

Original Research Article

SOCIETAL PERSPECTIVE OF COST ANALYSIS OF AN EARLY INTERVENTION PROGRAMME FOR AUTISM CHILDREN AND ITS CONTRIBUTING FACTORS IN KLANG VALLEY MALAYSIA, 2019

ABSTRACT

Autism spectrum disorder (ASD) is a public health concern globally, characterized by impairments in cognitive process, social functioning, communication skills, behaviours and interests. These problems require a multidisciplinary approach through an early intervention programme (EIP) which is remarkably expensive. To date, the knowledge of EIP costs for ASD children in Malaysia remains unknown. This study estimated the annual average costs of EIP for ASD children from the societal perspective (total average costs, education costs, treatment costs, and living costs), the relationship between the total annual average costs of EIP and its contributing factors, and the contributing predictors. A cross-sectional cost analysis study using simple random sampling according to proportion was conducted among 280 parents of ASD children receiving EIP services in Klang Valley. The data were collected using interview-based questionnaire and proforma. The Activity-based Costing Technique and Microsoft Excel 2016 were used to estimate the EIP-related costs. The relationship between variables and the total annual average EIP costs were carried out using Statistical Package for Social Sciences version 25. The result showed that the total annual average costs of EIP, average education costs, average treatment costs, and average living costs per ASD child were estimated at RM15,158; RM3,896; RM1,948; and RM4,409 respectively. There was a significant association ($P = .05$) between total annual average costs of EIP, and its contributing factors; family income, ASD level, programme characteristics, and family support. Multiple regression analysis showed that 27% of the total annual average costs of EIP can be predicted by the combination of these factors; EIP types, EIP frequency, centre-based type, ASD level and family income. In conclusion, the total annual average costs of EIP, average education costs, average treatment costs, and average living costs per ASD child were estimated at RM15,158; RM3,896; RM1,948; and RM4,409 respectively, and it can be predicted by the ASD level, family income, and EIP structure in term of its programme type, frequency, and centre-based type. This information is beneficial as evidence to assist the decision on future resource allocation by the policy maker.

Keywords: cost-analysis, total annual average costs, education costs, treatment costs, autism spectrum disorder, early intervention programme

1. INTRODUCTION

Autism spectrum disorder (ASD) is a public health concern globally with increased prevalence over the last decade one of the commonest developmental disability affecting children worldwide [2][3], ASD is characterized by impairments and restrictions in cognitive process, social functioning, communication skills, behaviors and interests [1][4][5]. The term "spectrum" reflects the wide variation in challenges and strengths possessed by each child with ASD. These problems

Comment [m1]: Check this statement carefully, there seems to be some repetition.

Comment [m2]: Stop mentioning software used for analysis and state specific estimation techniques employed by the study.

Comment [m3]: Repeation

Comment [m4]: Check all your headings and sub headings for consistency. While some are in caps throughout, others are in sentence cases or each word beginning with caps.

mandated multidisciplinary assessment and management by a multidisciplinary team consisting of family medicine specialist, pediatrician, psychiatrist (child and adolescent), psychologist (clinical and educational), counsellor, occupational therapist, speech-language therapist, medical social worker, and education officers [6].

In managing ASD cases, an early intervention programme (EIP) is an important intervention to determine the magnitude of disease outcomes in terms of cognitive, behavioral and social adaptability [6][7][8]. EIP should be offered to all children diagnosed with ASD as early as 36 months old to improve the outcomes. The models of EIP included home-based or centre-based program [9]; which involved integration of physical therapy, speech and language therapy, occupational therapy, psychological therapy, behavioral therapy, special education, social welfare and medical interventions [6][10]. To achieve the best outcome for children with ASD across lifespan, EIP can be one of the alternatives [11].

Regarding ASD's assessment, diagnosis and treatment, it requires comprehensive, intensive and multisectoral management approach [6][10]. Since it requires a multisectoral approach, EIP is a crucial component and it is very expensive [12]. Studies from developed countries such as United Kingdom and United States showed that, the total average costs of managing a child with ASD in a lifetime ranged from RM4.4 million to RM5.9 million [13]. However, the specific information on the total annual average costs of EIP per ASD child remains unknown in a centre-based EIP in Malaysia, as no study has been done before. Knowledge on average costs of programme (including average costs of EIP) is important for policy development and resource distribution [9][14][15].

1.1. Specific Objectives

- i. To determine the annual average costs of EIP per ASD child (total average costs, average education costs, average living costs) from the societal perspective.
- ii. To determine the relationship between the total annual average costs of EIP and its contributing factors.
- iii. To determine the predictors to the total annual average costs of EIP.

