

**DEVELOPMENT, VALIDATION AND  
APPLICATION OF QUALITY OF LIFE  
QUESTIONNAIRE IN CHILDREN WITH  
INFANTILE ESOTROPIA AND THEIR  
PROXY/PARENTS**

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PROXY/PARENTS**

by

**WAHEEDA AZWA BINTI HUSSEIN**

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## LIST OF ABBREVIATIONS

HRQoL	Health related quality of life
IXTQ	Intermittent Exotropia Questionnaire
Expert 1	NBH
Expert 2	SI
Expert 3	JCH
Expert 4	JR
Expert 5	SMK
Expert 6	NAW
Expert 7	SA
Expert 8	NK
Expert 9	AR
Expert 10	SAR
Expert 11	NZ
Expert 12	NAAW
Expert 13	NWS
Researcher 1	WAH
ANCOVA	Analysis of covariance

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Appendix XXXIII Soal Selidik Kualiti Kehidupan Kanak-Kanak Juling (Versi Ibu bapa) Proxy/parent Infantile Esotropia Health Related Quality of Life Questionnaire Final Draft

**PEMBINAAN, PENGESAHAN DAN APLIKASI SOAL SELIDIK KUALITI  
HIDUP DALAM KALANGAN KANAK-KANAK INFANTILE ESOTROPIA  
DAN IBU BAPA/PENJAGA MEREKA**

**ABSTRAK**

Kanak-kanak yang menghidap masalah *infantile esotropia* dan ibu bapa/penjaga mereka mengalami pelbagai masalah dalam kehidupan seharian. Namun demikian, tiada instrumen yang wujud untuk mengukur kualiti hidup mereka. Kajian ini bertujuan membina, mengesah dan menilai soal selidik dalam kalangan kanak-kanak *infantile esotropia* dan ibu bapa/penjaga mereka. Kajian ini dijalankan dari Julai 2016 sehingga Jun 2019 di tujuh buah lokasi. Pembinaan soal selidik dihasilkan daripada kajian literatur, kaji selidik, temubual serta perbincangan bersama pakar. Satu kajian pengesahan telah dijalankan ke atas kanak-kanak dan ibu bapa/penjaga mereka diikuti dengan kajian keratan rentas untuk menilai soal selidik tersebut. Hasil kajian dianalisa menggunakan SPSS versi 26.0. Satu soal selidik yang mengandungi tiga versi untuk kanak-kanak berumur 5-8 tahun (2 subtema, 10 item), 9-17 tahun (2 subtema, 10 item) dan ibubapa/penjaga (4 subtema, 25 item) telah dibina. Semua item dalam ketiga-tiga soal selidik mempunyai nilai *content evidence* yang memuaskan (*scale level-content validity index, averaging method* > 0.8), dan nilai *response process evidence* yang baik (*scale-level face validity index, averaging method* > 0.8). Semua soal selidik mempunyai nilai *internal consistency* yang tinggi. (Cronbach's alpha: 0.84-0.87 (5-8 tahun), 0.83-0.86 (9-17 tahun), dan 0.85-0.89 (ibu bapa/penjaga); nilai *intraclass correlation coefficients* yang baik ( $r=0.497$ ,  $p<0.01$  (5-8 tahun),  $r=0.728$ ,  $p<0.01$  (9-17 tahun) dan  $r=0.746$ ,  $p<0.01$ (ibu bapa/penjaga); serta korelasi signifikan dengan IXTQ ( $r=0.780$ ,  $p<0.01$  (5-8 tahun),  $r=0.602$ ,  $p<0.01$  (9-17 tahun)

dan  $r=0.444$ ,  $p<0.01$  (ibu bapa/penjaga). Perbezaan statistik signifikan didapati di antara skor purata kanak-kanak *infantile esotropia* dan ibu bapa/penjaga mereka, dibandingkan dengan kanak-kanak normal dan ibubapa/penjaga mereka ( $p < 0.001$ ). Tiada perkaitan yang signifikan didapati antara skor purata kanak-kanak *infantile esotropia* dengan faktor jantina, sudut deviasi dan *stereopsis* ( $p>0.05$ ). Pembedahan juling telah meningkatkan skor purata kualiti hidup kanak-kanak dengan signifikan (68.00 pra pembedahan kepada 89.36 selepas pembedahan ( $p < 0.001$ ) untuk kumpulan 5-8 tahun; 78.07 pra pembedahan kepada 90.21 selepas pembedahan ( $p < 0.001$ ) untuk kumpulan 9-17 tahun). Soal selidik dalam Bahasa Melayu yang sah dan dipercayai telah dibina untuk mengukur kualiti hidup kanak-kanak *infantile esotropia* dan ibu bapa/penjaga mereka. Kualiti hidup mereka adalah rendah berbanding kanak-kanak normal dan ibu bapa/penjaga mereka. Pembedahan juling didapati berjaya meningkatkan kualiti hidup kanak-kanak *infantile esotropia*.

