Parental Healthcare-Seeking Behaviors of Filipino boys with Duchenne Muscular Dystrophy

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Abstract

Background. Duchenne muscular dystrophy (DMD) is the most commonly encountered X-linkeddystrophinopathy. Affected children progressively develop symptoms. With prompt and proper interventions,complications are delayed or prevented. Appropriate health care-seeking behavior and effective medicallogistics would lead to early diagnosis. Hence, this study aimed to assess caregiver's health care seeking behavior for their children with DMD in a developing country.

Methods: This study utilized a prospective cross-sectional design to investigate the factors affecting the health seeking behavior of caregivers of genetically-confirmed DMD patients at the Multidisciplinary Neuromuscular Clinic of the University of the Philippines – Philippine General Hospital. Descriptive statistics were used to summarize the demographic and clinical characteristics of the patients and caregivers. Medians and Interquartile Range (IQR) were used for continuous exposures and Chi-Square Test in assessing associations of categorical exposures.

Results: A total of 55 parents participated in the study. There is a notable delay between the mean age when parents noticed the first symptom (4 ± 2.3 years old) and the mean age of the child on first medical consult (6.6 ± 2.3 years old). There is a 4.5 ± 1.1 year delay from the onset of symptoms to confirmation of DMD. Delay was brought by multiple factors. Majority opted observation and traditional medicine upon note of the first symptoms and eventually consulted only with worsening. Upon medical consultation, multiple variables also affect arrival to a diagnosis. Facilitating factors include initial physician doing a diagnostic evaluation, physician alleviating caregiver concern and peer pressure. However, impeding factors noted include cultural beliefs, financial constraints, alternative diagnosis by the healthcare provider and lack of available specialists in the region.

Conclusion: This study provided insights on the multiple factors affecting the health seeking behaviors of parents with DMD patients especially in a developing country. Timely diagnosis and intervention produces the best outcome. Yet, to achieve this, the factors identified in this study should be properly addressed.

Keywords: ***Duchenne muscular dystrophy, health-seeking behaviour***

I. INTRODUCTION

Duchenne muscular dystrophy (DMD) is the most commonly encountered pediatric X-linked recessive neuromuscular condition with an estimated prevalence of 1 in 3500 to 5000 male births[1]. Together with Becker muscular dystrophy (BMD), it is in the spectrum of a group of diseases called dystrophinopathies.

Dystrophinopathies are a result of genetic mutations, primarily exon deletions leading to out- or in-frame mutations resulting to absent or a partially functional dystrophin protein, respectively.[2] Due to this mutation, patients with DMD develop a progressive muscle weakening leading to increased disability. Despite the technological advances, the mean age at diagnosis (4.7 years old) has remain unchanged.[3]

Care for patients with DMD will require a multidisciplinary approach involving several specialists. The annual total direct medical cost of this disorder may range from €4420 (Php 24 300) in the Stage 1 to €68 968 (approximately Php 3.8 M) in the Stage 5 of the disease per patient quoted in a study in 2014 (Schreiber-Katz, 2014).[4] In a developing country like the Philippines, aside from the financial burden of this chronic illness, further challenges may include availability of the genetic tests, pharmacologic options and access to multidisciplinary care.

Early diagnosis provides the best opportunity for maximum benefit of the current standard of care and access to novel therapies.[5,6,7] However, certain challenges are present even before the patients arrive to the specialty clinic (e.g. Multidisciplinary Neuromuscular clinic UP-PGH). Hence, the investigators of this study conceptualized describing the parental health-seeking behaviors of the caregivers of boys diagnosed with DMD in order to identify challenges and recommend targeted strategies to facilitate early diagnosis, prompt intervention and optimal outcomes among Filipino DMD patients.

Health-seeking behavior is defined as all the activity undertaken by the individuals who perceive to have a health problem for the purpose of finding an appropriate remedy. The help-seeking behavior model was used to describe and characterize the health-seeking behavior of parents. The application of this theory derived from the behavioral model has provided an analytical assessment for which to examine the factors that may influence DMD diagnostic process. There are four stages of this help seeking behavior - 1) problem recognition, 2) decision to seek help, 3) service selection and 4) service utilization. This model suggests that all families pass through a unidirectional movement along each stages which is controlled by a complex interplay

of factors such as 1) predisposing characteristics, 2) social networks, 3) cultural factors, 4) enabling resources and 5) barriers to care.[9]

II. METHODS

This research was approved by the Review Ethics Board. Utilizing a prospective cross-sectional design, our study investigated the factors affecting the health – seeking behavior of parents of pediatric patients aged less than 19 years old diagnosed with Duchenne muscular dystrophy seen at the Multidisciplinary Neuromuscular Clinic of the University of the Philippines – Philippine General Hospital (UP- PGH) and private clinics of the senior investigator.With informed consent, the parents were requested to participate in a phone or video call interview or fill up an online Google Forms.

The study determined the demographic profiles of both patients and caregivers and identified the facilitating and impeding factors affecting the stages of their health seeking behaviors related to their boys with DMD. Secondary outcomes of the study include determining the association between the first symptom and the confirmation of the disease and the association of the age of patient at first consultation.

Descriptive statistics were used to summarize the demographic and clinical characteristics of the patients. Frequency and proportion were used for categorical variables.

Due to the small sample (n=55), medians and IQRs were used when presenting the continuous exposures (e.g., years of primary caregiver). Kruskal-Wallis test was used to assess the difference of these exposures.

To determine the trends between the select outcomes (e.g., age at first consultation and time interval of first symptom to confirmation) and the select categorical exposures (e.g., educational attainment, family type), cross-tabulationswere done. Chi-Square Tests were performed to formally assess the associations between these groups.

III. RESULTS

*Demographic Profile of the Patients*The study recruited 55 respondents who were parents of children with genetically-confirmed Duchenne Muscular Dystrophy.Majority of the patients belong to the 6 to 10 year old group (52.73%). A good number (40%) are already adolescents. Majority are Roman Catholics. Ninety-five percent are enrolled in a public institution. Two patients attend an individualized education program (See Table 1). Two out of the 10 children in this cohort are out of school.At the time of the study, 47% of the patients are already in the 4th stage of theirchronic illness.*Demographic Profile of the Parents*

About half of the parents belong to the young adult age group while the rest in the middle age group.About half of the family lives in the urban area within50-100 km distance from the multidisciplinary clinic at the UP-PGH. One-fifth of the cohort resides more than >250 km radius.Forty-three percent of parents are college-graduates while a third finished high school. Six out of ten families come from low to lower middle income families with an annual income less than Php 400, 000.