Comment [m5]: Same as objective iii

1.2. Research Questions

The following questions shall be the focus of this study

- i. What are the annual average costs of EIP per ASD child (total average costs, average education costs, living costs) in the centre-based EIP in Klang valley/Valley, Malaysia?
- ii. What are the contributing factors to the total annual average costs of EIP for ASD child?
- iii. What are the predictors to the total annual average costs of EIP for ASD child?

Comment [m6]: Don't use future tenses because the study has been completed.

Comment [m7]: These questions are the same.

1.3. Research Hypothesis

H₀: There is no significant relationship between the total annual average costs of EIP per ASD child and its contributing factors.

H₁: There is a significant relationship between the total annual average costs of EIP per ASD and its contributing factors.

Comment [m8]: This is contradictory because if something contributes to something, then is it statistically significant.

Rather you can state that "There is no significant relationship between the total annual average costs of EIP per ASD child on one hand and family income, ASD level, programme characteristics, and family support on another hand.

So the reverse would be for the alternative hypothesis.

Comment [m9]: Kindly drop the hypothesis and merge the objectives and research questions with the introduction using sentences.

Comment [m10]: State in full when first used.

1.4. Epidemiology of ASD

Based on epidemiological studies conducted over the past 50 years, the prevalence of ASD appears to be increasing globally, due to increase in screening tools with more detailed DSM-IV diagnostic criteria, which allow more comprehensive coding of the prevalence data. In a recent estimate by the World Health Organization (WHO), prevalence of ASD was estimated at 1 in 160 children [16]. The prevalence of ASD in the United States has increased tremendously over the past 5 years with the a rate of 16.8 per 1,000 (one_1 in 59) children aged 8 years in total as an overall [17]. In Malaysia, there is no official registry system available to monitor epidemiological data on the prevalence of ASD at present. However, a smaller scale study by the Ministry of Health on children between the ages 18 to 36 months showed a rate of 1.6 in 1000 children, or approximately 1 in 625 children [18][6]. In terms of cognitive impairments, about 30% to 50% of children with ASD was_were estimated to have intellectual disability with IQ below 85 and have poorer adaptive behavior that contribute to ineffective social communication [19]. As indicated in the available scientific evidence currently, environmental and genetic factors were involved with development of ASD, and no clear evidence of a causal relationship between measles, mumps and rubella vaccine, and ASD [11][16]. Older parents are at a higher risk to have children born with ASD, and parents who have ASD child have a 2 to 18 percent chance of having a second child who is_alsowould be affected by ASD [11].

84 | An individual with ASD has limited capacity to perform daily works and ~~having~~ ~~have~~ difficulties to participate in society,
85 | because of poor communication skills that may affect their educational and social attainments as well as employment
86 | opportunities. While some individuals with ASD can live independently, others have severe disabilities and require life-
87 | long care and support [5]. The children with ASD and their families may experience significant emotional and economic
88 | burden, by which children with more severe ASD require strong support for services accessibility and intensity. In the
89 | United States and United Kingdom, the lifespan costs to support a child with ASD was estimated at RM5.9 million and
90 | RM4.4 million, respectively [13]. Notwithstanding, with the presence of intellectual disability, higher costs were expected.
91 | Regarding medical expenditure, children with ASD incurred 4.1 higher cost compared to those children without ASD and it
92 | was expected to be increased exponentially following the rise in prevalence for the next decade, without an effective
93 | intervention [11]. In Malaysia, the total economic costs experienced by parents was estimated at RM35,365 per year [10].
94 |

95 | 1.5. Review of Factors Contributing to ASD-related Costs

96 | ~~The~~ ~~A~~ systematic review was conducted alongside with the research development to comprehensively assess the factors
97 | ~~contributed~~ ~~contributing~~ to the EIP-related costs for ASD children. The systematic search was conducted based on the
98 | research question on what ~~are the~~ contributing factors of the average cost of EIP among ASD children. The objective of
99 | the review ~~is was~~ to identify the average costs of EIP among ASD children from various sources. The search was
100 | conducted through electronic databases Ovid MEDLINE(R) and Epub Ahead of Print, In-Process & Other Non-Indexed
101 | Citations, Daily and Versions (R) from 1946 to November 16, 2018 using the combination of MeSH keywords and its
102 | related scopes available as follows; "Autism Spectrum Disorder" or "Autistic Disorder" and "Early Intervention (Education)"
103 | or "Early Medical Intervention" and "Costs and Cost Analysis". General search engine was used through Google Scholar
104 | to search for additional related articles using similar keywords combination. The detailed information on keywords and ~~the~~
105 | ~~their~~ combination was tabulated in **Appendix 1**. The search exercise was focused on the economic evaluation studies
106 | pertaining to ASD and its intervention, full text articles, scholarly journals and English articles that were published for the
107 | past 20 years. After selection, checking for duplicates, and hand search, 15 articles were finally selected, and their
108 | contents were then reviewed (~~see Appendix 2~~ – The search process according to PRISMA guideline). Two review
109 | authors extracted data independently from each study regarding type of study, type of intervention, ~~published~~ ~~year of~~
110 | ~~publication~~, journal, outcomes of the study, and the location of study. The quality of the reviewed articles was not further
111 | assessed as the credit of journals was considered enough. Based on the medical model of health determinants adapted
112 | from Dahlgren and Whitehead (1991) framework, the retrieved factors were categorized into socio-demographic factors
113 | (age of child, family income, residence location), medical condition factor (ASD level), health service factors (centre-based
114 | type, programme characteristics), and environmental factors (family support, EIP centre distance). The summary of
115 | results on factors contributing to ASD costs was tabulated in **Appendix 3**.
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117 | 1.6. Costs concept