**DEVELOPMENT, VALIDATION AND APPLICATION OF QUALITY OF  
LIFE QUESTIONNAIRE IN CHILDREN WITH INFANTILE ESOTROPIA  
AND THEIR PROXY/PARENTS**

**ABSTRACT**

Infantile esotropia is a common strabismus problem in Malaysia. Children with infantile esotropia and their proxy/parents suffered from various problems. However, there is no specific tool to measure their quality of life in our native language. The objective of this study is to develop, validate and evaluate new quality of life questionnaire in children with infantile esotropia and their proxy/parents. The study was conducted from 2016 to 2019. The questionnaire was developed based on literature review, survey, interview and brainstorming with experts. A validation study was conducted on the children and their proxy/parents followed by a cross-sectional study to test the questionnaire. The health related quality of life scores were compared to normal children and their proxy/parents, and between pre-operative and post-operative surgery. A new 3-version questionnaire in Bahasa Malaysia for child 5-8 years (2 subthemes, 10 items), child 9-17 years (2 subthemes, 10 items), and their proxy/parents (4 subthemes, 25 items) was developed. All items in the three version questionnaires had satisfactory content evidence (scale level-content validity index, averaging method  $> 0.8$ ), and good response process evidence (scale-level face validity index, averaging method  $> 0.8$ ). All questionnaires were found to have high internal consistency (Cronbach's alpha: 0.84-0.87 (5-8 years old), 0.83-0.86 (9-17 years old), and 0.85-0.89 (proxy/parent); acceptable intraclass correlation coefficients ( $r=0.497$ ,  $p<0.01$  (5-8 years old),  $r=0.728$ ,  $p<0.01$  (9-17 years old) and  $r=0.746$ ,  $p<0.01$ (proxy/parents); and significant correlations with the IXTQ ( $r=0.780$ ,  $p<0.01$

(5-8 years old),  $r=0.602$ ,  $p<0.01$  (9-17 years old) and  $r=0.444$ ,  $p<0.01$  (proxy/parent). A statistical significant difference was found between the mean total scores in children with infantile esotropia and their proxy/parents, and normal children and their proxy/parents ( $p < 0.001$ ). No significant association was found between the mean total scores of the children and all factors (gender, angle of deviation and stereopsis,  $p>0.05$ ). Strabismus surgery had significantly improved the HRQoL scores (68.00 preoperatively to 89.36 postoperatively ( $p < 0.001$ ) for 5-8 years old group; and 78.07 preoperatively to 90.21 postoperatively ( $p < 0.001$ ) for 9-17 years old group). A new valid and reliable 3-version HRQoL questionnaire for children with infantile esotropia and their proxy/parents, consists of Child (5-8 years old), Child (9-17 years old) and Proxy/parent questionnaires has been developed. The children and their proxy/parents had lower HRQoL compared to normal children and their proxy/parents. Strabismus surgery successfully improved the HRQoL of the children.

# CHAPTER 1

## INTRODUCTION

### 1.1 Introduction

According to the recent World Report on Vision 2019 (World Health Organization, 2019), vision impairment influenced the children's quality of life significantly. Young children with early onset severe impairment can suffer from delayed motor, language, emotional, social and cognitive development (Warren, 1994), which may affect their entire life. School-age children with vision impairment may also have a lower academic achievement and self-esteem compared to their normal friends (Chanfreau and Cebulla, 2009; Toledo et al., 2010; Augestad, 2017).

Children with strabismus are known to have psychological and functional problems in their daily life (Mojon-Azzi et al., 2011a; Lin et al., 2014; Ribeiro et al., 2014). These problems may lead to low academic performance in school (Reed et al., 2004). Minto and Ho (2017) reported strong relationship between children's visual health, and the learning quality and achievement at school. They also experienced amblyopia leading to vision impairment (Magli et al., 2014). As a result, these problems will affect the children's future quality of life and economic productivity.

Interestingly, parents of children with strabismus also reported having problems in their daily life along with their children's problems. In a psychosocial study conducted on mothers of children with strabismus in Turkey, they were likely to be more depressed and had an unsupportive relationship with their children (Akay et al., 2005). The study reported that the depression levels of the mothers might be

increased because of their children's condition. Besides, the child's depression and anxiety might also be the factors influencing the same characteristics in the mothers.

Parents also play a role as a proxy for problems encountered by their children. In a study on parental understanding of strabismus, the parents of children who underwent strabismus surgery thought that their child's self-esteem was affected by strabismus (Eustis and Smith, 1987). Parents believed that the most critical problem of their child was the cosmetic effect of strabismus. In another parent-reported symptoms study, Chung et al. (2012) reported that intermittent exotropia was a contributing factor for attention deficit hyperactivity disorder in children and the symptoms improved after strabismus surgery. Therefore, the information from the parents is crucial to help in the clinical management of strabismus.

Currently, the indication of strabismus surgery is according to the clinical evaluation including the angle of deviation and stereopsis (Kim et al., 2015). However, Asadi-Lari et al. (2004) suggested health related quality of life (HRQoL) as an important tool to be included in the clinical assessment. Quality of life can act as another clinical evaluation to measure the effectiveness of the surgery. By evaluating the quality of life, clinicians can be assured that the health need of a patient is met and satisfactory care process has been performed.