Nuclear family set-up is observed in half of the participants. More than half of the patients with DMD have two to three caregivers at the same time. All of the primary caregivers in this study are parents of the patients. A few being accompanied by grandparents, siblings, aunts, uncles and hired staffas secondary caregivers. The mean duration of care the primary caregivers in this cohort has spent until the diagnosis was made is 7 years. Those with secondary caregivers have a mean duration of 3 years until confirmation of DMD.

Problem Recognition Profile of the Caregivers

The mean age of onset of the symptom of the patients included in this study is at 4 ($\pm$2.3) years old. Symptom onset is between age 3 to 6 years old in nearly half of boys.On the other hand, the mean age on first medical consult is at 6.6 ($\pm$2.31) years old. This is a 2.6 years delay from the mean age of the onset of symptoms. Ninety-five percent of the patients are in the first two stages of the disease on first medical consult. However, three patients were first brought to medical attention when they reached the Stage 3 of the illness.The most common first symptoms noted by their parents are the abnormal gait and the Gower's sign. Together with the motor symptoms, a good number of parents also noted that the patients also had non-motor problems that were also noted as concomitantly occurring initially. These include behavioral issues, cognitive delay, learning and attentional issues and speech delay. Nine out of 10 parents are the key persons to notice the problem. The mean time interval between first notice of these symptoms until confirmation of DMD is 4.5 ($\pm$1.1) years. One third of the patients had their confirmatory test in about a year to less than three years from the onset of signs and symptoms.

Factors Affecting Decision to Seek Help

Among the study participants, 23 (41.82%) opted to observe when they noticed the early signs and symptoms of DMD. Eventually, three-fourths opted to seek medical consult upon observation of symptom progression.Twelve out of 55 parents have waited a minimum interval of less than 1 year before bringing the patient to medical consultation. Caregivers' reasons for the delay of consult included uncertainty or hesitancy (78.18%), financial constraints (41.82%), difficulty finding time to schedule a physician (12.73%), lack of knowledge (7.27%) and preference to go to a traditional healer (1.82%).Around 40% of the patients were initially seen by a general pediatrician and approximately half of the patients were eventually referred to pediatric neurologists.

Factors Affecting Service Selection

Service selection is affected by several factors. In this cohort, half of the first physicians to see their respective patients already initiated a diagnostic evaluation eventually arriving at the proper diagnosis. Thirteen patients had a positive family history of DMD hence the medical diagnostics were facilitated after the first medical consult. It is also noteworthy that several factors have influenced pushing through with doing the medical diagnostics – 1) physician alleviating the caregiver concern, 2) therapists identifying the problem, 3) advice from family, friends and pre-school teacher.

On the other end of the spectrum, several factors have been identified that impeded moving towards the correct diagnosis. A good number of them would have the following reasons: 1) cultural belief that there is no problem, 2) financial constraints, 3) misdiagnosis, and 4) unavailable pediatric neurologist.

Association between the Different Caregiver Characteristics and the Age of the Patient at First Consultation

The different caregiver characteristics were evaluated for association with the age of the patient on first consultation and the time interval from the first symptom to confirmation.Considering the years of service of the primary caregiver, those who had older ages at first consultation tend to have caregivers with longer service years. Patients aged 10 years and above have caregivers who have median service years of about 15 years. Meanwhile, patients who are less than 3 years have caregivers with median service years of 2 years. There is sufficient evidence to say that years of service of a primary caregiver is varying based on the age of the patient at first consultation ($p<0.001$). (See Table 4)Patients who are aged 3 to less than 6 and 6 to less than 10 years old tend to have secondary caregivers whose years of service are the longest. These two groups had median service years of 2.75 and 2 years, respectively. This is compared to the youngest age group which had a median of 1 year and the oldest age group who had a median of 1.75 years. (See Table 5).

However, there is no significant association with the age of the patient at first consultation with these caregiver characteristics: 1) number of caregivers, 2) age of the caregiver, 3) educational attainment of the caregiver, 4) annual total family income, 5) family type, 6) residential area and 7) distance of the patient's home from the hospital. No particular trend in the distribution of the age was also observed. (See Table 6).

There is also no significant association with the time interval on the notice of the first symptoms and the confirmation of the diagnosis with the different caregiver characteristics (See Table 7).

IV. DISCUSSION

Boys with DMD typically present at two to five years old with abnormal gait, often as toe walking and in some as delayed gait onset.[9][10] In our study, the mean age when the caregivers first note of symptoms is at 4 (SD$\pm$ 2.3) years old. This finding is consistent with our cohort where the most commonly noted initial presentation is abnormal gait. The other observed signs and symptoms include Gower's sign, gross motor delay, calf pseudohypertrophy, inability to jump, decreased endurance, decreased head control when pulled to sit, difficulty climbing stairs, flat feet, frequent falling or clumsiness, hypotonia, inability to keep up with peers, loss of motor skills, muscle pain or cramping, toe walking and difficulty running or climbing. Non-motor symptoms have also been described and include behavioral issues, cognitive delay, failure to thrive or poor weight gain, learning and attentional issues and speech delay.[3][11] These were also noted in our patients (See Table 3).

The natural history of DMD has been described to be composed of 5 stages – 1) at diagnosis, 2) early ambulatory, 3) late ambulatory, 4) early non-ambulatory and 5) late non-ambulatory. Upon note of suggestive symptoms, a prompt referral to a child neurologist or a neuromuscular specialist should be done. A markedly elevated serum CK increases the index of suspicion. Pre-genetic counseling[5] before confirmatory testing with multiplex ligation-dependent probe amplification (MLPA)[12] or comparative genomic hybridization assay[13] should be done.

In our study, the mean age of confirmation of diagnosis is 7.4 ($\pm$ 2.7) years old. There is a 3 years gap from the time the symptoms were first noted to the confirmation of the diagnosis. Moreover, almost half of the patients are in their stage 4 of the illness wheelchair-bound.

At the time they get the proper medical attention for DMD, the physician and the caregiver are now faced with improving the quality of life, prognosis and survival. Other complications are expected at this point - respiratory insufficiency, scoliosis, and secondary cardiomyopathy.

At present, the overall mean for respiratory death is 17.7 years for those without ventilatory support. This is extended to 27.9 years for patients with ventilatory support. However, case reports of survival up to the third decade of life is possible but this is uncommon.[14] About 85-90% develop dilated cardiomyopathy at 18 years of age, low levels of pulmonary function and skeletal-muscle impairment making them poor candidates for heart transplant.[15,16] This progression highlights the cost of illness leading to a 16-fold increase of expenses from direct and indirect costs.[17]

The mainstay of treatment includes physiotherapy and glucocorticoid administration yielding the best outcome if started at an early age. Prednisone is often prescribed however some experts routinely use deflazacort for DMD owing for its more favorable side effect profile compared with prednisone particularly weight gain.[18] Benefits for both types of steroid include prolonged ambulation, preserved cardiac, respiratory and upper limb function and delay in the need of scoliosis surgery. Mutation-specific drugs (e.g. ataluren,[19] eteplirsen[20] and viltolarsen[21]) have been studied and showed proven efficacy.

