ABSTRACT

Title of Dissertation:	PARENT PERSPECTIVES ON DIAGNOSIS OF AND SERVICES FOR CHILDREN WITH CORTICAL VISUAL IMPAIRMENT
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Medical advances in recent years have increased survival rates of infants born prematurely and/or infants and children that present with life-threatening conditions (Good et al., 1994; Khetpal & Donahue, 2007; Murphy & Carbone, 2011). These increased survival rates are associated with an increase in the number of children who have severe and/or multiple disabilities, including those conditions that are associated with cortical visual impairment. Children with typical or nearly typical eye exams, but having observable visual impairment are those generally diagnosed with cortical visual impairment, or CVI (Jan, Groenveld, Sykanda, & Hoyt, 1987). Delayed or lack of diagnosis of CVI can lead to missed opportunities for learning, and especially missed sensitive periods during which recovery can occur faster (Hubel & Wiesel, 1970; Roman-Lantzy, 2018). Without diagnosis, children may not be eligible for

funding assistance for educational materials (American Printing House for the Blind, n.d.*b*). The purpose of this study was to explore parents' experiences in getting a diagnosis of CVI for their children. For example, whether there were lapses in time between suspected vision difficulties and diagnosis, and what information was provided when diagnosis was obtained. The research questions guiding this investigation included: What are parents' experiences in seeking a diagnosis for their child's suspected vision challenges? What needs do parents recall related to information and supports while seeking a diagnosis for their child's suspected vision challenges? What kind of information is offered or readily available to parents upon diagnosis of CVI? The primary data source for this study was interviews with parents of children having diagnosed CVI. Secondary data sources included interviews with ophthalmologists, teachers of the visually impaired, and records review.

PARENT PERSPECTIVES ON DIAGNOSIS OF AND SERVICES FOR CHILDREN WITH CORTICAL VISUAL IMPAIRMENT

by

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Dedication

This project is dedicated to all of the children that I have been fortunate to call “my kids” these last 12 years, and the families who let me call those children “mine”. Those families and children have always provided me with enthusiasm, a valid reason for asking questions, and they are the “heart” behind the “work”.

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Finally, I need to thank my loving husband, who reminded me to eat and sleep during writing marathons, hugged me when I needed hugs, and gave me space when I needed space. I feel very lucky that he has supported me all this time.

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List of Abbreviations

APH – American Printing House for the Blind

CP – cerebral palsy

CNS – central nervous system

CVI – cortical visual impairment

CT – computed tomography

fMRI – functional magnetic resonance imaging

HIE – hypoxic ischemic encephalopathy

ONH – optic nerve hypoplasia

PET – positron emission tomography

PVL – periventricular leukomalacia

ROP – retinopathy of prematurity

SPECT – single photon emission computed tomography

TVI – teacher of the visually impaired

VI – visual impairment

CHAPTER I

INTRODUCTION

Medical advances in recent years have increased survival rates for infants born prematurely and/or infants and children that present with serious medical or health conditions (Good et al., 1994; Khetpal & Donahue, 2007; Murphy & Carbone, 2011). These increased survival rates have led to an increased number of infants who have potential health-related difficulties, including epilepsy, cerebral palsy, hemiparesis, microcephaly, hydrocephaly, hearing loss, vision loss, learning disabilities, behavioral difficulties, motor impairment, cognitive impairment, and multiple disabilities. (Khetpal & Donahue, 2007; Murphy & Carbone, 2011). Conditions that impact the brain may be associated with cortical visual impairment (CVI) (Groenveld, 2003; Khetpal & Donahue, 2007). Medical advances have reduced the number of children who have blindness due to ocular conditions, leading to an emergence of CVI as the most common cause of visual impairment in children (Good et al., 1994; Groenveld, 2003; Hyvärinen, 2005; Khetpal & Donahue, 2007).

Parent Perspectives on Diagnosis of Disability

Parents of children who have disabilities are partners with child development and medical professionals in the decision-making processes throughout their children's lives, and should be treated as important members of their children's teams. Studies suggest that families prefer to hear accurate information about the condition of their children, rather than be left with the uncertainty of no diagnosis (Adix, Adix, & Rosenthal, 1984; Howie-Davies & McKenzie, 2007; Klein et al.,

2011; Watson, Kieckhefer, & Olshansky, 2006). Unfortunately, several studies have found that many parents suspect their child may have developmental abnormalities long before they receive a diagnosis, and that when they express concerns they are sometimes dismissed or disregarded, only to find later that their suspicions were founded (Baird, McConachie, & Scrutton, 2000; Davies et al., 2003). This suggests that professionals should take parents' concerns seriously. Additionally, for parents whose children's diagnoses are delayed, some parents report that such a delay causes more stress and frustration than the diagnosis itself (Davies et al., 2003). In the absence of a diagnosis, appropriate information and assistance may be more difficult for parents to obtain. These studies are reviewed in more detail in Chapter II.

Theoretical Basis

Sensitive periods and neuroplasticity. To understand the impact of children's cortical visual impairment on parents and families, it is useful to apply the concepts of neuroplasticity and sensitive periods in human development and the theoretical framework of ecological and bioecological systems proposed by Urie Bronfenbrenner (1977, 2001). This theory provides a foundation for explaining how the developing child impacts and is impacted by the child's environments, the resources that exist in those environments, and how the environments interact with one another, and the child. The concepts of sensitive periods of visual development and neuroplasticity are important to the discussion of CVI diagnosis and intervention. Neuroplasticity refers to the ability of the brain to change and reorganize due to experiences. This can be stimulated by learning new skills and can be regulated over a sensitive period, in which sensory experience has a greater influence on behavioral and cortical

development (DeMaster et al., 2019; Silva et al., 2018). In the context of a child's vision, the concept of a sensitive period refers to times during a child's growth when visual development, including learning to use and understand the information that the eyes take in, is at its highest, and when neuroplasticity is greatest.

The work of Lewis and Maurer (2005) examined the concept of sensitive periods, and found evidence to support multiple periods of sensitivity in visual development in humans, varying for different aspects of visual acuity, movement detection, peripheral light sensitivity, and face recognition (Lewis & Maurer, 2005). They also examined the similarities and differences between sensitive periods of "normal development", of "damage" (such as visual deprivation after typical visual development is already in progress), and of "recovery", aiming to discover information about how brain plasticity changes for different forms of visual processing over time. Their work supported the notion that most of the sensitive periods of visual development occur during early childhood (Lewis & Maurer, 2005)

The critical point in research about neuroplasticity and sensitive periods in visual development, as related to CVI, is that children can recover more quickly and gain more usable vision when opportunities for intervention are provided as early as possible during the child's life (Hoyt, 2003; Pallagrosi, 1993; Roman-Lantzy, 2018). This suggests that earlier diagnosis and intervention may be more effective than intervention provided later, which aligns with fundamental concepts related to neuroplasticity. Neuroplasticity refers to the ability of the neurological system to change in "response to new information, sensory stimulation, development, damage, or dysfunction" (Rugnetta, 2019). As a child matures and receives intervention, she

can gain visual skills as intact areas of the brain change or gain function. Since neuroplasticity is greater in younger children, and the sensitive period for visual development is within early childhood, the prognosis for visual progress in children with CVI is greater when they are provided with earlier intervention (Cohen-Maitre & Haerich, 2005; Good et al., 1994; Good, Jan, Burden, Skoczenski, & Candy, 2001; Hoyt, 2003; Huo, Burden, Hoyt, & Good, 1999; Khetpal & Donahue, 2007; Lam, Lovett, & Dutton, 2010; Lueck, 2010; Malkowicz, Myers, & Leisman, 2006).

Bioecological Systems theory. In his original ecological systems theory, Bronfenbrenner suggested that to understand human development, a broader, systems approach is necessary that places the child in the context of a larger, nested system (Bronfenbrenner, 1977). This nested system is composed of several levels, all of which have bidirectional influences. The first level, which Bronfenbrenner called the *microsystem*, includes the immediate settings or environments in which the child functions. Children may participate in several microsystems – for example the family home comprises a micro-system, as does the classroom, day-care setting and other settings in which the child participates directly. Microsystems have direct effects on the child – for example, the diagnostic process for a child may be directly impacted by the family home situation and the family's resources and ability to pursue a diagnosis. A child with CVI can also be affected by the quality of intervention he receives, the adaptations made by his teacher, and the amount of sensory input in the classroom. Also, as stated by Bolinger and Bolinger in Holbrook (1996), a child's visual impairment can have influences on the family context – for example, the resources that a family uses to manage the obstacles associated with the disability

may have an impact on parents and other family members (Holbrook, 1996). Holbrook argues:

> Having accurate, up-to-date information about your child's [Visual Impairment] in general, and how it affects her specifically, can help you feel more in control when it comes to decisions about her care and future. You can at least agree or disagree knowledgeably with professionals' recommendations, and possibly begin to offer some recommendations of your own (Holbrook, 1996, p132).