Patient's perception can give meaningful information to help in the management of strabismus. The functional and physical concern obtained from the children and their proxy/parents can assist clinicians in strabismus management and decision making. Clinicians including the ophthalmologist, optometrist and orthoptist

can identify the actual problem of strabismus. As a result, the management of strabismus can be improved and the burden of strabismus on the patients' quality of life can be minimized.

The use of questionnaire in the field of optometrist and ophthalmologist is well-known including the Convergent Insufficiency Symptom Survey (Borsting et al., 1999), The Pediatric Quality of Life Inventory (Varni et al., 2001) and National Eye Institute Visual Function Questionnaire (NEI VFQ-25) (Mangione et al., 2001). It is a hope that the newly developed questionnaire will give an impact for the better management of the patient with strabismus. The implementation of the new questionnaire in the optometry and ophthalmology clinics will benefit the patients in measuring their satisfaction and outcome of treatment.

## **1.2 Problem statement**

Esotropia is a common strabismus problem among children attended Ophthalmology Clinics in Malaysia. In a hospital based study in 2016, 1025 cases of esotropia from 3227 strabismus cases were recorded from all hospitals with Ophthalmology services in the Ministry of Health Malaysia (The Cencus Committee of Optometry Profesion Ministry of Health Malaysia, 2017). In addition, 41.8% patients were diagnosed with esotropia, and 28.6% with infantile esotropia in a hospital based study performed at the Hospital Universiti Sains Malaysia, Kubang Kerian, Kelantan (Waheeda-Azwa et al., 2020).

A population based study among pre-school children carried out in Sitiawan District, Perak, Malaysia reported 1 % prevalence of strabismus (Hussin et al., 2009). Whereas a study among primary school children aged 7-12 years old conducted in Petaling Jaya, Selangor, documented a higher prevalence of 2.5% (Reddy and Hassan 2006).

Researcher found that children with esotropia were reported to have more learning difficulties (Uretmen et al., 2003) and perceived as more distressing compared to exotropia (Mojon-Azzi et al., 2011b). Children also showed negative behaviors towards esotropia compared to exotropia (Paysse et al., 2001).

The problems encountered by children with infantile esotropia and their parents will impact their HRQoL. Nowadays, the measurement of HRQoL is gradually becoming important in the management of strabismus and clinical outcome measures. Measuring effects on HRQoL may help to determine the indications for intervention along with clinical assessment. Strabismus is well-known to affect children's HRQoL, but the current clinical practice did not include the formal assessment of HRQoL. American Academy of Ophthalmology reported that patients' perspective was better to represent the visual function, self-esteem and the social interactions problems in the adults with strabismus (Mills, 2004).

### **1.3 Justification of study**

Children with infantile esotropia suffered from various problems that affected their HRQoL. The alarming burden reported including the social, emotional, physical, and functional problems needed to be understood and measured to help in the management and clinical decision. In addition, the increasing level of depression of the mothers is very alarming and needs urgent attention by the clinician and the public. Thus, a study is needed to understand and measure their problems to improve their HRQoL.

At present, there is extremely limited strabismus-specific questionnaire in children worldwide, to measure the HRQoL except for the Intermittent Exotropia Questionnaire (Hatt et al., 2010). Based on our literature search in various database, specific tool to measure HRQoL in infantile esotropia is still not available until now. Therefore, this study is strongly needed to develop a strabismus-specific questionnaire that measure the HRQoL for infantile esotropia from the perception of the children and their proxy/parents.

### **1.4 Objectives**

#### **1.4.1 General objective**

To develop and validate a new strabismus specific HRQoL questionnaire and to evaluate the HRQoL in children with infantile esotropia and their proxy/parents.

### **1.4.2 Specific objectives**

1. To develop a new strabismus specific HRQoL questionnaire for children with infantile esotropia and their proxy/parents
2. To measure the validity of the newly developed questionnaires
3. To measure the reliability of the newly developed questionnaires
4. To compare the HRQoL score between children with infantile esotropia and normal children using the newly developed questionnaire
5. To compare the HRQoL score between proxy/parents of children with infantile esotropia and proxy/parents of normal children using the newly developed questionnaire
6. To determine the factors affecting the HRQoL in children with infantile esotropia using the newly developed questionnaire
7. To compare the HRQoL score in children with infantile esotropia before and after strabismus surgery using the newly developed questionnaire

## **1.5 Research hypotheses**

1. Children with infantile esotropia have lower HRQoL compared to normal children.
2. Proxy/parents of children with infantile esotropia have lower HRQoL compared to proxy/parents of normal children.
3. Gender, angle of deviation and stereopsis affected the HRQoL in children with infantile esotropia.
4. Children with infantile esotropia have better HRQoL after strabismus surgery.

## **CHAPTER 2**

### **LITERATURE REVIEW**

#### **2.1 Strabismus**

Strabismus is a condition of the eye where one or other visual axis is not directed towards the fixation point (Rowe, 2013). The incidence of strabismus is between 0.8 to 8 % of the population (Friedman et al., 2009; Chia et al., 2010; Bruce and Santorelli, 2016; Chen et al., 2016; He et al., 2020). Asian countries such as Singapore, South Korea and Iran reported lower incidence (1.19 % to 1.68 %) compared to the western countries (Chia et al., 2010; Hashemi et al., 2015; Han et al., 2018).