Care for patients with DMD will require a multidisciplinary approach involving specialists from the fields of pediatric neurology, neuromuscular, clinical genetics, cardiology, pulmonology, rehabilitation medicine, orthopedics, gastroenterology, nutrition and endocrinology.[5,6,7].

To achieve these benefits, early diagnosis is essential. In a developing country like the Philippines, certain challenges are present even before these patients arrive to the proper medical specialists. From the end of the caregiver, multiple aspects affect early and prompt diagnosis.

The first action taken by the caregivers in our study upon note of the symptoms was not an immediate medical attention. Yet the majority opted for observation and traditional medicine. Only 2 out of 10 of the caregivers in the study immediately sought medical help upon notice of the symptoms. Despite regulation for traditional medicines in the Philippines, there are no approved therapeutic options available for this disease.[22] Yet, in our study, a good number of caregivers would still choose these as the first step in addressing a noted problem with their children. We also noted that around 40% of caregivers chose to initially observe their patient before taking other actions. Four of 10 caregivers in the cohort eventually sought medical help with

progression of the symptoms. However, it is noteworthy that a few are already familiar with norms of child development and waited for their children to improve, some came across multiple media platforms (e.g. internet, television, prints), have a positive family history, had an incidental finding of markedly elevated serum creatine kinase and others as suggested by relatives.

Eighty percent consulted at least a year after the first symptom. Reasons like hesitancy, financial constraints, difficulty to schedule a physician consultation, distance of the nearby health facility and lack of knowledge were among their responses.

Upon initial consultation, only 10% get to see a pediatric neurologist and about 50% of the DMD patients were evaluated by a neurologist on their second consultation. Most of the caregivers originally sought consultation with a general pediatrician before being referred to a pediatric neurologist. Other specialists that were sought in the cohort include an orthopedic surgeon, a physiatrist and a general physician. The subsequent increase of pediatric neurologists seeing the patient reflects knowledge of the first physician that the patient has a neurologic problem.

Despite having a correct clinical impression or a differential diagnosis that could have arrived at the proper diagnosis, several factors would influence this.

Upon the first medical consultation, a diagnostic process is initiated by the physician in half of the cohorts. However, around 25% of the physicians started the diagnostic process only to alleviate the concern of the caregiver. Other facilitating factors include a positive family history, parents demanded the action, and the physiatrists were the ones who recognized the symptoms.

In a study by Daak-Hirsch et. al. the effect of a positive family history for DMD has delayed the process of seeking help due to parent's readiness to face the diagnosis while others perceived it as stigma.[23]

On the other hand, around 25% of the first physicians who saw the patient were reported to initially have an alternative diagnosis. Only 5% of the first physician who saw the patient took no action. However, several other reasons that impeded facilitating the medical service include parents not following through the referral done, unavailable pediatric neurologist, cultural belief that there is no problem, financial constraints and a pregnant mother.

V. CONCLUSION & RECOMMENDATIONS

Our study provided insights as to how the health seeking behavior of caregivers in a developing country and how it has unfolded the delay in the diagnosis. In the problem-recognition stage, much of the delay is contributed by observing the patient which has extended to more than a year. This is influenced by cultural beliefs that traditional medicine is an option in treating genetic diseases such as DMD. As the disease progressed to a more serious stage, this now became the catalyst to seek medical consultation. This information should ignite debate among administrators, DMD advocates, scientists, and healthcare providers to explore other possibilities for early diagnosis of DMD such as Newborn screening and possibly genetic testing for suspected female carriers. However, there is still no equivalent for a well-informed community as the primary intervention for DMD.

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APPENDICES

Appendix A: Tables

Table 1: Demographic Profile of Patients

Characteristics			%
Age *(n = 55)*	0 to less than 3 years old	0	0
	3 to less than 6 years old	4	7.27%
	6 to less than 10 years old	29	52.73%
	10 to less than 19 years old	22	40%
Ethnicity *(n = 55)*	Bisaya	5	9.09%
	Bicolano	6	10.91%
	Ilocano	3	5.45%
	Ibanag	1	1.82%
	Tagalog	40	72.73%
Religion *(n = 55)*	Roman Catholic	41	74.55%
	Christian	9	16.36%
	Iglesiani Cristo	2	3.64%
	Jehovah's Witness	1	1.82%
	Muslim	1	1.82%
	Protestant	1	1.82%
Current Form of Education *(n = 55)*	School	0	0
	Homeschooling	41	74.55%
	Individualized Education Program	2	3.64%
	None (out of school)	12	21.82%
Type of school *(n = 43)*	Private	2	4.65%
	Public	41	95.35%
Current Stage of Illness *(n = 55)*	Stage 1	2	3.64%
	Stage 2	14	25.45%
	Stage 3	13	23.64%
	Stage 4	26	47.27%

	Stage 5	0	0

Table 2. Characteristics of caregivers			
		n	%
Number of caregivers the patient had from birth to time of diagnosis	1	23	41.82%
	2 to 3	32	58.18%
	4 to 5	0	58.18%
	More than 5	0	0
Relationship of caregivers to the patient	Parent	55	100%
	Siblings	1	1.82%
	Grandparents	3	5.45%
	Aunt and Uncle	1	1.82%
	Other relatives	0	0
	Not related to patient	1	1.82%
	Multiple caregivers	0	0
Mean Duration (years) per caregiver (*For patients with >1 caregiver)*	Primary Caregiver	7.44	
	Secondary Caregiver	2.94	
Demographic Profile of Caregiver			
Characteristics			
Number of caregivers	1	23	41.82%
	2 to 3	32	58.18%
	4 to 5	0	0
Age	Young adult (20 less than 40 years old)	28	50.91%
	Middle age (40 to less than 60 years old)	27	49.09%
	Old age (60 years old and above)	0	0
Type of Residential Area	Urban	30	54.55%
	Rural	25	45.45%
Distance away from hospital	<20 km	5	9.09%
	20 – 50 km	5	9.09%
	50 – 100 km	25	45.45%
	100 – 200 km	8	14.55%
	>250 km	12	21.82%
Educational Attainment	Elementary undergraduate	0	0
	Elementary graduate	0	0
	Highschool undergraduate	2	3.64%
	Highschool graduate	19	34.55%
	College undergraduate	8	14.55%
	College graduate	24	43.64%
	Vocational course	2	3.64%
Annual Total Family Income	<Php 250 000 (20 000/month) (Low income)	14	25.45%
	Php 250 000 – Php 400 000 (20 000 – 33 000) (Lower middle income)	21	38.18%
	Php 400 000 – Php 800 000 (33K to 66K) (Middle income)	7	12.73%
	Php 800 000 – Php 2 000 000 (66K – 166K) (Upper Middle income)	13	23.64%
Family type	Nuclear	29	52.73%
	Solo Parenting	5	9.09%
	Extended	21	38.18%