Bronfenbrenner described the interrelationships between microsystems and labeled those interactions the *mesosystem*, including the relationships between participants and the major settings that contain the developing person, including the interactions between family and the school. In effect, a *mesosystem* is a system of *microsystems*. An example of how the mesosystem might affect a child with CVI is the level and style of communication between parents and school staff, especially about concerns and progress in use of vision. Another important effect of a mesosystem is the family-professional relationship, which may vary greatly depending on either the parents or the service provider and result in the parents' satisfaction or dissatisfaction with a certain level of care. Absence of an accurate medical diagnosis can also determine how microsystems interact, as medical and service professionals may not recognize parent concerns without a diagnosis.

The third level, termed *exosystem*, includes an extension of the *mesosystem* and includes larger systems in which *micro-* and *mesosystems* are embedded. At this level, Bronfenbrenner includes larger agencies and policies that may affect the child

and family by influencing events that occur in the *microsystem* and *mesosystem*, but in which the child is not directly involved. Examples of such include a parent's workplace, and government agencies, all of which affect the developing child even though the child is not a direct participant. For a child with CVI, this may mean that policies dictated by government agencies affect the child's access to services and interventions (e.g., whether the diagnosis of CVI is included under the term *legal blindness*, enabling individuals with CVI to receive services for the visually impaired). The lack of appropriate early intervention may create greater need for assistance from society as the child develops; a need that may be impacted by and itself may impact policy and society.

The fourth level of this ecological systems theory is the *macrosystem*, which refers to the overall patterns of culture and subculture in which the other systems exist. For example, the way a society or culture views disability in children may affect the availability of services.

Bronfenbrenner also included time as a dimension, labeling it the *chronosystem.* It is included to account for historical time, as well as changes that occur over time at each level of the system, and how each level may, in turn, influence other levels over time (Bronfenbrenner, 2001). An example of how this can affect a child with CVI is that certain environments (e.g. home; classroom) may (or may not) provide specific forms of visual stimulation, which may influence the visual functioning of the child over time. The effects of and on the environment and society may become more pronounced as the child ages. Following Bronfenbrenner's theory, the effects of and upon the child (or children) progress to affect family, policy, and

culture over long periods that extend beyond the child's lifetime. In later years, Bronfenbrenner expanded this theoretical approach and renamed it *Bioecological Systems Theory*, to include the continuity and evolution in the biological characteristics of human beings as individuals and as groups. It extends throughout the lifetime, generations, and historical time (Bronfenbrenner, 2001). This relates directly to the biological nature of visual impairment and individual characteristics of the developing child, and his or her participation in his or her own development (Rosa & Tudge, 2013).

As his theory continued to evolve, Bronfenbrenner also introduced the concept of *proximal processes*, which proposes that complex, persistent, reciprocal interactions with and between those persons, objects, and symbols closest to an individual are the means by which that individual develops (Rosa & Tudge, 2013). It may be said, therefore, that the family of an individual not only affect the individual as a *microsystem*, but also in terms of the interactions of family members as *proximal processes*. *Process* became part of Bronfenbrenner's *Person-Process-Context-Time* (*PPCT)* model of development. *Person* refers to the biological and genetic aspects of the person, and is divided into the characteristics he named *demand, resource,* and *force*. *Demand* characteristics are those that act as a stimulus to other persons, such as what others can see, which may influence expectations. *Resource* characteristics are those that refer to mental and emotional resources such as past experiences and skills, as well as material resources such as quality housing, food, and educational opportunities. *Force* characteristics are those related to temperament and motivation. *Process* refers to proximal processes, *Context* to environmental factors (such as the

layers of the bioecological system), and *Time* represents multiple facets of time: *microtime, mesotime, and macrotime. Microtime* refers to what is occurring during an activity or interaction, *mesotime* is related to the frequency and consistency of these episodes of activity, and *macrotime* refers to the *chronosystem*, including historical events in society and the passage of time (Rosa & Tudge, 2013; Tudge, Mokrova, Hatfield, & Karnik, 2009).

Viewing the concepts of sensitive periods and neuroplasticity within the context of Bronfenbrenner's Bioecological Systems Theory, one can more easily understand the importance of family and professional partnerships in human development. The concepts of neuroplasticity and sensitive periods are bioecological in nature; that is, they are biological phenomena that affect the overall development of human beings, impacting all other systems. When a diagnosis is delayed, withheld, or never determined to explain a child's characteristics, as is often the case in children with CVI, it may impact all other levels or systems, which may be further compounded by missed sensitive periods and reduced neuroplasticity. Considering the *chronosystem*, the effects of all other systems on one another become more complex. For example, regarding diagnosis, one can consider the impact of delayed diagnosis over time – impacting the child's ability to take in information about his environment (which greatly reduces conceptual development over time), personal and social interactions with others, and family life.

Taken together, the concepts of neuroplasticity and sensitive periods in visual development along with Bronfenbrenner's Bioecological Systems Theory provide a framework from which to consider the importance of early diagnosis and intervention

for children with CVI, and parent perceptions of this process. As children age and pass beyond the sensitive periods of visual development, the impact of CVI on the nested, interacting systems may become greater due to decreased pace of the child's progress, and the impact of the systems on the child also become more pronounced due to the child's (generally) increased awareness of the environment. For the purposes of this paper, the framework provided by these concepts and this theory exemplifies the importance of early diagnosis and provision of services for children with CVI, in addition to the importance of professional regard of parent input and concerns about their child's use of vision early in their child's life.

Definitions

To provide clarity, it is important that several terms be defined. *Visual impairment* (VI) is typically defined as any functional limitation in the visual system, due to disorder or disease, and is not fully correctable by eyeglasses, contact lenses, or surgery. VI can be mild to severe, including total blindness or functional blindness where no useful vision remains (Freeman, Cole, Faye, Freeman, Goodrich, & Stelmack, 2010). *Functional vision* refers to a person's ability to use their vision in performing daily tasks. Functional vision can vary greatly over time for any individual due to factors such as fatigue, lighting, and other environmental factors. *Visual acuity* is the ability to see fine details, (Freeman et al., 2010). *Visual fields* refer to the part of a person's vision that enables the individual to see what is happening in a particular peripheral location with respect to his or her body, such as lower field (immediately in front and down), upper field, and right and left fields (peripheral) (Freeman et al., 2010). The term *ocular visual impairment* refers to VI

due to dysfunction of the structures of the visual system, including the eyes and nerves.

In the United States, where typical vision is 20/20 (meaning a person can see at 20 feet what most people see at 20 feet), *legal blindness* is defined as a visual acuity lower than or equivalent to 20/200 in the better eye with the best correction (meaning a person can see at 20 feet what a person without VI can see at 200 feet), or a visual field loss of 20 degrees in diameter (sometimes referred to as "tunnel vision") (Freeman et al., 2010). *Cortical visual impairment* (CVI) is a neurological visual disorder, defined for educational purposes as difficulty in processing the information received from the eyes, resulting in unique visual responses to people, educational materials, and to the environment (Roman-Lantzy, 2018). Students who exhibit these unique visual responses (which will be discussed at a later point in this paper), have sustained insult or injury to the brain, and are judged by their performance to be visually impaired are considered to have CVI (Roman-Lantzy, 2018). Individuals can have CVI, ocular VI, or both.

Diagnosis of CVI

Cortical visual impairment has emerged as a leading cause of visual impairment in developed countries (Good et al, 2001; Groenveld, 2003; Hatton, Schweitz, Boyer, & Rychwalski, 2007; Hyvärinen, 2005; Jan, Good, & Hoyt, 2004; Khetpal & Donahue, 2007; McKillop & Dutton, 2008). CVI is, in the opinion of some medical professionals, difficult to medically diagnose due to lack of definitive medical testing, and it can sometimes be similar in appearance to other conditions. Parents often suspect a problem with their child's vision, but in the absence of ocular

visual impairment, professionals often do not know what to do. Few professionals currently understand CVI and what can be done about it, but timely diagnosis and referral for intervention is crucial to rapid recovery and improvement of skills that affect many areas of life (Roman-Lantzy, 2018). These issues combine to create a situation: CVI has effects that can be greatly mitigated by early detection and intervention, but is difficult for many professionals to diagnose, and a lack of professional knowledge about CVI, may cause delayed or absence of a diagnosis. This creates additional issues for parents of children who have CVI, as evidenced by the literature on undetermined or undisclosed diagnosis of other developmental conditions from the parent perspective. Therefore, it is important to determine how delayed or unknown diagnosis of CVI affects parents of children who have this condition.