##### **2.1.1 Classification**

Classification of strabismus is according to the direction of deviation and time of onset. According to the direction of deviation, strabismus can be divided into several types including esotropia, exotropia, hypertropia, hypotropia and cyclotropia. Esotropia is a condition in which one or the other eye deviates inwards into the eyes while exotropia is the outward deviation of the eye (Rowe, 2013). Hypertropia occurs if one visual line of the eye is higher than the other. Relatively, hypertropia happens when the visual line is lower than the other eye. Cyclotropia happens when there is a misalignment of one or both eyes around the sagittal axis that produces clockwise (incyclotropia) or counter clockwise (excyclotropia) rotations of the globe (Von Noorden and Campos, 2002).

Another classification is by the time of onset of deviation, in which strabismus is divided into congenital or acquired type. A deviation noted at birth or in the first months of life is termed congenital. However, because it is difficult to document the deviation at birth, the congenital term has been replaced or used together with the infantile term. Infantile include all forms of strabismus with an onset during six months of life. Any form of strabismus that arises after that is known as acquired or primary heterotropia (Von Noorden and Campos, 2002).

Strabismus can also be classified by latency, either phoria or tropia. Phoria is a latent strabismus. When phoria is present, both visual axes are directed towards the fixation point but deviate on dissociation. When there is no deviation on dissociation, the condition is called orthophoria. Among types of phoria are esophoria, exophoria, hyperphoria, hypophoria, and cyclophoria (Rowe, 2013). Tropia is a manifest strabismus. It is a condition which one or other visual axis is not directed towards the fixation point. Tropia can be classified into comitant or incomitant strabismus. The classification of horizontal strabimus was divided into Figure 2.1 for esotropia and Figure 2.2 for exotropia.