Table 3: Problem Recognition			
Factor		n	%
Age of patient at the onset of	0 to less than 3 years old	15	27.27%

symptom	3 to less than 6 years old	27	49.09%
	6 to less than 10 years old	13	23.64%
Age of patient on first medical consult	0 to less than 3 years old	4	7.27%
	3 to less than 6 years old	9	16.36%
	6 to less than 10 years old	40	72.73%
	>10 years old	2	3.64%
Stage of Illness on 1st medical consult:	Stage 1	30	54.55%
	Stage 2	22	40%
	Stage 3	3	5.45%
	Stage 4	0	0
	Stage 5	0	0
First symptom noted	MOTOR		
	Abnormal gait	46	83.64%
	Calf pseudohypertrophy	10	18.18%
	Decreased endurance	4	7.27%
	Difficulty climbing stairs	7	12.73%
	Frequent falling or clumsiness	21	38.18%
	Gower's sign on rising from floor	32	58.18%
	Gross motor delay	4	7.27%
	Hypotonia	4	7.27%
	Inability to keep up with peers	1	1.82%
	Loss of motor skills	1	1.82%
	Muscle pain or cramping	2	3.64%
	Toe walking	25	45.45%
	Difficulty running or climbing	4	7.27%
	NON-MOTOR		
	Behavioral issues	17	30.91%
	Cognitive delay	7	12.73%
	Poor weight gain	12	21.82%
	Learning and attentional issues	7	12.73%
	Speech delay or articulation difficulties	2	3.64%
Person who first noticed the problem	Parents / Caregivers	49	89.09%
	Relatives	6	10.91%
	Friends	1	14.29%
	Non-physician healthcare professional	1	5.56%
	Others, please specify	0	0
Time interval between note of symptoms and confirmation of Duchenne muscular dystrophy	Less than 1 year	0	0
	One to less than 6 months	1	1.82%
	Six to less than 12 months	9	16.36%
	One year to less than three years	17	30.91%
	Three to less than six years	16	29.09%
	More than 6 years old	12	21.82%

Table 4. Differences among Caregiver Characteristics (Continuous) based on Age of Patient at First Consultation

	Age of patient at first consultation								
	0 to < 3 years		3 to < 6 years		6 to < 10 years		10 years and above		
Characteristics	Median	IQR	Median	IQR	Median	IQR	Median	IQR	P-value
Years of Caregiver 1/primary	2	2 - 3.5	5	4 - 6	8	7 - 9	14.5	12 - 17	< 0.001
Years of Caregiver 2	1	1 - 2	2.75	1.5 - 3.5	2	2 - 5	1.75	1.5 - 2	0.143
Age of Caregiver	39.5	33.5 - 43.5	35	31 - 40	40	35.5 - 44.5	36.5	36 - 37	0.249

Table 5. Differences among Caregiver Characteristics (Continuous) based on Time interval of first symptom to confirmation

Time interval of first symptom to confirmation				
1 month to < 6 months	6 months to < 12 months	1 year to < 3 years	3 years to < 6 years	> 6 years

Characteristics	Median	IQR	Median	IQR	Median	IQR	Median	IQR	Median	IQR	P-value
Years of Caregiver 1/primary	2	2 - 2	6	5 - 9	7	5 - 8	8	8 - 7.5	9	8.5 - 12	0.004
Years of Caregiver 2	1	1 - 1	2.5	2 - 3	2	1 - 4	2	2 - 2	3	2 - 4	0.599
Age of Caregiver	44	44 - 44	36	35 - 43	38	35 - 42	41	41 - 32.5	38.5	35.5 - 42	0.826

Table 7. Cross-Tabulation of Select Exposures Based on Time interval of first symptom to confirmation

		Time interval of first symptom to confirmation										
		1 month to < 6 months		6 months to < 12 months		1 year to < 3 years		3 years to < 6 years		>= 6 years		
Characteristics	Categories	Count	%	Count	%	Count	%	Count	%	Count	%	P-value
Number of caregivers the patient had from birth to time of diagnosis	1	0	0.00%	5	55.60%	6	35.30%	8	50.00%	5	41.70%	0.801
	2 to 3	1	100.00%	4	44.40%	11	64.70%	8	50.00%	7	58.30%	
Type of Caregiver	As the solo caregiver	0	0.00%	4	44.40%	6	35.30%	8	50.00%	5	41.70%	0.922
	As the primary in multiple	1	100.00%	5	55.60%	11	64.70%	8	50.00%	7	58.30%	
Relationship of caregivers to the patient (PRIMARY)	Parent	1	100.00%	9	100.00%	17	100.00%	16	100.00%	12	100.00%	
Relationship of caregivers to the patient (SECONDARY)	Parent	1	100.00%	4	80.00%	8	72.70%	7	87.50%	6	85.70%	0.734
	Siblings	0	0.00%	0	0.00%	1	9.10%	0	0.00%	0	0.00%	
	Grandparents	0	0.00%	0	0.00%	2	18.20%	0	0.00%	1	14.30%	
	Aunt and Uncle	0	0.00%	0	0.00%	0	0.00%	1	12.50%	0	0.00%	
	Other relatives	0	0.00%	1	20.00%	0	0.00%	0	0.00%	0	0.00%	
Number of caregivers	1 to 2	1	100.00%	9	100.00%	17	100.00%	16	100.00%	12	100.00%	
Age group of caregiver	Young Adult (20 to < 40 years)	0	0.00%	5	55.60%	10	58.80%	5	31.30%	8	66.70%	0.251
	Middle age (40 to < 60 years)	1	100.00%	4	44.40%	7	41.20%	11	68.80%	4	33.30%	
Type of Residential Area	Urban	0	0.00%	7	77.80%	7	41.20%	8	50.00%	8	66.70%	0.264
	Rural	1	100.00%	2	22.20%	10	58.80%	8	50.00%	4	33.30%	
Distance away from hospital	< 20 KM	0	0.00%	0	0.00%	2	11.80%	2	12.50%	1	8.30%	0.986
	20-50 KM	0	0.00%	1	11.10%	1	5.90%	2	12.50%	1	8.30%	
	50-100 KM	0	0.00%	5	55.60%	8	47.10%	6	37.50%	6	50.00%	
	100-200 KM	1	100.00%	1	11.10%	2	11.80%	3	18.80%	1	8.30%	