Cortical visual impairment is considered difficult to diagnose for several reasons. Sometimes, CVI is mistaken for other developmental disorders due to the behavioral similarities that accompany sensory processing difficulties and cognitive impairments, including visual attraction to movement, fixation on specific sensory stimuli, facial recognition difficulties, eye contact avoidance, improved performance in familiar environments, and difficulty with combining use of vision with grasp/reach (Davis, Bockbrader, Murphy, & O'Donnel, 2006; Morse, Pawletko, & Rocissano, 2000). Medical imaging is able to identify damage to brain tissue that is often associated with CVI, but is not definitive specifically to CVI. However, CVI does have a set of very specific behaviors that tend to accompany it, which is currently the basis for diagnosis. Cortical visual impairment is caused by a

malformation of or an insult or injury to the brain, and the most common causes are lack of oxygen or excess carbon dioxide in the brain; lack of oxygen resulting in low blood flow causing irritation of the brain; bleeding into the brain; injury to or death of white brain tissue; neonatal stroke; infection; structural abnormalities; metabolic/mitochondrial conditions; chromosomal and genetic disorders; and trauma (Good et al., 2001; Khetpal & Donahue, 2007; Roman-Lantzy, 2018). CVI is often accompanied by a normal ocular visual exam though it may co-exist with ocular visual issues. For example, children with CVI do not seem to use their vision as well as children with typically developing vision skills, often relying on peripheral vision, requiring that a visual target be moving (or using the motion of their own body to facilitate use of vision), or gazing excessively at lights or lighted targets (Jan, Groenveld, Sykanda, & Hoyt, 1987). Children with CVI typically "act as though they cannot see" and their difficulty in using vision cannot be accounted for by an ocular issue, or the difficulties are greater than expected for the level of visual acuity determined (Roman-Lantzy, 2018, p 3). A significant difference between CVI and ocular VI is that children with CVI often have the ability and potential, given the appropriate interventions, to improve their functional vision skills and use their vision more productively and functionally, due to neuroplasticity (Cohen-Maitre & Haerich, 2005; Good et al., 1994; Good et al.,. 2001; Hoyt, 2003; Huo at al., 1999; Khetpal & Donahue, 2007; Lam et al., 2010; Lueck, 2010; Malkowicz et al., 2006). In other words, the use of vision can be learned as other skills are learned, given appropriate instruction and accommodation (Dutton et al., 1996; Geruschat, 2005; Hoyt, 2003).

Medical imaging. Medical imaging is sometimes used to aid in the diagnosis of CVI-related conditions. However, thus far most imaging techniques can reveal damage to the brain itself, but not specifically to the visual areas in the brain because the visual system is widespread throughout the brain (Thompson & Kaufman, 2003; van Genderen et al., 2012). Vision depends upon intact visual pathways, including the eyes, optic nerves, optic chiasm, and multiple parts of the brain including the primary visual cortex, referred to collectively as the geniculostriate pathways. Complex visual interpretation also requires the association cortex. Cortical visual impairment is caused by any condition that affects these structures. Therefore, medical imaging that can demonstrate such damage can sometimes be helpful in diagnosing CVI, through the diagnosis of associated conditions (Good et al., 1994).

The area of the visual system that is affected can influence the extent that CVI has an impact on each visual component (i.e. color, fields, movement). There are a number of diagnostic imaging options that may help guide the diagnostic process for individuals with CVI. The electroretinogram (ERG) provides information about retinal function, and should be normal in children with CVI and no other vision conditions (Afshari, Afshari, & Fulton, 2001; Good et al., 1994; Whiting et al., 1985). Ultrasound, computed tomography (CT), magnetic resonance imaging (MRI), positron emission tomography (PET), single photon emission computed tomography (SPECT), and functional MRI (fMRI) are options that can reveal damage to brain tissue (Flodmark et al., 1990; Good et al., 1994; Good et al., 2001; Whiting et al., 1985). These imaging techniques may help identify the causes of CVI, though do not themselves result in CVI diagnosis. Because of its portability, ultrasound is the best

method to detect periventricular leukomalacia (PVL), a condition that is frequently associated with CVI. MRI is also used to detect PVL and to assess asphyxia and damage to the white matter of the brain (Good et al., 2001). PET and SPECT can provide information about delivery of blood to the white matter and visual cortex, and the damage that results when blood perfusion to a particular area decreases. Magnetic Resonance and CT have been found to be less useful for diagnosis immediately following the injury due to high water content of the newborn brain. However, during the chronic phase of brain damage, these tests can show enlarged ventricles with irregular margins and decreased white matter between the ventricles and cerebral cortex (Good et al., 1994; Good et al., 2001). Due to the lack of diagnostic imaging techniques that definitively and conclusively demonstrate the existence of CVI, as a condition it is difficult to diagnose, and the diagnosis process is not straightforward. Therefore, particular attention must be paid to the behavioral characteristics that aid in the diagnostic process.

Unique visual behaviors of children with CVI. Unique visual behaviors of individuals who have CVI include specific color preferences, attraction to movement, visual field preferences, difficulties with visual complexity and novelty, visual attraction to light (light gazing), visual latency, atypical visual reflexes, atypical or absence of visually guided reach and movement, and non-purposeful gaze (Edelman et al., 2006; Good et al. 1994; Good, 2001; Jan et al., 1987; Roman-Lantzy, 2018; Smith, 2007). Due to the large number of causes and symptoms of CVI, no two children with this condition may exhibit the signs in the same way, yet the presence of these behaviors is one of the most reliable ways to diagnose CVI (Roman-Lantzy,

2018). Fatigue, complexity in the environment (including sensory input channels other than vision, for example, noise), and health status may all have a profound impact on a child's success in using his or her vision. Often, children will close their eyes when they are listening, presumably to block out the visual stimuli and focus on auditory processing (Whiting, Jan, & Wong, 1985). Investigators in one study found that the most effective way to assess the children was to play with them and observe how they used their vision (Jan et al., 1987). They also found that familiar environments helped children use their vision functionally, as did verbal explanation of what they should look for and where it was located.

Older children with CVI (preschool age and older) often show signs of impaired facial recognition and social gaze (avoidance of looking at faces), difficulty in identifying shapes and objects, impaired visual-motor function (visually guided reach), impaired attention, difficulty with orientation, increased visual fatigue, and non-use of vision when the surrounding environment is complex with regard to sensory stimulation, impaired visual attention, and lack of visual curiosity (Jan et al., 1987; McKillop & Dutton, 2008). Children with CVI also typically have difficulties at school, affecting their ability to sit still, focus on schoolwork, and look at presented materials, including reading, so it is sometimes misinterpreted as Attention-Deficit/Hyperactivity Disorder or a learning disability. The characteristics of CVI can also have an impact on social development, due to lack of facial recognition because facial expressions, hair styles, makeup, and other facial changes which increase complexity, and children with CVI often do not sustain eye contact. A person with lower field impairment, for example, may not notice an outstretched hand

for a handshake, may not see the food closest to him/her on a plate, or may not see items on the ground as they walk (Lam et al., 2010). Since visual difficulty is not the most common cause of such behavior, physicians and educators often do not consider vision as a potential cause. As a result, these factors also impact the diagnostic process (Febel, 2006). If the existence of CVI is not discovered or pursued, children may not receive the instruction and intervention that will enable recovery of the visual processing system (Good et al., 2001; Lueck, 2010). It is important that CVI be diagnosed and interventions be provided to minimize the impact of the condition as much as possible on the development of the child.

Significance of this Study

Although the diagnostic process may be difficult for parents, studies have demonstrated that parents prefer that information be shared in an open, honest, and direct manner. Uncertainty from professionals about the diagnosis and lack of information about future possibilities leaves parents feeling uncomfortable and uncertain (Adix, Adix, & Rosenthal, 1984; Graungaard & Skov, 2006; Howie-Davies & McKenzie, 2007; Klein et al., 2011; Watson et al., 2006). Prior to receiving a diagnosis, parents' suspicion of child disability, should be taken seriously by professionals (Baird et al., 2000; Davies et al., 2003). Professionals need to check in with parents on a regular basis to be certain that the parents feel they are receiving the information and care they need, as parents do not always feel they are receiving care in the same way that professionals believe they are providing it (Klein et al., 2011). This study sought to examine experiences with the process of getting a diagnosis of

CVI in children, from parents' perspectives, to gain understanding about the level of difficulty and what results from the diagnosis.

Rationale

Since the literature on family experiences with cortical visual impairment is limited, in the following chapter I discuss family perspectives and experiences with diagnosis of disability in general, including a review of the literature, followed by a discussion of family perspectives on disability diagnosis, including CVI. First is a discussion of families and their experiences with disability and diagnosis. Then, I provide a theoretical basis to help explain why it is important to understand family perspectives about diagnosis of CVI. Next, I review the definition and description of cortical visual impairment and other key terminology. Then I discuss characteristics of CVI and the diagnostic process.