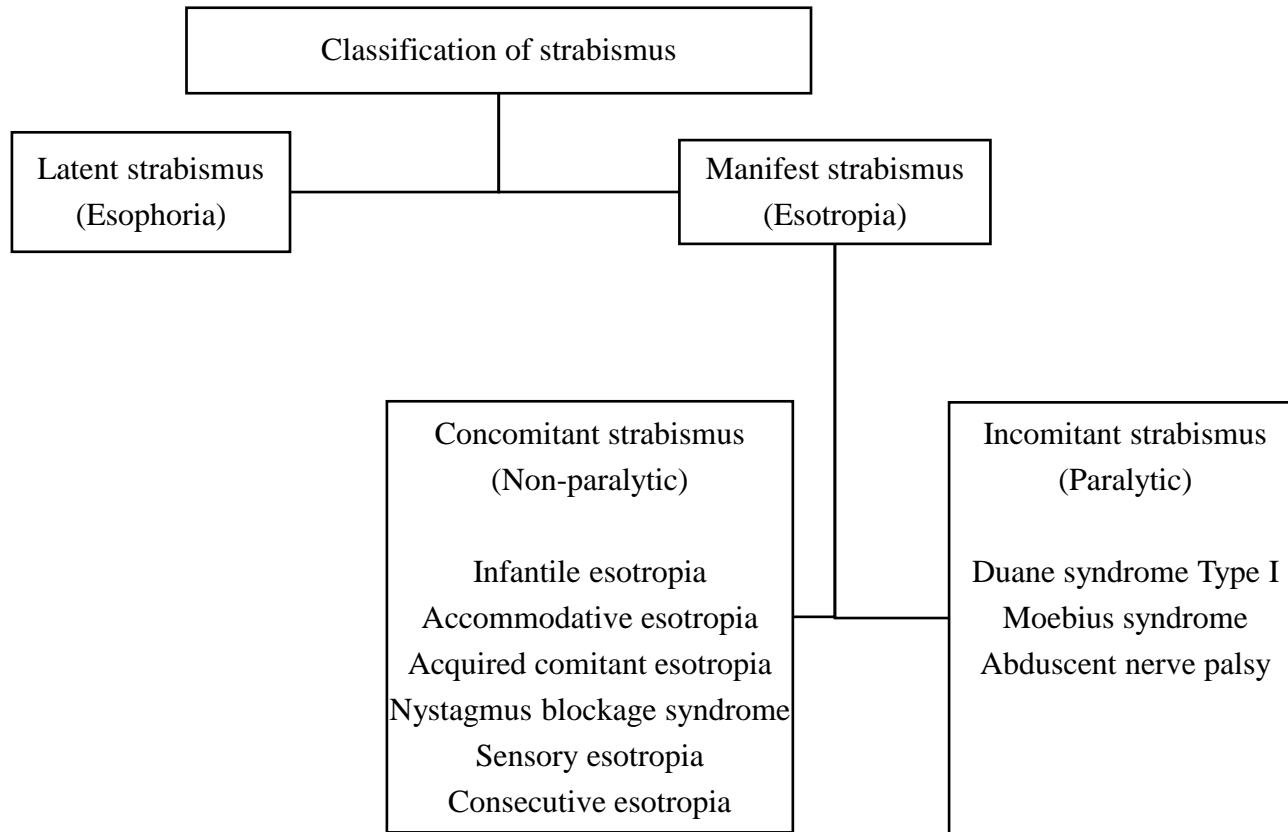


Figure 2.1 Classification of esotropia

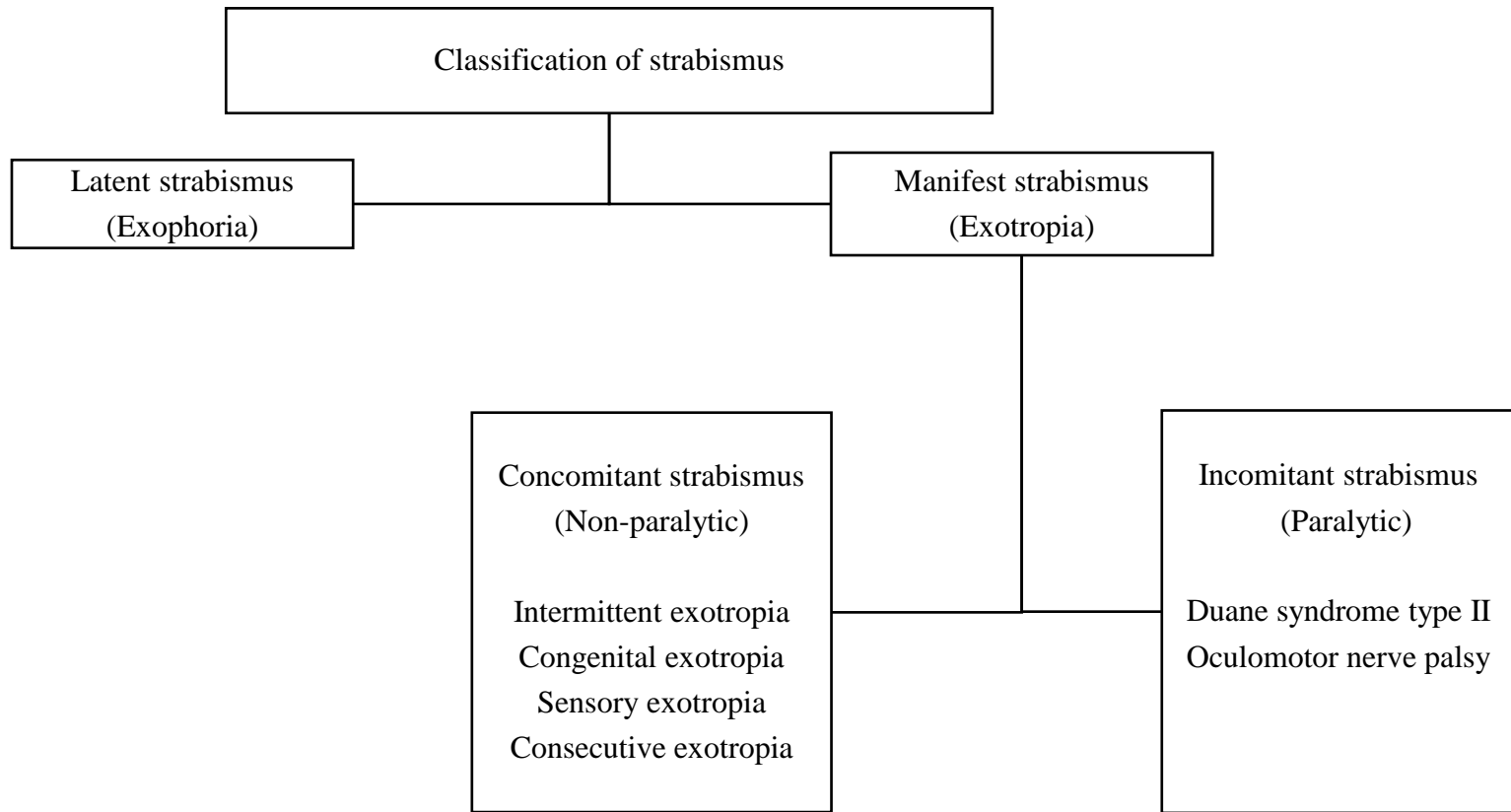


Figure 2.2 Classification of exotropia

### **2.1.2 Concomitant strabismus**

Comitant strabismus or non-paralytic strabismus include strabismus with equal deviation in all positions of gaze (Rutstein et al., 2011). It is more frequent in childhood strabismus. This means that the angle of deviation of the strabismus will remain the same at all direction of gaze when examined at a particular test distance. (Billson, 2003).

Among common types of concomitant esotropia are infantile esotropia, accommodative esotropia, acquired comitant esotropia, nystagmus blockage syndrome, sensory esotropia and consecutive esotropia (Figure 2.1). Common types of concomitant exotropia are intermittent exotropia, congenital exotropia, sensory exotropia and consecutive exotropia (Figure 2.2).

### **2.1.3 Incomitant strabismus**

Incomitant strabismus or paralytic squint is present when the angle of deviation measured at a certain test distant varies (Rutstein et al., 2011). This is subject to the direction of gaze and will be greatest in the direction of the affected muscle movement. Incomitant strabismus occurs in adult onset strabismus rather than childhood strabismus (Billson, 2003). Among common types of incomitant esotropia are Duane's retraction syndrome Type I, Moebius syndrome and Abducent nerve palsy (Figure 2.1). Common incomitant exotropia include Duane syndrome type II and Oculomotor nerve palsy.