	> 200 KM	0	0.00%	2	22.20%	4	23.50%	3	18.80%	3	25.00%	
Educational Attainment	Highschool Undergraduate	0	0.00%	0	0.00%	1	5.90%	0	0.00%	1	8.30%	0.791
	Highschool Graduate	0	0.00%	3	33.30%	8	47.10%	5	31.30%	3	25.00%	
	College Undergraduate	0	0.00%	0	0.00%	2	11.80%	3	18.80%	3	25.00%	
	College Graduate	1	100.00%	6	66.70%	6	35.30%	7	43.80%	4	33.30%	
	Vocational Course	0	0.00%	0	0.00%	0	0.00%	1	6.30%	1	8.30%	
Annual Total Family Income	< Php 250,000	0	0.00%	2	22.20%	7	41.20%	4	25.00%	1	8.30%	0.332
	Php 250,000 - Php 400,000	1	100.00%	1	11.10%	5	29.40%	7	43.80%	7	58.30%	
	Php 400,001 - Php 800,000	0	0.00%	2	22.20%	3	17.60%	1	6.30%	1	8.30%	
	Php 801,000 and above	0	0.00%	4	44.40%	2	11.80%	4	25.00%	3	25.00%	
Family type	Nuclear	0	0.00%	5	55.60%	7	41.20%	11	68.80%	6	50.00%	0.341
	Solo parenting	1	100.00%	1	11.10%	2	11.80%	0	0.00%	1	8.30%	
	Extended	0	0.00%	3	33.30%	8	47.10%	5	31.30%	5	41.70%	

Table 8. Age of patient at first consultation

		0 to < 3 years	3 to < 6 years	6 to < 10 years	10 years and above
Number of caregivers the patient had from birth to time of diagnosis	1	1	1	22	0
	2 to 3	3	8	18	2
Relationship of caregivers to the patient (PRIMARY)	Parent	4	9	40	2
		0 to < 3 years	3 to < 6 years	6 to < 10 years	10 years and above
Relationship of caregivers to the patient (SECONDARY)	Parent	3	8	14	1
	Siblings	0	0	1	0
	Grandparents	0	0	2	1
	Aunt and Uncle	0	0	1	0
	Other relatives	0	0	1	0
		0 to < 3 years	3 to < 6 years	6 to < 10 years	10 years and above
Type of Caregiver	As the solo caregiver	1	1	21	0
	As the primary in multiple	3	8	19	2
		0 to < 3 years	3 to < 6 years	6 to < 10 years	10 years and above
Number of caregivers	1 to 2	4	9	40	2
	3 to 5	0	0	0	0
	> 5	0	0	0	0
		0 to < 3 years	3 to < 6 years	6 to < 10 years	10 years and above
Age group of caregiver	Young Adult (20 to < 40	2	6	18	2

	years)				
	Middle age (40 to < 60 years)	2	3	22	0
		0 to < 3 years	3 to < 6 years	6 to < 10 years	10 years and above
Type of Residential Area	Urban	2	6	20	2
	Rural	2	3	20	0
		0 to < 3 years	3 to < 6 years	6 to < 10 years	10 years and above
Distance away from hospital	< 20 KM	0	2	3	0
	20-50 KM	0	2	3	0
	50-100 KM	2	2	19	2
	100-200 KM	2	2	4	0
	> 200 KM	0	1	11	0
		0 to < 3 years	3 to < 6 years	6 to < 10 years	10 years and above
Educational Attainment	Highschool Undergraduate	0	0	1	1
	Highschool Graduate	1	3	15	0
	College Undergraduate	0	0	7	1
	College Graduate	3	6	15	0
	Vocational Course	0	0	2	0
		0 to < 3 years	3 to < 6 years	6 to < 10 years	10 years and above
Annual Total Family Income	< Php 250,000	1	2	10	1
	Php 250,000 - Php 400,000	1	1	18	1
	Php 400,001 - Php 800,000	1	1	5	0
	Php 801,000 and above	1	5	7	0
		0 to < 3 years	3 to < 6 years	6 to < 10 years	10 years and above
Family type	Nuclear	3	5	21	0
	Solo parenting	1	1	3	0
	Extended	0	3	16	2

Table 9: Time interval of first symptom to confirmation

		1 month to < 6 months	6 months to < 12 months	1 year to < 3 years	3 years to < 6 years	> 6 years
Number of caregivers the patient had from birth to time of diagnosis	1	0	5	6	8	5
	2 to 3	1	4	11	8	7
		1 month to < 6 months	6 months to < 12 months	1 year to < 3 years	3 years to < 6 years	> 6 years
Relationship of caregivers to the patient (PRIMARY)	Parent	1	9	17	16	12
		1 month to < 6 months	6 months to < 12 months	1 year to < 3 years	3 years to < 6 years	> 6 years
Relationship of caregivers to the patient (SECONDARY)	Parent	1	4	8	7	6
	Siblings	0	0	1	0	0
	Grandparents	0	0	2	0	1
	Aunt and Uncle	0	0	0	1	0
	Other relatives	0	1	0	0	0
		1 month to < 6 months	6 months to < 12 months	1 year to < 3 years	3 years to < 6 years	> 6 years