Purpose Statement

There are many ways to research the experiences of parents of children who have CVI. However, in an effort to capture all details that parents feel are important, in-depth, personal interviews that allow parents to tell their whole story may best result in data that speaks to professionals in the field of visual impairment. Therefore, a qualitative research design was used for this study. Qualitative research is particularly useful to gain knowledge about particular individuals' perspectives, experiences, and emotions. The goal of this research was to understand what parents experienced when they expressed concerns to professionals about their children's vision. These concerns could not be explained by ocular vision differences, alone, so

the purpose of this research was to address concerns about timely diagnosis and referral for children with suspected CVI.

CHAPTER II

REVIEW OF LITERATURE

In this chapter, I review the literature related to parent experiences and perspectives on receiving their child's disability diagnosis, and parent needs and concerns when their children have disabilities. First, I provide a review of CVI and associated conditions in order to discuss prevalence of the condition. Diagnosis of conditions associated with CVI can influence the diagnosis of CVI, depending on professional awareness of the associations. Following the discussion of CVI, I examine selected articles about parent perspectives on childhood disability with regard to diagnosis. This is followed by a discussion of the limited literature concerned with the impact that CVI has on parents and families.

Literature Review Search Method

I located the studies included in this literature review using several search strategies. First, I conducted an electronic search of EBSCOhost, ERIC, PsychINFO, PsychARTICLES, Dissertations & Theses, Social Sciences Citation Index, Family & Society Studies Worldwide, SocIndex, Academic Search Premier, National Center for Health Statistics Data Warehouse, Web of Science, and. I searched these databases for articles concerned with cortical visual impairment and all combinations and variations of these words: difficulty getting service, child disability, parent perspectives, child disability diagnosis, parent satisfaction, parent professional/physician relationship, delayed diagnosis, family support. In addition, I performed journal searches for the same word combinations in Developmental

Medicine & Child Neurology, Functional Neurology, Pediatric Neurology, Eye, Seminars in Neonatology, Exceptional Parent, Intellectual and Developmental Disabilities (previously Mental Retardation), and British Journal of Ophthalmology. Next, I searched the Journal of Visual Impairment and Blindness for the terms "professional knowledge" and "cortical visual impairment". Additionally, I conducted a search on http://www.spedex.com/napvi/ for additional resources, using the same search terms.

Furthermore, I performed an ancestral search in the reference lists of literature reviews and other research articles and dissertations in the field of cortical visual impairment. An additional search strategy I used was to review the APH website for the following subjects: MEDICAL education; PEOPLE with disabilities; DEVELOPMENTALLY disabled; Colleges, Universities, and Professional Schools; Offices of Physicians (except Mental Health Specialists); PHYSICIANS; MEDICAL care; and PEOPLE with mental disabilities – Care.

These literature searches resulted in over 450 articles, so the abstracts were reviewed and results filtered to eliminate those without relevance to this specific topic of study, such as articles that were solely about parent stress, family-centered care, and parent views on disability in general, educational programming, specific diagnoses, or literacy. The focus of this literature review is Parent Perspectives on CVI Diagnosis and Services, but since very little information exists on this specific topic, the review was expanded to include parent perspectives on disability diagnosis and services, and also includes literature that offers explanations about CVI, its diagnosis, and services. Once the literature that was irrelevant to this review was

removed, 23 studies remained (11 that give recommendations for supporting families, four that discuss delivery of disability diagnosis, two that relate to parental needs, one that specifies best practices for professionals, one that discusses medical terminology, specifically, and four that describe parent-professional partnerships). Additionally, four studies review the prevalence of CVI and conditions that are associated with it. One article was found that describes a study of parent perceptions of CVI and its diagnosis and services, and one book describes the process of the author's family obtaining a diagnosis and services for their child with CVI. In subsequent sections of this Chapter, I review studies conducted in each of these areas and follow that with a discussion of where the literature is lacking, problems with studies in general is they relate to the topic at hand, and what needs to be done to remedy the situation.

Prevalence of CVI and Associated Conditions

Four studies were identified that focus on prevalence, characteristics, and diagnosis of CVI. These studies involved the review of records from three vision clinics (Dutton et al., 1996; Huo et al., 1999; Khetpal & Donahue, 2007), and one national registry (Babies Count - The National Registry for Children with Visual Impairments), (Hatton et al., 2007) to obtain information about the etiology, prevalence, and prognosis of CVI. In Dutton's study, children referred to the Pediatric Vision Assessment Clinic in Glasgow, Scotland were examined over a 30-month period to produce a clinical description of the population they serve. Visual acuity was found using a range of assessments such that each child was assessed using an appropriate tool. The author also used assessments for visual fields, evaluations from a developmental pediatrician, and parent interviews to gather

information about each child's visual behavior, acuity, cognitive functioning, and social behavior (Dutton et al., 1996).

Keeping in mind that the children referred to the clinic were those with conditions that were difficult to diagnose, or with unusual visual behavior for which a plan of action was needed, 90 of 130 children (69%) seen at the clinic during the time frame in question had a vision difficulty that was attributed to damage to the cerebral cortex – they showed evidence of having cortical visual impairment. Because the children referred to this clinic had complex and difficult-to-diagnose conditions, this percentage of children with CVI may be slightly higher than average rate compared with studies of the general population of visually impaired children (Dutton et al., 1996). The children in this study all had vision issues tending toward the more severe. The findings from this study, obtained through interviews with parents and consultation with each participant including a 30-60 minute vision assessment and observation period, suggest that children with CVI have impairment of recognition, orientation, depth perception, perception of movement, and simultaneous perception, characteristics that are now frequently used to describe – or diagnose - CVI. The combination of observation and parent interviews yields information that can be checked for consistency, adding to reliability of the conclusions. Dutton and his colleagues, referring readers to other studies about CVI, describe the importance of recognizing that CVI is not static, and that children with CVI may develop sufficient vision skills to read print, if provided with targeted intervention. At the time this study was summarized for the article, they suggested a need for more specific assessment tools (rather than observation without consideration for specific

characteristics) and conditions under which the assessments will be used, and development of appropriate interventions to enhance visual recovery in children with CVI. This study is limited to a very specific population and therefore the results should not be applied to other groups. The authors used the information they gathered from this study to develop a prospective questioning strategy, so future studies would not rely solely on information volunteered by parents and caregivers and unguided observation.

Like Dutton and his colleagues, Hatton and her associates sought to answer questions about age of diagnosis and referral for children with severe, uncorrectable visual conditions, and what conditions were most prevalent at the time, as well as the characteristics of those conditions (Hatton et al., 2007). Intake coordinators at each participating clinic, who had expertise in early childhood VI, invited newly referred families to participate in the registry and obtained written consent. Data were recorded on a specific data collection form, using information collected via parent interviews and record review. The study included only children ages birth to 39 months, with significant VI at the time they were referred to specialized programs for children with VI. Twenty-nine states reported data to the registry, totaling 2155 children within the age range specified. Data regarding age at diagnosis was available for 1979 participants, and findings indicated that diagnoses were made for children with structural defects at the youngest age (mean age 1.5 months) and CVI was diagnosed much later (mean age 7.6 months). On average, children were not referred for intervention until 4.5 months following diagnosis, and there was an average of one month between referral and entry into specialized programs for

children with VI. Age of referral varied, however, based on the condition diagnosed. Only the primary visual condition from each child's ophthalmologic report was used for those children with multiple diagnoses, and the most prevalent visual condition in the sample was CVI, at 23.6% of the participants (n=509). Another important finding of this study of data is the percentage of participants for whom legal blindness status was unknown or missing. The status of Legal Blindness directly impacts the provision of specialized services and resources, and the status was unavailable for 36% of this study's sample, in part due to the acuity criteria required for classification as legally blind at the time, paired with the difficulty in obtaining acuity scores for infants and toddlers.

This study may suffer from sampling bias, because the study sample included only volunteers, and included just 29 of 50 states (Hatton et al., 2007). Also, the study only included infants and toddlers diagnosed and in programs before 39 months old – it did not include children that were not diagnosed, who acquired VI later, where VI was not suspected due to lessened severity, or where families declined participation. This greatly reduces generalizability of findings for several reasons: speed of diagnosis and service implementation may vary between states and would change statistics on obtaining diagnosis and services; children who were not yet diagnosed could not be included and they may have had different experiences (and the addition of these children may change prevalence and diagnostic data); and agencies that were responsible for data collection did not provide data about the number of families that declined participation, nor their reasons for refusal. In addition, the data entered into the registry was done without supervision of the

researchers, and some information (for example, parent refusals to participate), and some of the required information was not complete (the authors do not specify the information that was skipped). It is important to keep in mind that these data were a snapshot of causes of severe visual impairments in 2007, and that it is possible that the status of these things has changed since that time. Still, the prevalence of CVI in the sample is noteworthy, as are the statistics on mean age at diagnosis of CVI (7.6 months) and average time between diagnosis and referral, and referral and intervention (4.5 months and 1 month, respectively). This demonstrates time lost during the sensitive period of visual development.