## **2.2 Infantile esotropia**

Infantile esotropia is a type of esotropia with onset before the age of six months. For decades, it had been considered the most recognized type of strabismus with a 1% incidence of the population and 28 to 54% of all types of esotropia (Major et al., 2007). The prevalence varied from 0.1 to 2% of the population (Von Noorden and Campos, 2002; Greenberg et al., 2007, Abadom et al., 2014). Infantile esotropia was additionally represented around 8.1% of esotropia, influencing 1 in each 100 to 500 people (Mohny, 2007). However, there is no study conducted on the prevalence of infantile esotropia in Malaysia. According to a retrospective study on 98 strabismus patients attending Hospital USM, Kubang Kerian, Kelantan, Malaysia, 28.6% (26 patients) were diagnosed with infantile esotropia (Waheeda-Azwa et al., 2020).

Patient with infantile esotropia had a history of affected parents or siblings. Infantile esotropia must be differentiate from other type of strabismus including sensory and accommodative esotropias, other early-onset esotropias, Moebius syndrome, Duane syndrome, congenital abducens nerve palsy, and pseudoesotropia (Christenson et al., 1990).

### **2.2.1 Clinical characteristics**

Patients with infantile esotropia may exhibit clinical characteristic which includes, constant and large-angle esotropia which normally at 40 to 60 prism diopter at distance and near that occurs before six months. In addition, they also display other clinical characteristics such as refractive errors of low hyperopia. However, 50% were reported with hyperopia of more than two diopter. Forty percent of the patients were diagnosed with amblyopia (Rutstein et al., 2011).

Other characteristics are the high frequency of ocular motility disorders. These include dissociated vertical deviation, less possibility for normal binocular vision and higher levels of stereopsis, limited abduction, latent or manifest nystagmus, inferior oblique overaction and asymmetry monocular nasotemporal-pursuit optokinetic nystagmus (Rutstein et al., 2011; Simonsz and Kolling, 2011). It is also related with cross-fixation, a condition of using the right eye to view targets at the left field of gaze and vice versa. This condition would interfere will interfered severely the normal binocular vision development (Christenson et al., 1990).

Interestingly, the ocular alignment is not stable in infantile esotropia. Commonly at the age of six months, more than 25% of infants with esotropia diagnosed at two to four months of age, become orthophoric spontaneously. The esotropia appears to be small-angle, intermittent, and variable. However, this is not happened in constant and large of more than 40 prism diopter (Beck, 2002). A patient with infantile esotropia may achieve satisfied binocular vision with the angle of deviation of less than 10 prism diopter after the strabismus surgery. Stereopsis can be developed if the angle of deviation is less than four prism diopter after the surgery (Leske and Holmes, 2004).

### **2.2.2 Risk factors**

Researchers reported risk factors including male, prematurity, low birth weight, systemic disorders, secondary ocular history, family history, perinatal or gestational complications, neonate oxygen usage, systemic medications, and low Apgar scores (Mohney et al., 1998; Major et al., 2007; Simonsz and Kolling, 2011).

The risk factors identified will help the clinician to detect infantile esotropia early and provide the appropriate management.

### **2.2.3 Management**

Infantile esotropia should be managed to improve the ocular misalignment and optimise the potential for binocularity. Previously, Ing (1993) suggested post-surgery alignment within ten dioptres by two years old for the best prognosis of binocular vision. However, in a review on management of infantile esotropia by Hug (2015), paediatric ophthalmologist used preferences according to their training, previous studies, and personal experience in the decision making of the surgery.

Cerman et al. (2014) performed a retrospective review to determine optimal age of surgical intervention. They used the development of stereopsis as the treatment goal. The study reported that patients operated on before 13 months of age developed some level of stereoacuity. In contrast, none of the patients operated on after 39 months of age developed stereopsis. The study reported high sensitivity (66.7%) and specificity (92.3%) in predicting existence of stereopsis, when surgical success was defined as within five prism diopters of orthotropia.

A systematic review by Elliott and Shafiq (2013) had also evaluated the effectiveness and the best time for the treatment of infantile esotropia. The review concluded that it was unrealistic to determine the discussions with respect to the age of intervention, non-surgical management, and type of surgery.

The management of infantile esotropia maybe non-surgical, surgical, or combination of both treatments. The non-surgical management include the correction of refractive error, patching treatment and botulinum toxin. Patient with significant hyperopia should be treated prior to the surgery. However, in most cases, the refractive error is insignificant and the large esotropia persists despite the corrective lenses and repeated cycloplegic refraction (Rutstein et al., 2011).