118 | The cost concept is defined as the analysis of comparative costs of alternative treatments or health care programmes
119 | [20]. A costing exercise starts with the formation of a well-defined decision problem, including the objectives of costing,
120 | the perspective of costing, and the time horizon, as well as the description of a service. The perspective for an analysis
121 | may be that of a specific provider or providing institution, the patient or groups of patients, a third-party payer (public or
122 | private), or a broad perspective (i.e. all costs and consequences to whomsoever they accrue). After the service for costing
123 | has been defined in detail, the costing methodologies follow three distinctive steps: the first step is identification of
124 | resources used to deliver the service, second is the measurement of resource utilization in natural units, and ~~the third~~
125 | ~~one is~~ attaching monetary value to resource used.
126 |

127 | 1.7. Costing methodology

128 | The common costing methods used in healthcare, ~~including~~ ~~are~~ the traditional step-down approach (macro-costing),
129 | micro-costing and activity-based costing (ABC) [20]. Macro-costing is ~~the a~~ top-down approach, ~~it~~ involves the process of
130 | allocating ~~the~~ overhead cost to ~~the to the~~ departments or the units of services based on service volume to estimate the
131 | average unit costs. Micro-costing approach involves the collection of data about resource use by each individual service
132 | (item-by-item and line-by-line). This bottom-up approach can be used where the accuracy of resource measurement is
133 | important, contingent on ~~the~~ data availability, ~~and~~ whether it is feasible in an economically sensible way. Micro-costing is
134 | the most precise estimation compared to the traditional macro-costing approach, which ~~is~~ based on average estimation
135 | including case-mix, disease cost per diem and average cost per diem. The ABC method ~~involved~~ ~~involves~~ the step-down
136 | approach at first. Next, the shared activities need to be identified (activity-centre) and the total cost will be allocated down
137 | to the departments in proportion according to their consumption of resources (cost driver). The department costs will then
138 | be divided ~~into~~ its service volume to estimate the unit costs. The cost calculated will be the average cost and presented as
139 | average cost per contact point or per visit or per service.
140 |

Comment [m11]: Correct the use of caps

Comment [m12]: Are these sources also?

Comment [m13]: State in full when used for the first time

Comment [m14]: There is no appendix attached.

Comment [m15]: Source

Comment [m16]: Source. If same source as 20, then indicate it at the end of the paragraph and delete that of the first sentence.

2. METHODOLOGY

A cross-sectional costs analysis study using simple random sampling according to proportion was conducted among 280 parents of ASD children receiving EIP services in the Klang Valley, which include governments and non-governments EIP centres. Parents of all ASD children aged less than 7 years old which enrolled in EIP centres in the Klang Valley were included, and those ASD children with other co-morbidities were excluded from the study. The required sample size was calculated according to the hypothesis testing formula for two groups by Lemeshow et al. (1990). To get an adequate sample unit from the sampling frame, 2 governments and 2 non-governments-EIP centres that match the centre-based criteria were selected using the lottery method. The data on contributing factors were collected using interview-based questionnaire and a proforma was used to obtain the data on EIP-related costs. The research instrument was administered and retrieved personally by the researcher. The instrument for the study was submitted to the health economics experts for validation. The Cronbach's Alpha and Intra-Class Correlation were used to determine the reliability of the instrument. Both descriptive and inferential statistics were performed. The relationship between the total annual average costs of EIP, its contributing factors and its predictors were carried out using Statistical Package for Social Sciences version 25.

Comment [m17]: State the specific inferential estimation technique employed.

The cost components included were the direct costs for EIP-related intervention. The indirect costs such as parents' productivity loss, time-off work, and opportunity cost were excluded from the estimation. The provider costs (including capital, staff, utilities, maintenance) were estimated using an ABC method that estimates the total annual average costs of EIP according to the proportion of resources allocated for EIP services. The patient cost was estimated from the reported annual out-of-pocket expenditure (OOP) by the parents, and the breakdown of cost was estimated based on the proportion of annual OOP for ASD-related intervention [21]. From the societal perspective, the total annual average costs of EIP in each centre were estimated using the formula as follows:

$$\frac{\sum(Capital^1 + staff^2 + utilities^3 + maintainence^4 + patient)}{Total\ number\ of\ enrolled\ students}$$

Comment [m18]: Is it not the total number of ASD patients?

