start Denver Model; OT/PT = occupational/physical therapy.

For mean difference.

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