A similar study by Khetpal and Donahue in 2007 looked at the etiology, prognosis, neurological, and ophthalmologic records for patients at a particular pediatric ophthalmology facility from 2002 to 2005. Ninety-eight patients were identified as having CVI, and their charts were examined to reveal the conditions with which the patients presented. The most common associated conditions were perinatal hypoxia (35%), prematurity (29%), hydrocephalus (19%), structural central nervous system (CNS) abnormalities (11%), and seizures (10%). Many of the children in the study had multiple associated conditions. Neurological conditions that co-existed were primarily seizures, cerebral palsy, PVL, hemiparesis, and hearing loss. The record review showed that during the time these patients were seen clinically, 57% showed some level of improvement in visual function. The study is limited by participant attrition, as 63 of the participants had no record of follow up at the clinic. While this limits the interpretation of data related to changes in functional use of vision, it does not affect the data regarding presentation or co-existing conditions.

The results of this study suggest that assessment for CVI may be performed on children with these commonly co-existing conditions to decrease the delay in diagnosing CVI.

To compile information about CVI, Huo et al. (1999) performed a record review of 7200 patients seen from 1979 to 1994 in a large pediatric ophthalmology clinic. At the time of referral to the clinic, CVI was diagnosed based upon vision loss without anterior pathway disease, and which greatly exceeded that which would be expected based on ocular examination. Children were excluded if poor visual function could be attributed to non-cortical deficits. Record review in this manner resulted in 170 cases of CVI for deeper review. Huo and his colleagues assessed the participants' functional vision on a scale as follows: *level 1* indicated light perception, only; *level* 2 meant that the participant could occasionally visually fixate on large or moving objects; *level* 3 if visual function was highly variable but moments of ability to see small objects or fixate on faces; *level* 4 if the participant reliably fixate on small objects or if visual acuity could be measured in the 20/400 to 20/200 range; *level 5* if the participant had reliable visual fixation or visual acuity not better than 20/50; and *level* 6 if the participant had completely normal sensory vision. This scale allowed functional vision improvement to be measured. Level One and Level Two combined included 70.59% of the study participants. Average functional vision score was 2.1, meaning that the population had very dysfunctional vision, and the 56.5% of original participants returned for follow-up and further evaluation after an average of 5.9 years (n=96). The follow-up group, a subset of the initial study group, averaged slightly more visually impaired than the original study population, as there was a

higher concentration of follow up subjects that had initially scored in the lower levels. The average improvement in functional vision for those that participated in the follow up was 0.9 levels using the scale developed by the researchers, indicating that almost one full level of improvement was attained over an average of 6 years (range of 3 months to 15 years); with 40% improving by one level and 20% improving by more than one level. Those participants who were more than three years old before they were first diagnosed averaged less than 0.5 levels of improvement, which indicates that later diagnosis may limit the amount of potential improvement in functional vision. Perinatal Hypoxia (lack of oxygen immediately after birth), Cerebral Vascular Incident (stroke), Meningitis/encephalitis, and acquired hypoxia accounted for over 58% of the participants' known causes of CVI. Seventy-five percent of participants also had noted neurological abnormalities, the most common being seizures which occurred in nearly 53% of participants. However, Cerebral Palsy (CP) was broken down into multiple, more specific categories, and total for those categories indicated that CP existed in over 58% of the population sample. There was no significant correlation between etiology of CVI and prognosis, but earlier diagnosis did seem to predict greater functional improvement. Results from this study suggest that professionals need to be aware of the conditions that tend to accompany CVI, and refer as appropriate and as early as possible. One limitation to the utility of the findings from this study is that the functional assessment scale was created retrospectively and contained little or no information on actual acuity measures. Although the study is older (1994), the longitudinal nature of the study adds value to its consideration for inclusion in this review, because data on visual

recovery is provided in a same-subject manner. It also clarifies the importance of early diagnosis and prompt referral for intervention for maximum visual skill recovery. Since the publishing of this article, a new assessment scale has been developed (Roman-Lantzy, 2018) and tested for reliability (Newcomb, 2009). Limitations of the Huo et al. (1994) study include that 9.4% of cases studied had unknown causes of CVI. The authors cite that the retrospective nature of the study may account for this in part, because there was no avenue to examine to find the possible causes for these cases, but also state that there is a need for further research into mechanisms causing CVI. Another limitation is that participants were obtained through partnership with an ophthalmology practice, which may skew the results for percentage of the participant population for co-morbidity of ophthalmological difficulties, and may not have included potential participants that have milder cases of CVI, or who had already shown improvement. Another drawback to the investigation presented here is that they used their own scale, developed for the study. This classification system was created to provide the researchers with rapid assessment results, but it provides limited data on acuity measures.

Summary and methodological issues. The aforementioned studies suggest that cases of CVI in children have a specific set of coexisting neurological conditions and etiologies. The studies also collectively suggest that children with CVI can recover some additional functional vision, and all agree that at the time of the studies, additional assessment procedures and interventions needed to be developed. The studies support the idea that earlier diagnosis and immediate intervention is vital to functional vision skills recovery. All of the studies had similar limitations. Each had

a limited or biased selection process, as the participants were volunteers or clients seen at very specific vision clinics, therefore participants included only those with suspected visual impairment whose guardians agreed to participate (Dutton et al., 1996; Hatton et al., 2007; Huo et al., 1999; Khetpal & Donahue, 2007). However, the studies were never intended to apply to the entire population of visually impaired persons. In the case of Dutton's study, the participants that were referred to that particular clinic were those that were difficult to diagnose from other clinics. The records that were reviewed did not always contain complete information and the data were often provided via caregiver input, which is a factor to consider when interpreting results. When considering the results of these studies, one must also consider the age of the studies, and that advances in assessment and intervention techniques may impact the relevance of such information. Obtaining parent perspectives and interpretations of child behavior, and understanding family experiences with CVI, diagnosis, and comorbidity may be key to understanding how to recognize and diagnose CVI in children earlier, thereby allowing for earlier intervention.

Families' Experiences with Disability and Diagnosis

Parent satisfaction with disclosure. While research has expanded in the arena of CVI assessment and diagnosis, there has not been research conducted with regard to parent perception of the assessment and diagnostic process when their children have CVI. As a result, this literature review includes examination of parent perspectives of the diagnostic process in children with other types of disabilities. In the process of searching the literature for studies concerned with family experiences

with diagnosis and disclosure, approximately 43 articles were located. Disability categories included primarily cerebral palsy, motor difficulties, specific learning difficulties, hearing impairment, and Autism. The number of articles was narrowed to exclude those that focused on intervention, rather than diagnosis. Eight articles remained for review.

In a study that examined parental satisfaction with disclosure of diagnosis in families of children with Cerebral Palsy (CP), Baird and associates (2000) interviewed parents of 107 children using semi-structured interviewing techniques that focused on structure, manner, and information that parents perceived, as well as asking the parents what they would have liked to happen at the time of diagnosis (Baird et al., 2000). The researchers included open-ended questions about disclosure of their child's diagnosis (including how the news was delivered, who was present during receipt of the news, the disposition (e.g. directness, friendliness, caring attitude) of the person who delivered the diagnosis, and the extent of information that-was provided at the time), problems the families were experiencing regarding caring for their child, hospital admissions, family circumstances, and other matters not reported in the published article about this study. The interviewer assigned an overall rating to the level of satisfaction based on the interview results, using a four-point scale, and checked to ensure that parents agreed with the score their interview received. Demographic information was also collected, and mothers completed two questionnaires to measure current levels of depression (Edinburgh Postnatal Depression Scale) and coping (Ways of Coping Inventory). The depression scores are presented as a continuum, rather than as above/below the cutoff point for

depression because the questionnaire cutoff points were developed for use within the first several weeks of the child's life, rather than a year or more after birth. During home visits, Baird examined each child and recorded level of severity of physical disability and intellectual impairment. Baird also extracted information such as birth weight, gestational age, cause of CP, and date of confirmation of diagnosis from the children's medical records, with parental permission.