¹costs of building and asset

²salary and allowances for the multidisciplinary team and administrative officers

³electrical bills, water bills, internet bills, phone bills, security services

⁴cleaning services, laundry, water filter, waste management

3. RESULTS AND DISCUSSION

In this study, the overall response rate is was 93.3%. The distribution of continuous variables was determined statistically. The Shapiro-Wilk test is was significant ($P = .05$) for all continuous variables, showing the data is not normally distributed. Thus, the non-parametric tests were used to determine the relationship between the contributing factors and the study outcome.

3.1. Descriptive Statistics

The median age of ASD children was 37 months old, IQR (30,48). The range of children's age was 69 months with the youngest at 15 months old and the oldest at 84 months old. The provision of EIP in this study was limited to preschool children age less than 84 months old. In general, EIP services were provided by most EIP centres among preschool age group of ASD children, because EIP is believed by many to be most effective when provided at an early age [8][10]. In Malaysia, the EIP service provision in the public centre-based was limited to the preschool age to date, and most EIP centres were concentrated in urban areas due to because demand for services is higher compared to the rural areas. The gender for both male and female were not equally distributed with the percentages of being 85.3% and 14.7% respectively. The finding was corresponding corresponds with the global prevalence of ASD that showed males have are four times of more likely to be diagnosed with ASD than females [11][17]. The family income distribution shows that 79.8% of the respondents were earning less than RM100,000 per year. The distribution of ASD severity among respondents' children was defined by the level of support, according to the CPG Malaysia, (2014) which adopted the DSM-IV criteria. The severity of ASD was categorized into three level; Level 1 (Requiring support), Level 2 (Requiring substantial support) and Level 3 (Requiring very substantial support). The data on parent's support was retrieved during parents' interview on sharing their experience in handling and managing ASD children in the public and community activities. The reported support level for ASD children, mostly fall within the category of Level 1 and 2 ASD (88.7%), followed by a small portion of ASD children fall within Level 3 (11.3%).

Comment [m19]: State in full before acronym

Comment [m20]: Then why the oldest children bieng 84 months old as stated above?

Comment [m21]: State in full before acronym

The features of health services involved the types of EIP consumed, programme intensity (EIP frequency and duration), and centre-based type. As a multidisciplinary approach intervention, EIP incorporated various types of interventions such as Applied Behavioural Analysis (ABA) therapy, speech therapy, occupational therapy, sensory integration and music therapy [6]. The finding shows that 76.9% of respondents' child received more than 1 EIP types per session, 53.8% received more than 2 EIP sessions per week and 58% received more than 5 hours of EIP sessions per week. Around 60.9% of ASD children received EIP at a public EIP centre. Regarding the distance of EIP centre, 51.3% of the respondents stay near (less than 20km). The range of the nearest and the farthest EIP centre distance was 60km. In assessing EIP-related services, home relocation was reported by 29.8% of respondents due to logistic reasons such as change to a new workplace that was near to the EIP centre, moving to a house that nearer to the EIP centre and other reasons.

Comment [m22]: There is no Table showing the descriptive statistics for us to cross check

3.2. Estimation of Total Annual Average Costs of EIP

3.2.1. Annual provider costs

The summary of the annual average costs of EIP from the provider perspective can be seen in **Table 1**. The detail information on cost estimation for each category (staff emolument, utilities, maintenance, and capital) was tabulated in **Appendix 4**. The component of staff emolument is the highest burden estimated for the total annual average costs of EIP from the provider perspective (75% to 95%).

Table 1. The estimation of annual average EIP costs from provider perspective (N=238)

EIP centre	Staff ¹ (RM)	Utilities ² (RM)	Maintenance ³ (RM)	Building ⁴ (RM)	Total ⁵ (RM)
Centre 1	3,371.88	135.91	148.65	858.81	4,515.25
Centre 2	5,425.00	64.00	70.00	404.42	5,963.42
Centre 3	4,368.00	56.32	61.60	187.17	4,673.09
Centre 4	4,352.40	53.46	58.47	-	4,464.33

¹Average annual staff costs per child

²Average annual utilities costs per child

³Average annual maintenance costs per child

⁴Average annual building costs per child

⁵Total average annual costs of EIP per child

3.2.2. Annual patient costs

The summary of the annual average costs of EIP from the patient perspective can be seen in **Table 2**. The detail information on cost estimation for each category (education costs, treatment costs, and living costs) was tabulated in **Appendix 4**.