Patient with amblyopia should be treated before the surgery. The amblyopia therapy or patching treatment should be prescribed on the preferred eye for two hours daily based on amblyopia study. The children with infantile esotropia who undergo the patching treatment should be monitored for every four to six weeks (Chen and Cotter, 2016). When the amblopia has resolved, the maintainance of the patching therapy maybe required (Repka et al., 2003)

Early surgery is indicated for infantile esotropia to increase likelihood of the development of stereopsis. Approximately 40% of surgically treated infantile esotropia achieved stereopsis (Donahue, 2007). The surgical ocular alignment should be considered when the angle of deviation is large and non-accommodative. Most surgeons choose to intervene before 24 months of age, and as early as six months to establish the binocular vision (Rutstein et al., 2011). Surgical management to correct the esotropia involves the horizontal extraocular muscles, including the bilateral surgery, unilateral surgery, and three or more muscle surgery. The bilateral surgery is conducted by weakening the medial rectus of both eyes. The unilateral surgery is performed by the medial rectus recession and the lateral rectus resection for dense amblyopia. While the three or more muscle surgery is the combination of resections

and recessions of the muscles. It is a choice for angle of deviation of more than 50 prism diopter

The botulinum toxin is used to block the muscle contractions for the non-surgical management. It is injected into the medial recti to weaken their action and paralyse the muscles temporarily. This allows the lateral recti to act unopposed. The ocular alignment will be improved when the paralytic effect of the muscles disappears after several months.

According to Hug (2015), the role of botulinum toxin remains questionable although it has been used as augmentation with medial rectus recession, and as treatment for residual esotropia. A recent Cochrane review by Rowe and Noonan (2017) studied the randomized controlled trials of the use of botulinum toxin for the treatment of strabismus. Two studies reported no treatment outcome difference in the retreatment of acquired and infantile esotropia. One trial showed a poorer response for the use of botulinum toxin than surgery for the treatment of horizontal strabismus in patients without binocular vision. Interestingly, the complication rate including ptosis, vertical deviation, diplopia, and subconjunctival hemorrhage was reported to be between 24% and 55% in the patients.

### **2.3 The impact of strabismus**

Strabismus had been reported to impact adult and children psychosocially and functionally. These impacts may occur both in adult and children. However, some impacts exist according to age.

In a study conducted by Sim et al. (2014), strabismus children were found to have significant decrease in the quality of life scores compared to normal children. The study done on 60 children with strabismus in Singapore measured the psychological and functional impact of strabismus. The impact of strabismus on the children's quality of life was measured using the Intermittent Exotropia Questionnaire (IXTQ) and the Adult Strabismus 20 Questionnaire (AS20). The study reported that children with strabismus scored lowest in items including having to wait for their eyes to clear up, being worried about their eyes, bothered about people's wondering what may be wrong with their eyes, and worried about what people thought of them. The study concludes that strabismus has an adverse effect on the quality of life of the children. However, the effect on the change of policy in Singapore was not documented.

Parents act as a proxy to their children since they identify and describe the problems faced by their children. The majority of parents reported public ignorance and low communication skills of the children compared to their friends. Some of the parents also stated that their child were angry when they were commented to have strabismus (Kothari et al., 2009). The parents also said that their child was very bothered because of individuals' comments, seriously disliked, had extreme trouble in

communication and coping. Such information is important to understand the problems of strabismus and its consequences to the children.

Parents of children with strabismus were also affected by their children's problems. Kothari et al. (2009) conducted a study on 93 children with strabismus and their parents in India. They found significant negative emotional and psychosocial effect of the childhood strabismus. The majority of the parents were bothered because of strabismus, individuals' comments and many were incredibly stressed.

### **2.3.1 Psychological impact**

Psychosocially, adults with strabismus suffer a variety of problems including mental illness, schizophrenia and depression, social phobia and self-image, poor job opportunities (Coats et al., 2000), interpersonal relationship (Satterfield et al., 1993) and difficulties in finding partner (Satterfield et al., 1993, Coats et al., 2000, Mojon-Azzi and Mojon, 2009, Olson et al., 2011). Strabismus influenced negatively the individuals' opportunity to obtain work and therefore has an impact on their economic status (Satterfield et al., 1993, Mojon-Azzi and Mojon, 2009).

Interestingly, strabismus was also reported to give remarkable impact to children. Strabismus is often socially seen as stigmata among the society. Among problems faced by the children were poor interpersonal relationship, reduced self-image, ridicule, negative social bias, depression, and social anxiety (Satterfield et al., 1993, Coats et al., 2000; Mohny et al., 2008; Mojon-Azzi et al., 2011; Lin et al., 2014). In a study by Lukman et al. (2011), children were reported to refuse sitting beside a strabismus person, specifically the exotropia type.

### **2.3.2 Functional impact**

Functionally, adults with strabismus reported abnormal facial appearance and experienced symptoms such as diplopia and problems in depth perception (Von Noorden, 1985). Some are also visually impaired. They will suffer from loss of vision if the strabismus was not treated (Von Noorden, 1985). As a result, these problems will affect them in work, hobbies, sports and driving (Satterfield et al., 1993).

## **2.4 Measuring health-related quality of life in strabismus**

The idea of health-related quality of life had increased in popularity as an significant patient-reported outcome, as it originated from individuals' feelings and perceptions about the health status (Mruthyunjaya et al., 1996). The information on the quality of life (QoL) often assists policy makers and health professionals to understand and assess the cost-effectiveness of the treatment and patients' satisfaction. QoL is different between individuals with similar sickness and within a person after some time, as it depends on personal perception and impacted by mental, social, and physical elements (Wang et al., 2018).