The participants for this portion of the study were among the first 180 children to be included in a larger, longitudinal study that looked at joint problems in 660 children with CP that were referred to the study, conducted in the Southeast Thames region of the United Kingdom (Baird et al., 2000). Families were excluded if the child in question had a primary disability other than CP, if CP was acquired postnatally due to injury or illness, if the child was fostered, if the child had visible signs of difficulty at birth, and if the child had multiple significant learning difficulties. The part of this study included in this article aimed to determine guidelines for good practice in disclosing diagnosis of CP to parents. The researchers conducted the interviews in the parents' homes, discussing their experiences with disclosure of their child's disability. Three of the children's parents received no diagnosis, and three had not been told directly but had discovered the diagnosis by reading documentation about their children (one read their child's medical notes, one received a letter from the social services disability worker, and the other had read the diagnosis first in a letter from the local public housing authority). The results on satisfaction ratings were analyzed, along with characteristics of the relationships with the child and the family, using statistical tests (χ^2 and *t)* to determine which results

were classified as either satisfied or dissatisfied. During the course of the interview, many parents openly reported they were satisfied with the delivery of the diagnosis, but 86% (96 parents) also mentioned that they suspected that something was wrong before the diagnosis was explained to them. Forty-four mothers (41%) in the study were angry at the time, because the diagnosis came so long after they had expressed their concerns. Parents who received diagnoses later were less satisfied than those whose children were diagnosed earlier. In the cases of parents who had suspicions before the diagnosis was confirmed, further probing during the interview process clarified that dissatisfaction with disclosure was frequently expressed because physicians seemed to disbelieve the concerns that parents conveyed, or delay accompanied by the feeling that "everyone knew before I did" (Baird et al., 2000). During the course of this study, parents suggested that they could have benefitted from written information about their child's disability and services they could seek out. Based on these findings, the authors suggest that information that may be difficult for families to process should be explained tactfully, and extra time should be allowed so the family can ask questions. Professionals should be aware of the potential that parents will need the information repeated. For this reason, the authors suggest that providing information in writing may be helpful. Professionals should not pare down information to avoid repeating themselves, and information should be repeated as often as necessary to ensure parent comprehension. The most important findings for the purposes of this review are that parents' concerns should be taken more seriously, as in some cases the concerns may lead to diagnosis sooner (Baird et al., 2000). In addition, provision of more information for parents, preferably in

writing due to the difficult nature of disclosure, may help parents obtain services for their child more easily and quickly. This study had significant sampling bias, as many families were excluded (for example, if CP was acquired postnatally, if the child was fostered, and if the likelihood of problems was obvious at birth). Disclosure may affect these families differently. The sample is also limited to a very specific diagnosis, with specific criteria, in addition to the geographic limitations, which restricts the generalizability of the results.

Another interview-based, descriptive, qualitative study by Klein et al., (2011) examined parent perceptions of providers' methods for delivering diagnostic information and revealed a severe disconnect between parents' and professionals' perceptions. Twelve professionals from a preschool diagnostic assessment clinic completed open-ended surveys describing what information was typically provided to parents during a consultation. This information was used to guide interview questions for the second part of the study - nine couples whose children attended the clinic volunteered to participate in open-ended interviews to discuss their perceptions of the information they received. These interviews were conducted four to six weeks after their child's assessment, to enable parents to remember the assessment and its findings, and to allow time for families to implement recommendations that resulted from the diagnostic assessment. Interview transcripts were analyzed for common themes. An important finding is that parents' and professionals' perceptions of the information conveyed did not match. Parents indicated that not all of their needs were met during the professional consultation, while professionals believed that all information was conveyed successfully. The authors suggest that in lieu of

professionals asking parents if they have any further questions that professionals provide parents with a guideline of questions that parents frequently ask, since parents may not have sufficient information and be comfortable enough in the diagnostic situation to think of and/or ask questions (Klein et al., 2011). Results were synthesized to produce strategies for professionals to better support parents during such assessments. All parents mentioned the need for diagnostic results and information in writing. However, the authors suggest that parent readiness to receive information from professionals – meaning parents have the emotional resources and communication receptiveness- is individualized and begins with preassessment preparation to ensure parents are capable of processing the information provided. The authors do not provide detail about this preassessment procedure, but state that readiness varies based on many factors including age of child, diagnoses already received, and length of time parents had been seeking diagnosis; and the authors suggest that professionals take this readiness into account when delivering news and assessment findings. Parents expressed the need for information about their children's strengths and challenges, and words of encouragement. Parents also indicated a need for information that would prepare them in advance for the assessment process, such as questions that are typically asked during the process and the most common procedures that are used during the process. Parents commented that parent-friendly language should be used in lieu of medical terminology. To increase this study's credibility and dependability, the transcript of each interview was coded by each researcher involved independently, followed by team-based discussion of the codes to reach conclusions supported by all researchers. The

authors acknowledge the limited transferability of the results of this study, but it is useful in this literature review as similar needs regarding parent needs likely exist for parents of children with CVI.

Another study focused on parent satisfaction with the delivery of their child's diagnosis, and what factors influenced their satisfaction with this disclosure (Hasnat & Graves, 2000). Hasnat and Graves conducted semi-structured interviews (starting with a description of how disclosure occurred, followed by the parent rating satisfaction with the disclosure process and specific elements of the process (such as manner of the disclosing professional, the people present, the information received, and the subsequent follow up) on a five-point scale, from very satisfied to very dissatisfied) of 26 parents (representing 23 families) who volunteered out of a total attempt to recruit 80 parents. The researchers later reduced the satisfaction scale to three levels for ease of analysis. They analyzed results using chi-squared analysis, and Fisher's exact test was used to accommodate the small sample size. The data were categorized and tables were constructed to investigate possible relationships between variables. The researchers concluded that parent satisfaction with disclosure of a diagnosis was greater when they were provided with more information, and when the diagnosis was provided in a direct, understanding manner. They also found that parents who were overwhelmed by the amount of information provided were more satisfied with the professional level of care at diagnosis than parents who received an "adequate" amount of information, indicating that parents prefer that professionals provide as much information as possible, without being overly concerned that it is overwhelming or upsetting. Participants in the study suggest that the parents

themselves should decide the line between “adequate” and “too much”. Results are important to the discussion of CVI diagnosis because parents in general prefer too much information to not enough, so the diagnosis of CVI, or suspicion of CVI, should be shared as soon as it exists. For example, when parents suggest that their child “just does not seem to see well”, the professionals involved should address this immediately, rather than using a “wait and see” approach that can delay diagnosis and intervention (Roman-Lantzy, 2018).

This study had limitations, including a small sample size, which was not representative of all parents who have children with disabilities due to the selection process (though accommodated statistically using Fisher’s exact test). This process involved eighty mailed invitations to participate, of which only 26 accepted. This creates a sample of volunteers drawn from a small pool of candidates, all of whose children must have been treated at a very specific clinic (Monash Medical Center) before they were contacted to participate. In addition, both parents were available for three of the 23 interviews. For the remaining 20, only the mothers were interviewed. This potentially creates bias in the answers to interview questions, as mothers may have different interpretations of events than fathers. The disabilities of the children in the sample do not have the same distribution as children with disabilities in society in general (for example, 56.6% of respondents had children diagnosed with Autism), so the results have limited generalizability (Hasnat & Graves, 2000). The authors offered no speculation as to why the number of respondents was so low (29% of invited families).

Summary and methodological issues. The three studies reviewed above, which all examined parent perceptions on disclosure of diagnosis for their children, had similar study designs and similar flaws. All three used semi-structured interviews, and all found similar results in that parents express the desire to hear their child's diagnosis in a compassionate, but direct, manner. All three studies had limited sample sizes and limited sampling methods. Baird and associates (2000) limited their sample by including only those parents who had children with CP which was not obvious at birth, was not acquired postnatally, and if their children were not fostered. Klein et al. (2011) used a convenience sample of parents whose children attended a specific clinic. Hasnat and Graves (2000) used mailed invitations to invite parents to participate, and only 23 of 80 families agreed to participate. All of the studies had volunteers participate, which introduces bias, but cannot be avoided in studies of this type. None of these studies looked at parents of children with CVI, and no such studies were found when the literature search was conducted. The question remains, however, whether similar results would be found in cases where children have CVI.

Parental needs and satisfaction with information. In addition to methods of information disclosure, the information that professionals share when a family receives a diagnosis for a child, such as resources for more information, matters significantly. Some evidence suggests that parents need more from professionals than simply direct, understanding disclosure procedures. Stallard and Lenton (1992) describe parental satisfaction with information and services they received for their children with special needs. The study involved structured interviews/questionnaires consisting of 25 items (fixed choice questions, with opportunity to elaborate) with 41

parents (of 26 boys and 11 girls (32.5% of the total population of pre-school children identified as having special needs in the District); 21 children with severe learning difficulties, six with CP, two with both severe learning and motor conditions, and eight with other conditions (such as deafness, tuberous sclerosis, etc.) recruited via an open letter that was sent to the local playgroups in which the children were involved, requesting that parents volunteer for the study. Twelve impartial interviewers were obtained through the Community Health Council. Although overall satisfaction with services was generally high for questions such as convenience of appointment locations and times, the results from the interviews demonstrated a relatively high rate (44 percent) of dissatisfaction with the amount of information the families received about help available to them. Thirty-nine percent of parents in the study felt unsure of or that they were not given a clear explanation about the difficulties their child was facing, and 61 percent had not had the opportunity to discuss their concerns about their child's future needs with a professional. This relates to CVI because as a condition that is considered difficult to diagnose, the amount of information that parents receive about their child's vision difficulties – even if they do receive a diagnosis – is demonstrated to be lacking, and most parents feel they must do research on their own to find out what they should do (Jackel, Wilson, & Hartmann, 2010; Tallent, Tallent, & Bush, 2012). The survey results may not be generalizable to the population of families of children with disabilities, due to sampling bias, as self-selected participants may paint an overly positive or overly negative picture. Furthermore, answers to the survey questions allowed answers that were positive, negative, and neutral or unsure. The authors questioned whether the neutral answers

can be considered as dissatisfaction, as typically satisfied individuals are not unsure of their level of satisfaction, and given the tendency of consumer surveys to reflect a more positive perspective due to the "people pleasing" nature of respondents. If this is the case, a higher percentage of respondents are likely to have expressed dissatisfaction with the amount of information received, and a higher percentage may have felt unsure of or that they were not given a clear explanation about the difficulties their child was facing. To help counteract this, the researchers used unbiased interviewers for the families in the study. Although this study is older, it was included in this review because it does present an interesting point, applicable to families of children with CVI. Specifically, parents state they had not been presented with an opportunity to discuss concerns about their child's condition and future with a professional.