Table 2. The estimation of annual average EIP costs from patient perspective (N=238)

Cost components	Proportion*	Centre 1 (RM)	Centre 2 (RM)	Centre 3 (RM)	Centre 4 (RM)
Education costs	0.38	1,839.20	6,992.00	2,574.50	4,180.00
Treatment costs	0.19	919.60	3,496.00	1,287.25	2,090.00
Living costs:	0.43	2,081.20	7,912.00	2,913.25	4,730.00
Total**	1.00	4,840.00	18,400.00	6,775.00	11,000.00

*Proportion of total OOP expenditure on EIP-related activities; adapted from Roddy et al., 2014

**Reported by parents of ASD children from 4 different EIP centres in Klang Valley

Comment [m23]: Make the summation of figures are correct

3.2.3. Total annual average costs of EIP

The summary of the total annual average costs of EIP from the societal perspective can be seen in **Table 3**. The total annual average costs of EIP, education costs, treatment costs, and living costs per ASD child were estimated at RM15,158; RM3,896; RM1,948; and RM4,409 respectively. The estimated total annual average costs of EIP per ASD child in selected government EIP centres ranged from RM 9,358 to RM24,384, and from RM11,437 to RM15,458 for non-government EIP centres. As a partial government sponsored EIP centre, the lowest cost was estimated for Centre 1. In

Comment [m24]: Then why did you state that you chose two government and two non-government centres? Where would this one fall?

Comment [m25]: Where are these figures from? From which tables can they be seen?

contrast with the other government EIP centre, namely Centre 2, highest cost was estimated due to the special EIP structure with lower staff-student ratio and different financing structure under Ministry of Education. For non-government EIP centres, higher cost estimation was expected due to profit-related components (Centre 3 and Centre 4).

Comment [m26]: Check this sentence for accuracy

Table 3. The estimation of total annual average EIP costs from societal perspective (N=238)

EIP centre	Patient costs ¹ (RM) (a)	Provider costs ² (RM) (b)	Total EIP costs ³ (RM) (a) + (b) = (c)
Centre 1	4,840.00	4,515.25	9,355.25
Centre 2	18,400.00	5,963.42	24,363.42
Centre 3	6,775.00	4,673.09	11,448.09
Centre 4	11,000.00	4,464.33	15,464.33
Total annual average costs ⁴ of EIP per child			15,157.77

¹Total annual average costs of EIP reported by the parents of ASD children in 2018

²Total annual average costs of EIP (Provider) = Total costs (staff + utilities + maintenance+ capital)

³Total annual average costs of EIP per child (each centre) = Provider costs +Patient costs

⁴Total annual average costs of EIP per child from 4 centres

Comment [m27]: Is this same as cost of building? If so, use building for consistency.

In this study, the estimated amount is very much lower compared to the findings reported in other countries, apart from the currency exchange rate (Table 4). In the Netherlands, the outcome of interest is the societal cost of childhood ASD and the relationship between state and family OOP expenditure. The cost estimation was 15.5 times higher compared to the current study, because the bottom-up prevalence-based cost-of-illness methodology was adopted from a societal perspective [21]. The cost of special needs assistants and the parent's time-off were incorporated in the cost estimation. In addition, the a similar study involved the cost estimation from an older age-group, of which 2 to 18 years old ASD children were involved, and mostly received private ASD-related care services.

Comment [m28]: Which study are you referring to?

Comment [m29]: Which study are you referring to?

~~In contrast~~ Also, higher cost was demonstrated in Egypt because home-based EIP was practiced widely as an intervention of choice, due to limitation of an EIP-related services by the state provider [12]. In view of the above, the parent's involvement in providing EIP as a home-based programme was estimated as the full time equivalent (FTE) cost per hour based on the salary rate of EIP staff in centre-based setting. The cost parameters were estimated solely based on the home-based EIP resource utilization by ASD child's family. The total estimated cost was mostly influenced by staff cost due to low staff to student ratio observed in the home-based EIP, of which 1 private therapist to 1 ASD child ratio per home EIP session existed.

Comment [m30]: Source

In Australia, the A cost analysis study was conducted to compare the expenses between families with immediate ASD diagnosis following an identification of atypical neuro-developmental delay with families that experienced a delay between first identification and ASD diagnosis [22]. The estimated total ASD-related cost was 12.6 times higher compared to the current study, because the components of treatment-related travel cost, family productivity loss and medical costs were included in the cost estimation, apart from the ASD-therapeutic cost. The total average cost of ASD-related treatments from this study were contributed mainly by loss of income of the parents and caregivers (89%), followed by ASD-related travel costs (3%) and treatment costs (8%).

Comment [m31]: Which study are you referring to?