HRQoL studies were introduced to ophthalmology since the 1980s to discover the impact of the disease on the overall quality of life on patients, either physically or mentally. HRQoL questions have become a critical segment of public health surveillance and are commonly viewed as valid indicators of requirements and management outcomes. Self-assessed health status is a more remarkable predictor of mortality and morbidity than numerous objective health measures. HRQoL measures may demonstrate the impact of health on quality of life scientifically (Centers for Disease Control and Prevention, 2016).

The assessment of HRQoL is crucial for health outcomes research and assessment of the medical intervention effectiveness. HRQoL assessments are progressively applied in healthcare to measure the patients' quality of life. These measures help the clinician to make clinical decision through the evidence-based practice. Currently, the HRQoL is measured by the general and specific questionnaires.

#### **2.4.1 Questionnaire**

Questionnaire is a research tool to gather information from respondents. Questionnaires can be administered face to face, by telephone, computer or post. It is a cheap, fast and effective way on collecting information from the respondents. It is an effective method to measure the behaviour, attitudes, opinions, preferences and intentions of large numbers of participants more cheaply and quickly than other methods. Usually, a questionnaire includes both open and closed questions for collecting data. This is helpful as it obtained both quantitative and qualitative data (McLeod, 2018).

A questionnaire consists of a set of standardized questions called items (Lavrakas, 2012). The items should be in order to collect individual data about one or more specific topics. The response scales are the indexes that measure the types of variables that are not directly observed, but are inferred from the other variables that are directly measured. The Likert scale is the most popular type of scale in a questionnaire.

When a Likert-type response scale is used, the points on the scale should represent the entire measurement range. The responses should be presented in an ordinal sequence without any uncertainty (Boateng et al., 2018). Each point on the response scale should be meaningful and interpreted the similar way by each participant to ensure data quality.

Krosnick and Presser (2009) reported that response scale with two to three points have lower reliability than Likert-type response scales with five to seven points. Hence, response scales with five points are recommended for unipolar items, for example the response scale representing relative degrees of a single item response quality (For example “not at all satisfied” to “very satisfied”). Seven response items are recommended for bipolar items, for example those reflecting relative degrees of two qualities of an item response scale (For example “completely dissatisfied” to “completely satisfied”).

#### **2.4.2 Development of questionnaire**

The development of questionnaire involves the identification of domains and the item generation (Boateng et al., 2018). The first step is to identify the domains. A domain means the concept, attribute, or unobserved behavior that is the objective of the study (Haynes et al., 1995). It should be decided and defined before any item process (Streiner et al., 2015). A well-defined domain will provide the information of the phenomenon, specify the limitations of the domain, and aid the process of item generation and content validation.

McCoach et al. (2013) suggested several steps for the identification of domain. These includes specifying the purpose of the domain to be developed, and confirming that there are no available instruments that will provide the same objective. If there is similar questionnaire, the researcher should justify the need of developing the new questionnaire, as well as the difference between the existing and new one. Next step is to describe the domain, provide preliminary conceptual definition and specify the dimensions of the domain if there is any theory underpinning the study. The domains are classified as a priori if there is an established theory guiding the study, but a posteriori if none occurs. Finally, if the domains are identified a priori, the conceptual definition for each domain should be detailed.

The item generation involved identifying appropriate questions that fit the identified domain. There are two methods in this step which are the deductive methods and the inductive methods. The deductive method or known as “classification from above” is based on the information of the relevant domain and the identification of items. This method can be conducted through literature review, evaluation of available questionnaires and indicators of the domain (Hinkin, 1995).

The inductive method or “classification from below” involves the generation of items from the responses of participants (Hinkin, 1995). In this method, the qualitative data is gained through direct observations, focus groups and individual interviews. Hence, the domain items are inductively identified from the participants’ responses (Morgado et al., 2017).

### **2.4.3 Validation of questionnaire**

Validity means the degree to which an evaluation measures what it is supposed to measure. There are three types of validity including the content validity, criterion-related validity, and construct validity (DeVellis, 2003). The validation of questionnaire is conducted to ensure that it measures accurately what it supposed to measures. A valid questionnaire increases the credibility of data by collecting better quality data. The questionnaire must have the following criteria including simplicity and practicality, reliability and accuracy in the words, sufficient for the problem proposed to measure, and reflect underpinning theory to be measured and ability to measure (Kazi and Khalid, 2012).

#### **2.4.3 (a) Content validity**

Content validity is the ability of a questionnaire to represent the domain of interest and the conceptual definition of a construct. Fayers and Machin (2007) stated that there is no formal statistical testing to claim content validity, however, the item generation process should include a literature review of published data, interviews from targeted patients and an expert panel to approach item relevance. Inclusion of at least five experts in that field (Burns and Grove, 2001) would be useful to judge the content domains of a scale through the use of rating scales.

In contrast, Polit and Beck (2006) suggested the content validity index (CVI) as evidence for the content validity of the questionnaires. The CVI was calculated based on the experts' scores of item relevance. Among the advantages of the CVI is the ease of calculation, understandability, focus on the agreement of relevance instead of agreement per se, focus on consensus instead of consistency, the establishment of