Type of Caregiver	As the solo caregiver	0	4	6	8	5
	As the primary in multiple	1	5	11	8	7
		1 month to < 6 months	6 months to < 12 months	1 year to < 3 years	3 years to < 6 years	> 6 years
Number of caregivers	1 to 2	1	9	17	16	12
	3 to 5	0	0	0	0	0
	> 5	0	0	0	0	0
		1 month to < 6 months	6 months to < 12 months	1 year to < 3 years	3 years to < 6 years	> 6 years
Age group of caregiver	Young Adult (20 to < 40 years)	0	5	10	5	8
	Middle age (40 to < 60 years)	1	4	7	11	4
		1 month to < 6 months	6 months to < 12 months	1 year to < 3 years	3 years to < 6 years	> 6 years
Type of Residential Area	Urban	0	7	7	8	8
	Rural	1	2	10	8	4
		1 month to < 6 months	6 months to < 12 months	1 year to < 3 years	3 years to < 6 years	> 6 years
Distance away from hospital	< 20 KM	0	0	2	2	1
	20-50 KM	0	1	1	2	1
	50-100 KM	0	5	8	6	6
	100-200 KM	1	1	2	3	1
	> 200 KM	0	2	4	3	3
		1 month to < 6 months	6 months to < 12 months	1 year to < 3 years	3 years to < 6 years	> 6 years
Educational Attainment	Highschool Undergraduate	0	0	1	0	1
	Highschool Graduate	0	3	8	5	3
	College Undergraduate	0	0	2	3	3
	College Graduate	1	6	6	7	4
	Vocational Course	0	0	0	1	1
		1 month to < 6 months	6 months to < 12 months	1 year to < 3 years	3 years to < 6 years	> 6 years
Annual Total Family Income	< Php 250,000	0	2	7	4	1
	Php 250,000 - Php 400,000	1	1	5	7	7
	Php 400,001 - Php 800,000	0	2	3	1	1
	Php 801,000 and above	0	4	2	4	3
		1 month to < 6 months	6 months to < 12 months	1 year to < 3 years	3 years to < 6 years	> 6 years
Family type	Nuclear	0	5	7	11	6
	Solo parenting	1	1	2	0	1
	Extended	0	3	8	5	5

Appendix B: Graphs

6. Cross-Tabulation of Select Exposures Based on Age of Patient at First Consult

Characteristics	Categories	Age of patient at first consultation								P-value
		0 to < 3 years		3 to < 6 years		6 to < 10 years		10 years and above		
		Count	%	Count	%	Count	%	Count	%	
Number of caregivers the patient had from birth to time of diagnosis	1	1	25.00%	1	11.10%	22	55.00%	0	0.00%	0.032
	2 to 3	3	75.00%	8	88.90%	18	45.00%	2	100.00%	
Type of Caregiver	As the solo caregiver	1	25.00%	1	11.10%	21	52.50%	0	0.00%	0.047
	As the primary in multiple	3	75.00%	8	88.90%	19	47.50%	2	100.00%	
Relationship of caregivers to the patient (PRIMARY)	Parent	4	100.00%	9	100.00%	40	100.00%	2	100.00%	
Relationship of caregivers to the patient (SECONDARY)	Parent	3	100.00%	8	100.00%	14	73.70%	1	50.00%	0.718
	Siblings	0	0.00%	0	0.00%	1	5.30%	0	0.00%	
	Grandparents	0	0.00%	0	0.00%	2	10.50%	1	50.00%	
	Aunt and Uncle	0	0.00%	0	0.00%	1	5.30%	0	0.00%	
	Other relatives	0	0.00%	0	0.00%	1	5.30%	0	0.00%	
Number of caregivers	1 to 2	4	100.00%	9	100.00%	40	100.00%	2	100.00%	
Age group of caregiver	Young Adult (20 to < 40 years)	2	50.00%	6	66.70%	18	45.00%	2	100.00%	0.386
	Middle age (40 to < 60 years)	2	50.00%	3	33.30%	22	55.00%	0	0.00%	
Type of Residential Area	Urban	2	50.00%	6	66.70%	20	50.00%	2	100.00%	0.635
	Rural	2	50.00%	3	33.30%	20	50.00%	0	0.00%	
Distance away from hospital	< 20 KM	0	0.00%	2	22.20%	3	7.50%	0	0.00%	0.262
	20-50 KM	0	0.00%	2	22.20%	3	7.50%	0	0.00%	
	50-100 KM	2	50.00%	2	22.20%	19	47.50%	2	100.00%	
	100-200 KM	2	50.00%	2	22.20%	4	10.00%	0	0.00%	
	> 200 KM	0	0.00%	1	11.10%	11	27.50%	0	0.00%	
Educational Attainment	Highschool Undergraduate	0	0.00%	0	0.00%	1	2.50%	1	50.00%	0.244
	Highschool Graduate	1	25.00%	3	33.30%	15	37.50%	0	0.00%	
	College Undergraduate	0	0.00%	0	0.00%	7	17.50%	1	50.00%	
	College Graduate	3	75.00%	6	66.70%	15	37.50%	0	0.00%	

	Vocational Course	0	0.00 %	0	0.00%	2	5.00%	0	0.00%	
Annual Total Family Income	< Php 250,000	1	25.00 %	2	22.20%	10	25.00%	1	50.00%	0.32
	Php 250,000 - Php 400,000	1	25.00 %	1	11.10%	18	45.00%	1	50.00%	
	Php 400,001 - Php 800,000	1	25.00 %	1	11.10%	5	12.50%	0	0.00%	
	Php 801,000 and above	1	25.00 %	5	55.60%	7	17.50%	0	0.00%	
Family type	Nuclear	3	75.00 %	5	55.60%	21	52.50%	0	0.00%	0.288
	Solo parenting	1	25.00 %	1	11.10%	3	7.50%	0	0.00%	
	Extended	0	0.00 %	3	33.30%	16	40.00%	2	100.00 %	

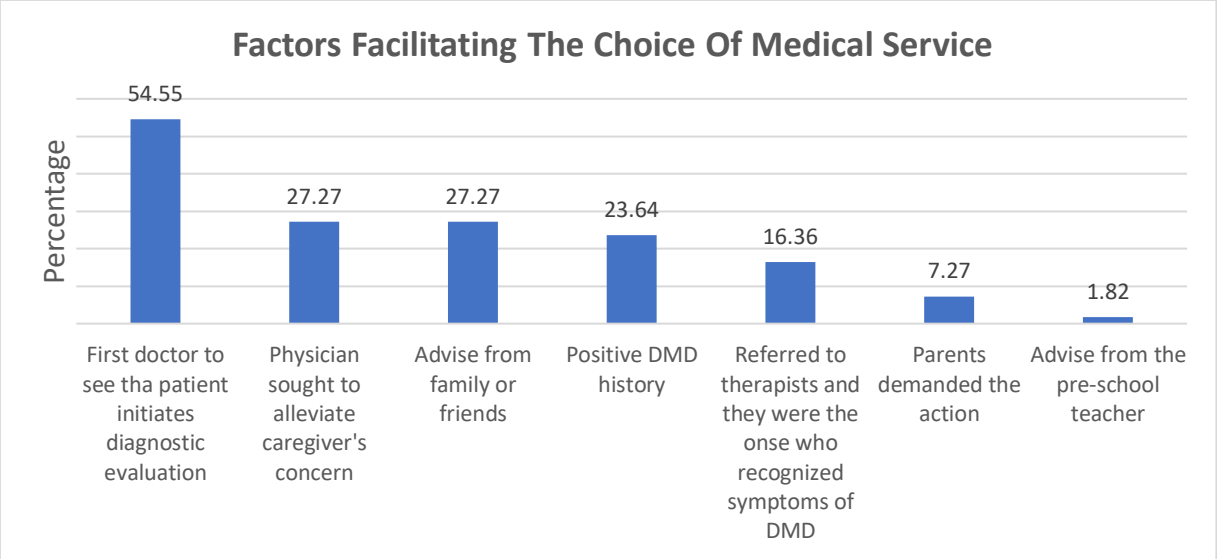

Figure 1: Factors facilitating the choice of medical service.