Table 4. Comparison of total annual average cost of EIP with other countries (N=238)

Country	Study	Average EIP cost* (RM)	Comparison (times higher)
Netherland	[21]	202,367.22	15.50
Egypt	[12]	169,476.99	12.98
Australia	[22]	164,584.28	12.61
United Kingdom	[13]	54,776.09	4.20
United States	[15]	42,253.85	3.24
Klang Valley, Malaysia	Present study, 2018	15,157.77	-

*Estimated annual cost per ASD child; adjusted with 3% discounting; all values converted to Ringgit Malaysia (RM) currency as at November 2018 (1USD=RM4.19, 1GBP= RM5.35, 1EUR=RM4.71)

Comment [m32]: In the title you used 2019

3.2.4. Sensitivity analysis

The Deterministic Sensitivity Analysis (DSA) was done to test the robustness of the conclusions, by determining how the values of cost parameters impact the total annual average costs of EIP in the next 5 years under a set of assumptions

290 as follows; fixed number of students in EIP center, fixed number of EIP staff, fixed EIP programme structure, and 3%
 291 discounting applies for the next 5 years. DSA was performed towards multiple parameters within the base value (total
 292 annual average costs of EIP per ASD child); to check the extent of the possible economic impact of current cost analysis.
 293 A Univariate SA was conducted to assess the parameters by varying $\pm 25\%$ of the base value (Figure 1). The DSA shows
 294 that the total annual average costs of EIP is most sensitive towards the living costs, staff cost, and education costs
 295 compared to others (about 14% to 15% positive incremental following increase 25% of the base values, and 7% to 10%
 296 negative incremental following decrease 25% of the base values).

Comment [m33]: Check for comprehensiveness

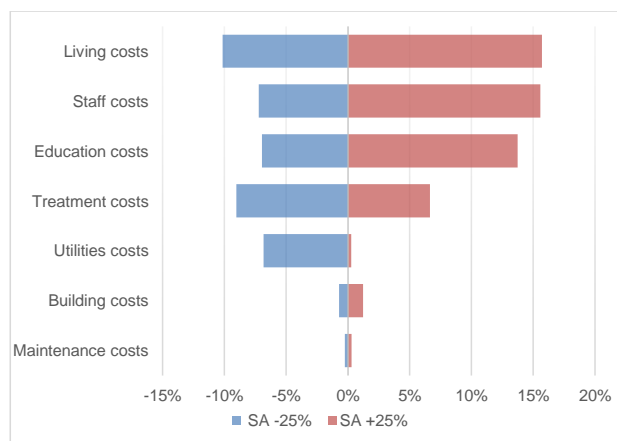


Fig. 1. Univariate Sensitivity Analysis for the average EIP cost (N=238)

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3.2. Relationship between Total Annual Average Costs of EIP and Its Contributing Factors.

As shown in Table 5, there is a significant relationship ($P = .05$) between the total annual average costs of EIP and its contributing factors as follows; family income, ASD level, EIP types, EIP frequency, EIP duration, centre-based type, home relocation and family support. Therefore, the null hypothesis for those factors were rejected.

Table 5. Summary of bivariate analysis

Contributing factors	Total annual average costs of EIP		
	X ²	df	Asymp. Sig. (2-sided)
Age of child	10.710	4	.030
Family income	39.486	4	.001**
ASD level	6.422	2	.040**
EIP types	32.87	4	.001**
EIP frequency	18.042	4	.001**
EIP duration			.012*
Centre-based type			.001*
Centre-based distance			.245
Home relocation			.005*
Family support			.020*

Notes: *P value at $< .05$; significant Mann-Whitney U test
 **P value at $< .05$; significant Kruskal-Wallis with post-hoc test

Comment [m34]: Do you mean you used different tests for different significance levels?

3.3. Predictors to the Total Annual Average Costs of EIP

Multiple linear regression (MLR) was used to identify the predictors for average EIP cost. The 'ENTER' and 'STEPWISE' methods were used. For the preliminary model, the multiple regression statistically significant predicted average of EIP

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cost, $F(7,957) = 12.506$, $P = .05$, adj. $R^2 = 0.280$. However, only five were statistically significant to the prediction ($P = .05$), except for EIP duration ($P = .257$), home relocation ($P = .172$) and family support ($P = .099$). The Five-five significant variables remained were EIP type, EIP frequency, centre-based type, ASD level, and family income. For the final model, the multiple regression model statistically significant predicted average EIP cost, $F(8,001) = 12.506$, $P = 0.05$, adj. $R^2 = 0.270$. The summary of multiple linear regression can be seen in Table 6.

Comment [m36]: Check for comprehension

The final predictive model was derived as follows: Total annual average costs of EIP = 7799.599 + 6593.020 (family income) + 5688.508 (ASD level) + 3731.508 (EIP type) + 3639.020 (EIP frequency) + 4300.819 (centre-based type).