Information provided to parents by professionals was a key question in a study by Howie-Davies and McKenzie (2007), who examined information provided to parents of children diagnosed with specific diagnoses and non-specific learning disabilities. The study was conducted via postal questionnaire, and used both a between-subjects and a within-subjects design. Subjects were recruited from seven schools serving the population in question, and 47 of 273 parents participated. They were tasked to rate how much information they had received from each source, and how satisfied they were with the information at the time, using a Likert scale. Questionnaires included information about parent and family problems, pessimism, child characteristics, physical incapacitation, and sources of support. Both tools that were used had internal consistency, and adequate reliability and validity. Results

suggest that professionals should ensure that information they provide to families is relevant, timely, and accessible. Parents of children with specific diagnoses (such as Autism Spectrum Disorder and Cerebral Palsy) indicated they had access to a larger number of sources of information than parents of children with non-specific diagnoses, but there were no significant differences between the amount of information itself or their satisfaction with the information received. Parents who received specific diagnoses accessed information from support organizations more than parents of children with non-specific diagnoses did. Overall, however, the amount of information parents received from professionals, and parental expression of satisfaction with the information received from the professionals, was not significantly different for the two groups of parents. Perceived stress was not different between the two groups, and no relationship was found between information, satisfaction, and stress. Satisfaction with the information received by parents was low, a difficulty that may be explained by lack of professional knowledge about specific conditions. The authors provide no explanation as to why professionals did not know about specific conditions (for example, professionals thought of "learning disability" as meaning dyslexia or physical disability in a time when information about that distinction was widely available). This suggests that professionals must make efforts to increase their knowledge to provide more timely and accurate information to parents.

This study supports the need to have a definitive diagnosis of CVI, as having a diagnosis is associated with parents' seeking information from support organizations. The study by Howie-Davies and McKenzie (2007) had several weaknesses, including

the low response rate (17.2%), which is not uncommon with postal questionnaire-based research (Galvan, J.L., 2006, p. 51). This level of response leads to a conclusion that the results are biased and therefore not generalizable, particularly regarding participant self-selection. For example, parents who were feeling the most stress at the time of the study may have opted to not participate. This would lead the results to be slanted toward the less-stressed parent participants' perspectives.

Summary and methodological issues. The three studies regarding parental needs and satisfaction with the information received from professionals provide insight into the needs of parents. Although information on CVI and parent needs is not covered, the information is useful and may influence future study into that population. These three studies were quite different from one another. Stallard and Lenton (1992) used structured interviews and questionnaires, recruited via an open letter sent to area playgroups. This creates a limited geographical sample, and self-selected participants. Bailey et al. (1992) used postal surveys, and the high response rate should be noted. Although the survey was completed by volunteers, hence self-selection again, the high response rate and relatively diverse sample (when compared to the other studies reviewed here) results in reliable and valid results. The third study in this section, by Howie-Davies and McKenzie (2007) is much more recent than the other two studies in this section. Like Bailey et al., they used postal questionnaires. However, the response rate was very low leading to a limited and potentially biased sample. Another consideration regarding parent satisfaction and needs is communication, and how effective communication skills can affect parents' perceptions and understanding of medical information.

Communication. Another difficulty that parents of children with disabilities face is that often, physicians present information using medical terminology and jargon that families do not understand (Davies et al., 2003). This study included exploration of parents' experiences of care by pediatricians before and during the period of time when their children were being diagnosed with life-limiting conditions. Fourteen couples and sixteen mothers, all from the U.K. and predominantly working-class, were interviewed; limiting generalizability to other locations and those of other socio-economic states. Findings suggest that parents continue to feel that medical terminology is confusing and meaningless, and parents suggested that clinical practices such as sensitivity were more important when receiving difficult information. All parents had suspected something was wrong prior to their child's diagnosis, and felt that the receipt of diagnosis allowed the families to plan accordingly. For parents whose children's diagnoses were delayed, some reported a sense of relief when the diagnosis was determined, and some reported that the delay caused more stress and frustration than the diagnosis itself. In some cases presented by this study, insensitive care may lead to delays in diagnosis due to dismissal of parents' concerns, and it was only due to persistence that the families received a diagnosis at all. In some cases, delayed diagnosis of CVI can result in loss of opportunity to make visual progress (Baird et al., 2000; Davies et al., 2003; Roman-Lantzy, 2018). In other words, if parents of children who demonstrate characteristics of CVI are dismissed when they request a diagnosis or reason for their child's vision difficulties, this could result in a delay in services, thereby creating lost opportunity. The overall findings from this study indicate high parental satisfaction with

physicians who exhibit technical expertise, good communication, and human sympathy and understanding. Low parental satisfaction was associated with lack of these skills. The authors of this article offer no specific information about the interview structure other than they were "in depth". Though the sample is claimed to be representative of "family life" at the time in the U.K., the sampling method suggests otherwise, as it was obtained through invitational letter so subject to selection bias as it is possible that, for example, only parents that were not feeling stressed, or those with stories they felt the need to tell, participated.

Summary and methodological issues. Harnett et al. (2009) used a mixed-methods design including parents and professionals, in focus groups, exploratory interviews, and a postal questionnaire to research best practices for professionals. The summary article did not explain how data was analyzed, or what the distribution of parents and professionals was in phase II of their study. However, their sampling method yielded a large range of participants.

Parents of children with cortical visual impairment. Families of children with visual impairments need information about resources and interventions, and need professional services right away to understand how the child will learn about the world (Kenney, 2005). In the search for literature regarding the study of parents whose children have CVI, only one article was located. Jackel et al. (2010) summarized an anonymous, online survey of 80 parents of children with CVI. The survey questions consisted of four basic categories, including parents' suspicion of a vision problem, diagnosis and information provided to parents, etiology and visual inventory, and education and related services their children are receiving. Two

hundred twelve individuals responded to the survey requests, 80 of them parents. The 80 parent participants came from 32 states, had children of wide age ranges with different etiologies. The survey results indicated that 55% of the parent participants were the first to suspect that something was wrong with their child's vision. Parents reported that their children displayed visual behaviors that are typical for CVI, including difficulty with visually guided reach, light gazing/non-purposeful gaze, atypical reflexes, generally typical eye examinations, and difficulty with complex visual environments. Half of the children in the study were diagnosed with CVI within the first year of life, 35% were diagnosed between ages 1-3, 9% between the ages of 4-7, 5% were between ages 8-12 and one child was between ages 13-18. Most often (49%) the children were diagnosed by an ophthalmologist, but 17% of these children were first seen by a Teacher of the Visually Impaired (TVI) and suggestions were made to the parents to have the child evaluated by a medical professional. Sixteen percent of the children were diagnosed by a neurologist, and 10% were diagnosed by their parents before receiving an official diagnosis.

Parents were asked how much and what sort of information they received at the time of diagnosis. Forty-five percent indicated they received very little information about how they should proceed, 28% indicated they received "some" information, and 22% said they were given no information at all (Jackel et al., 2010). Books, pamphlets, website addresses, professional journal articles, and information about online courses were the most common types of information that were provided to parents. Fifty-one percent of the parents found the information to be helpful, but 26% indicated the information was not helpful at all. Approximately half of the

parents that participated in this survey indicated that they received no information except the diagnosis at the time, and 69% of the parents said they got information through their own research.