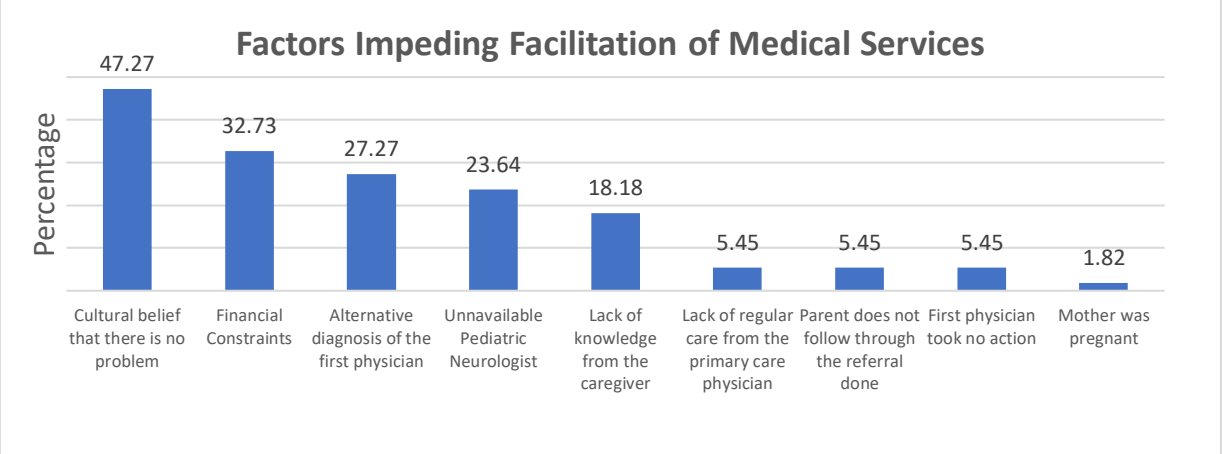

Figure 2: Factors Impeding Facilitation of Medical Services.

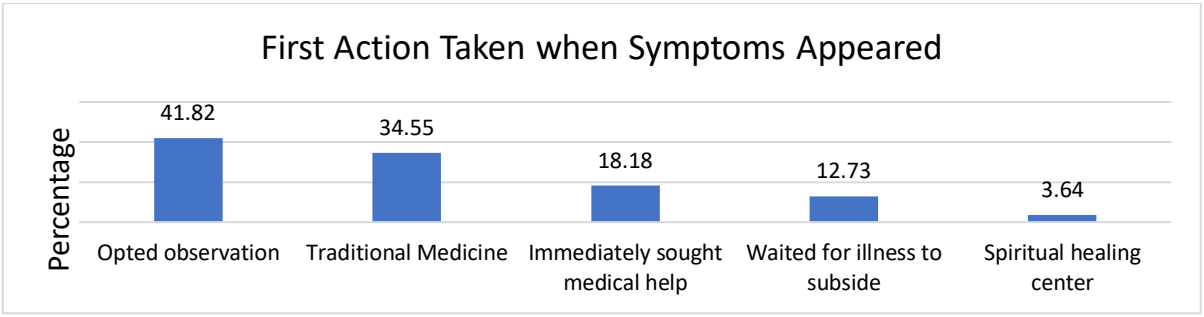

Figure 3: First action taken by the caregiver when symptoms appeared.

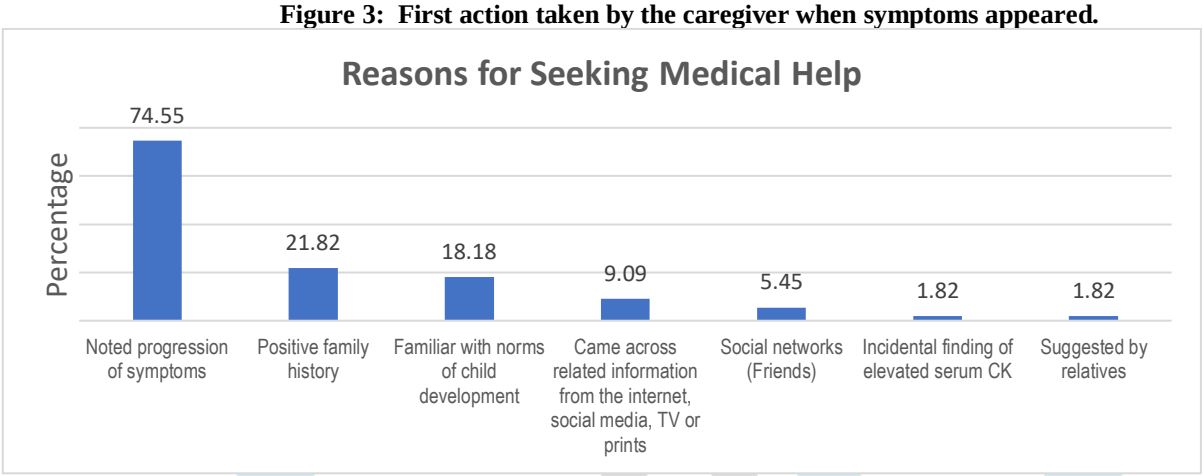

Figure 4: Reasons for seeking medical help.