Table 6. Summary of multiple linear regression (N=238)

Variable	Unstandardized coefficient		Standard coefficient	t	p-value	95% confidence interval (CI)	
	B	SE				Lower bound	Upper bound
Constant	7799.599	1258.275		6.199	.001	5320.493	10278.705
Family income	6593.020	1332.215	.283	4.949	.001	3968.234	9217.806
ASD level	5688.508	1668.629	.193	3.409	.001	2400.905	8976.112
EIP type	3731.508	1266.203	.168	2.947	.004	1236.782	6226.234
EIP frequency	3639.020	1062.830	.194	3.424	.001	1544.987	5733.054
Centre type	4300.819	1092.592	.224	3.936	.001	2148.148	6453.491

Comment [m37]: In the methodology, you must indicate how all these predictors were measured. Also the coefficients are too big which is not the best. The authors may consider dividing the numeric figures by a common denominator before running the regression. Also, since it is linear regression, tests such as heteroscedasticity, autocorrelation, and multicollinearity should be conducted to ascertain the robustness of the results.

Inference was made to reference group. Based on the final model, 27% of average EIP cost can be explained by the predictors. Respondents with annual family income of more than RM100,000 are expected to have RM6,593 higher cost than the families with lower annual family income. Respondents having more severe ASD children (Level 3) require RM5,688 higher cost in providing support compared to less severe ASD children (Level 1 and 2). Respondents with ASD children received more than 1 type of EIP per session require RM3,732 higher cost than the counterpart. Respondents with ASD children received 3 and more EIP sessions per week require RM3,639 higher cost than the counterpart. Respondents with ASD children in the private EIP centre-based require RM4,300 higher cost than children in the public EIP centre-based.

In determining the financial burden of families with ASD child, there was a significant difference between household income groups with services rehabilitation ($P = .05$). To improve the health outcome, the families with above average household income groups put a higher commitment to their child's therapy, compared to lower household income group [10]. Among the very low-income group, trade-offs were made between expenditures for the special needs' child and basic living expenses and between care of children and other demands. The complex relationships between disability, poverty, and welfare does expose the families with ASD children at greater risk of experiencing financial hardship [23]. The demands of autism diagnosis and treatment in lower household income group can seriously outstrip the family resources. In US, the annual cost of intervention for ASD child was estimated 3 to 5 times higher among very dependent and semi dependent group of ASD children, compared to normal children [24]. Due to severe restriction in cognitive, social communication and rigidity of behaviour, high ASD level usually require child assistance that able to monitor the child all the time. In UK, cost for ASD child with more severe levels (presence of an intellectual disability) require 2 to 3 times higher cost compared to ASD child without intellectual disability [25], of which greater resources incurred to the families and caregivers for an intensive EIP-related therapy. The largest contributors to total costs in across all age groups were special education, including early intervention services, and indirect costs, such as parental productivity loss. Regarding the programme characteristics, the school services were the biggest contributor to costs associated with childhood ASD ($P = .05$), due to the increased use of special education services [15].

4. CONCLUSION

The total annual average costs of EIP, average education costs, average treatment costs, and average living costs per ASD child were estimated at RM15,158; RM3,896; RM1,948; and RM4,409 respectively. There is a relationship between total annual average costs of EIP, and its contributing factors as follows; family income, ASD level, EIP types, EIP frequency, EIP duration, centre-based type, home relocation, and family support. Following the multiple regression analysis, the finding shows that 27% of an average EIP cost can be predicted by the combination of these factors; EIP types, EIP frequency, centre-based type, ASD level and family income.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

CONSENT

All authors declare that written informed consent was obtained from the patient (or other approved parties) for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editorial office/Chief Editor/Editorial Board members of this journal.

ETHICAL APPROVAL

This study was registered under the National Medical Research Register (Ref.: NMRR-19-936-45682). The ethical approval to carry out this research was obtained from the Human Ethical Committee of Universiti Putra Malaysia (Ref.: JKEUPM-2019-080). Permission was obtained from the facilities involved in the study [(Ref.: KURNIA.600-8/2/2(10) dated 22 April 2019); (UKM FND/237/2 dated 23 April 2019); approval feedback forms (Lampiran E dated 10 May 2019 and 17 May 2019)]. The consent from respondents (parents) was obtained prior to the data collection.

REFERENCES

- [1] T. A. Lavelle, "Examining Health and Economic Outcomes Associated with Pediatric Medical Conditions in the United States," Harvard University, 2012.
- [2] World Health Organization, "Children and Neurodevelopmental Behavioral Intellectual Disorders (NDBID)," in *Children's Health and the Environment WHO Training Package for the Health Sector*, 2011, pp. 1–51.
- [3] N. Adab *et al.*, "Children and neurodevelopmental behavioral intellectual disorders," *Int. J. Epidemiol.*, vol. 12, no. 3, pp. 1–51, 2014.
- [4] American Psychiatric Association, "Autism spectrum disorder," *Curr. Biol.*, vol. 15, no. 19, pp. R786–R790, 2013.
- [5] T. Ting, X. Neik, and L. W. Lee, "Prevalence, Diagnosis, Treatment and Research on Autism Spectrum Disorders (ASD) in Singapore and Malaysia," *Int. J. Spec. Educ.*, vol. 29, no. 3, pp. 82–92, 2014.
- [6] M. Ministry of Health, *Clinical Practice Guideline Management of Autism Spectrum Disorder in Children and Adolescent*. 2014.
- [7] Z. Cidav, J. Munson, A. Estes, G. Dawson, S. Rogers, and D. Mandell, "Cost Offset Associated With Early Start Denver Model for Children With Autism," *J. Am. Acad. Child Adolesc. Psychiatry*, vol. 56, no. 9, pp. 777–783, 2017.
- [8] Autism Speaks, "What Is Autism?," 2018. [Online]. Available: <https://www.autismspeaks.org/what-autism>. [Accessed: 14-Oct-2018].
- [9] C. M. Escobar, W. S. Barnett, and U. D. Goetze, "Cost Analysis in Early Intervention," *J. Early Interv.*, vol. 18, no. 1, p. 1, 1994.
- [10] S. Kamaralzaman, H. Toran, S. Mohamed, and N. B. Abdullah, "The economic burden of families with autism spectrum disorders (ASD) children in Malaysia," *J. ICSAR*, vol. 2, no. 1, pp. 71–77, 2018.
- [11] Autism Speaks, "Autism Facts and Figures," 2018. [Online]. Available: <https://www.autismspeaks.org/autism-facts-and-figures>. [Accessed: 17-Oct-2018].
- [12] Mendoza, "The Economics of Autism in Egypt," *Am. J. Econ. Bus. Adm.*, vol. 2, no. 1, pp. 12–19, Jan. 2010.
- [13] A. V. S. Buescher, Z. Cidav, M. Knapp, and D. S. Mandell, "Costs of autism spectrum disorders in the United Kingdom and the United States," *JAMA Pediatr.*, vol. 168, no. 8, pp. 721–728, 2014.
- [14] M. L. Ganz, "The Lifetime Distribution of the Incremental Societal Costs of Autism," *Arch. Pediatr. Adolesc. Med.*, vol. 161, no. 4, p. 343, Apr. 2007.
- [15] T. A. Lavelle, M. C. Weinstein, J. P. Newhouse, K. Munir, K. A. Kuhlthau, and L. A. Prosser, "Economic Burden of Childhood Autism Spectrum Disorders," *Pediatrics*, vol. 133, no. 3, pp. e520–e529, 2014.
- [16] WHO, "Key Facts," *World Health Organization Web Page*, 2018. [Online]. Available: <https://www.who.int/news-room/fact-sheets/detail/autism-spectrum-disorders>. [Accessed: 24-Sep-2018].
- [17] CDC, "Prevalence of Autism Spectrum Disorder Among Children Aged 8 Years — Autism and Developmental Disabilities Monitoring Network, 11 Sites, United States, 2014," *MMWR. Surveill. Summ.*, vol. 67, no. 6, pp. 1–23, 2018.
- [18] H. Azizan, "The Burden of Autism," *The Star Online-for write up*, 2008. [Online]. Available: <http://www.thestar.com.my/story/?file=/2008/4/27/focus/21080181&sec=focus>. [Accessed: 28-Sep-2018].
- [19] T. Charman, A. Pickles, E. Simonoff, S. Chandler, T. Loucas, and G. Baird, "IQ in children with autism spectrum disorders: Data from the Special Needs and Autism Project (SNAP)," *Psychol. Med.*, vol. 41, no. 3, pp. 619–627, 2011.

- 428 [20] M. F. Drummond, M. J. Sculpher, K. Claxton, G. L. Stoddart, and G. W. Torrance, *Methods for the Economic*
429 *Evaluation of Health Care Programmes (4th Edition)*. 2015.
- 430 [21] A. Roddy and C. O'Neill, "The economic costs and its predictors for childhood autism spectrum disorders in
431 Ireland: How is the burden distributed?," *Autism*, 2018.
- 432 [22] C. Horlin, M. Falkmer, R. Parsons, M. A. Albrecht, and T. Falkmer, "The cost of autism spectrum disorders," *PLoS*
433 *One*, vol. 9, no. 9, 2014.
- 434 [23] D. L. Sharpe and D. L. Baker, "Financial issues associated with having a child with autism," *J. Fam. Econ. Issues*,
435 vol. 28, no. 2, pp. 247–264, 2007.
- 436 [24] S. S. Motiwala, S. Gupta, M. B. Lilly, W. J. Ungar, and P. C. Coyte, "The cost-effectiveness of expanding intensive
437 behavioural intervention to all autistic children in Ontario," *Healthc. Policy*, vol. 1, no. 2, pp. 135–51, 2006.
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