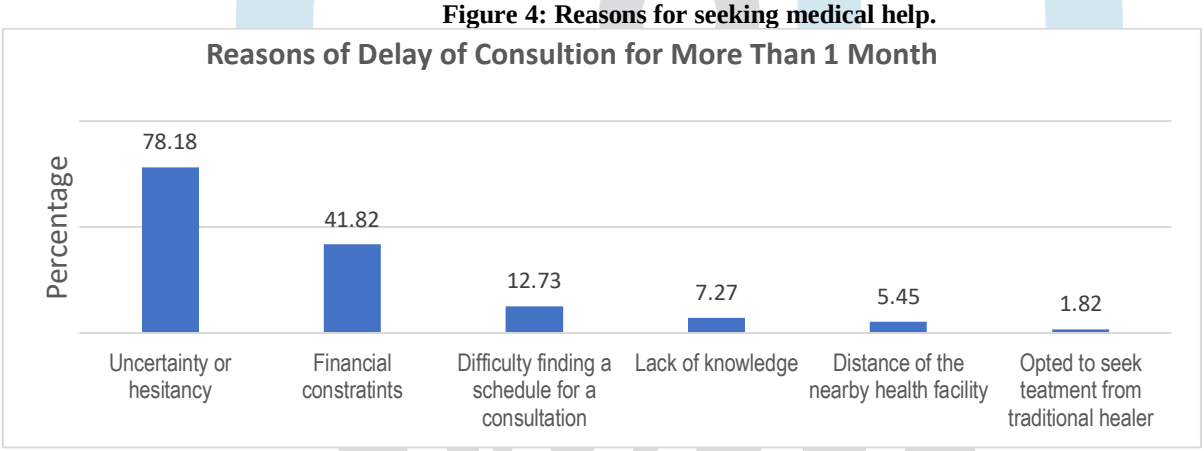

Figure 5: Reasons for delay of consultation for more than 1 month.

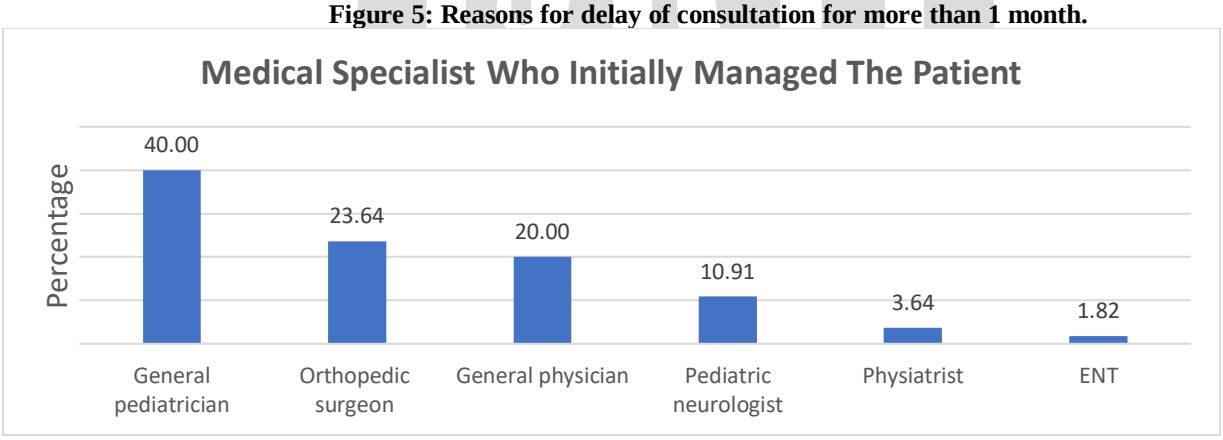

Figure 6: Medical specialist who initially managed the patient.

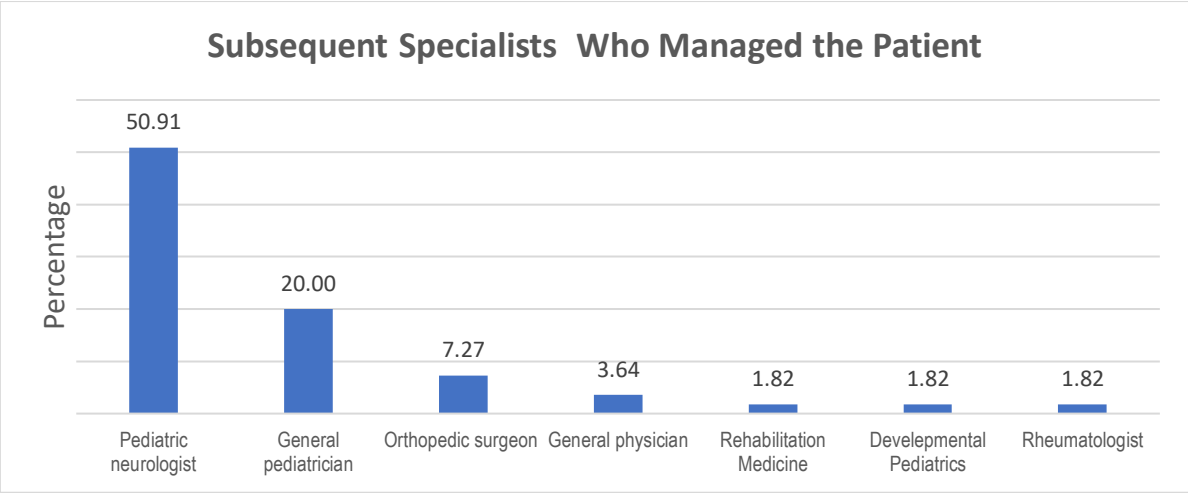

Figure 7: Subsequent specialist who managed the patient if not initially seen by a pediatric neurologist.

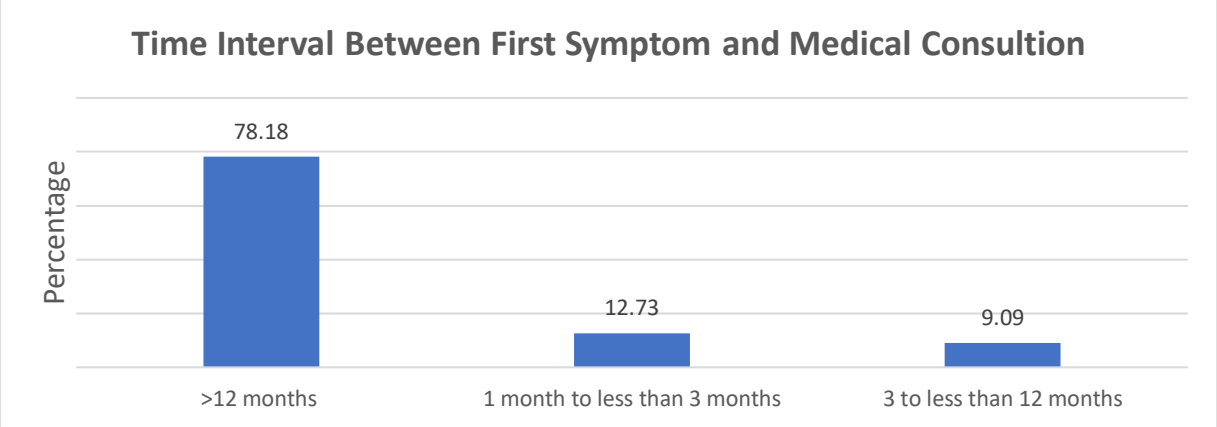

Figure 8: Time interval between first symptom and first medical consult.