When parents were asked about educational placement of their children, 27% said they were receiving services through early intervention programs, 24% attended the local or neighborhood school, 16% were placed in a special education school, and 9% participated in a regional special education program (Jackel et al., 2010). The remainder of the children were in a non-public program, a school for those with VI, or were home schooled. When parents were asked about level of difficulty in getting services for their child with CVI, 20% replied that it was very difficult, 15% that it was somewhat difficult, and 18% reported that it was not difficult. Thirty five percent of parents reported that their children had not received any services. Eleven percent reported that the question did not apply for a variety of reasons: low understanding of VI, appropriate services are not being received; the child is "too young"; the child is not in public school, and the families were "still not getting appropriate accommodations". Parents reported that the services that were provided to their children included small classroom setting, uncluttered work areas, lighting accommodations, preferential seating, large print, audio books, quiet learning areas, and real-life objects and examples. Parents reported that the primary reason for the difficulty they experienced in obtaining appropriate accommodations for their children with CVI was professionals' lack of understanding, knowledge, and training about CVI. Some parents responded that professionals indicated their children "see well enough", do not have "true VI" because their eye exams are typical, and that the

CVI would resolve on its own with no targeted intervention. Professionals also reportedly disregarded the VI due to other, concurrent disabilities. These statements indicate a need for increased education and awareness in the professional community.

This study is the only study that was found, which addresses the topic at hand (parent reactions to CVI and the process of its diagnosis) directly. This study has heavily influenced the research questions that I am exploring. One limitation of the aforementioned study is that the sampling method (an emailed invitation was sent to participants on electronic bulletin boards about CVI) created a biased, convenience sample (Jackel et al., 2010). This sample represents parents who felt empowered and able to actively seek out information and participate in a community of people that have similar concerns about their children. This means that parents who may not have access to the internet, or that may not feel empowered to seek out information independently, for example, were not included. The results could reflect very different conclusions had parents without these resources and/or abilities been able to participate. An additional source of sampling bias is that only 32 states were represented.

A non-rigorous, non-study, family perspective is provided in a book entitled *Little Bear Sees.* The parents and a grandparent of a child who experienced widespread brain injury due to Hypoxic Ischemic Encephalopathy (meaning at some point before birth, he did not receive enough oxygen) wrote it. At six weeks old, the child did not look at his parents; at four months old, the family took him to the ophthalmologist and were told he could see light but there was no way to know what else he could see and no way to help him improve. At seven months old, the family

connected with a teacher of the visually impaired who suspected CVI. The parents did an internet search to find out more, and the authors write, "securing knowledge, accurate information, and personal and professional support can seem nearly impossible when your child has [CVI]. Many doctors seem to know very little about CVI, and resources that do exist are not written for parents," (Tallent, Tallent, & Bush, 2012, p9). As of the publishing of the book, the child about whom the book was written had yet to receive an official diagnosis of CVI. However, the family believes the information they found on their own, and the interventions they therefore implemented, have helped their child learn to see.

The book is not a summary from a study; rather, it is an example of parent perspectives on CVI and the difficulty that families can experience in getting diagnosis and services to treat it (Tallent et al., 2012). As such, the material included is not held to the same standards as formal research is, and it is greatly biased, strictly from the parents' point of view. However, it does tell the story of a family's journey through the medical and early intervention system and the obstacles they faced in getting their son's visual difficulties addressed. The book is written for other parents who find themselves with questions about CVI and what to do when they discover their child might have the condition.

Summary and methodological issues. The study conducted by Jackel et al. (2010) consisted of an anonymous, email-invitational online survey of parents who have children with CVI, yielding a biased, convenience sample. However, the sample did consist of parents from 32 states, whose children with CVI had a range of

ages and etiologies, which improves the variation of the study sample. It is the only study that was located for this literature review, and though it has flaws, it is an important beginning to studies of this type including the population of families affected by CVI. The other publication included within this section is not a study, but one family's perspective about their experiences with their child who has CVI (Tallent et al., 2012).

Summary

The studies presented in this chapter discuss conditions associated with CVI, as well as prevalence and characteristics of CVI. All of the studies had limited sampling methods (mainly due to volunteering participants), so bias is present in them all (Dutton et al., 1996; Hatton et al., 2007; Khetpal & Donahue, 2007) though some are less affected than others (Huo et al., 1999). The studies found that the most prevalent visual difficulty in the populations studied was CVI. The studies that reviewed associated conditions (including perinatal hypoxia, infant stroke, hydrocephalus, CNS abnormalities, cerebral palsy, periventricular leukomalacia, hemiparesis, and seizures) had similar conclusions (Khetpal & Donahue, 2007; Huo et al., 1999). Those that examined prognosis agreed that earlier detection and diagnosis improves outcomes for individuals affected by CVI (Huo et al., 1999; Khetpal & Donahue, 2007).

Also, three studies are presented related to parent satisfaction with *disclosure* of diagnosis, for conditions other than CVI. All three studies used interviews, and all but one (Baird et al., 2000) had small sample sizes. The conclusions were very

similar – professionals should take parents' concerns seriously, professionals should consider that providing families with all of the information they have may not fully meet the needs of the families, and that parents are more satisfied with the disclosure of their child's diagnosis when the professionals provide them with as much information as possible (Baird et al., 2000; Hasnat & Graves, 2000; Klein et al., 2011).

Three additional studies examined parent satisfaction with *information* with which they were provided (Bailey et al., 1992; Howie-Davies & McKenzie; Stallard & Lenton, 1992). Stallard and Lenton used structured interviews and questionnaires and found that parents felt the need to discuss their child's condition and prognosis with a professional, and felt unsure about the information they received. Bailey and associates found that the needs of fathers and mothers – with regard to information provided – did not always match. This study had a large, widespread sample and used the well-established Family Needs Survey. The study conducted by Howie-Davies and McKenzie used a postal questionnaire to assess the level of satisfaction that parents felt about the information they had received. The results indicated that parents of children with specific diagnoses had access to a larger number of sources of information, and accessed support organizations, when compared with parents of children who had no diagnoses.

Delays in diagnosing disabilities in children create many negative consequences, especially delay in starting Early Intervention Services, resulting in loss of developmental potential (Halfon et al., 2004). Typically, parents are first to

suspect that something is “not right,” and often there is a period of time where physicians do not take their concerns seriously, they suggest a “wait-and-see” approach, suggest the child will “grow out of it”, or they do not see the delays in development as the parents see them (Addison, 2003; Davies, et al., 2003; Peters, 2010). Davies et al. also found that although the majority of pediatricians in their study were sensitive overall, physicians that were responsible for the delayed diagnoses for their study participants did not apologize, persisted in treating the families dismissively, and continued to give parents the impression that the physicians were insensitive and uncaring.

Knowing what CVI is and having it diagnosed are key aspects of providing intervention. This intervention may assist children in taking in and comprehending greater amounts of information from their environment, increasing learning in all developmental domains. Until diagnosis, intervention cannot be properly provided, which delays visual skill development thereby delaying information absorption and evaluation from the environment, ability to move as guided by vision, and acquisition of visually ascertained social skills.

There currently exists very little information about parental perspectives on the diagnosis disclosure process for children with CVI, despite its increased prevalence. Many professionals are unaware of any procedures for assessing a child for CVI and providing intervention when it is diagnosed. It is unclear whether professionals are fully aware that a diagnosis of any of the conditions associated with CVI could be a precursor to a diagnosis of CVI itself; that the existence of an associated condition should prompt the professionals to check for CVI. It is also

unclear whether professionals are aware that earlier diagnosis of CVI is paramount to visual recovery, and that apparent visual difficulties should not be ignored. This literature review examined journal articles and books related to disclosure for other disabilities, from parents' perspectives; the difficulties faced by parents when a diagnosis has not yet been determined for their child's condition; the disconnect between parents and professionals with regard to information sharing; and that parents desire an increased level of information sharing from professionals to help them better understand and help their children develop. Further examination of such things is needed with regard to CVI, to find out what the status is of services provided by professionals, and what parents are experiencing when their child has CVI, whether diagnosed or undiagnosed. Research is needed to discover what percentage of children currently display characteristics of CVI but have no diagnosis, and the reasons for this phenomenon. Furthermore, information should be obtained about how much information parents are receiving, and from what sources, when they seek out answers about CVI.

Research Questions

To address concerns about timely diagnosis and referral for children with suspected CVI, the following research questions are addressed:

1. What are parents' experiences in seeking a diagnosis for their child's suspected vision challenges?
2. What needs do parents recall related to information and supports while seeking a diagnosis for their child's suspected vision challenges?

3. What kind of information is offered or readily available to parents upon diagnosis of CVI?

CHAPTER III

RESEARCH METHODOLOGY

This study used qualitative methods to explore parent's perspectives on the process of receiving a diagnosis of CVI for their children. I begin with a brief description of qualitative inquiry and explain how this method of study is appropriate when researching topics such as parent experiences with disability in their children. After restating the proposed research questions, I provide a description of the recruitment process, data collection, and data storage and analysis. I conclude this chapter with a discussion of ethical and personal considerations as related to this study, specifically.

Qualitative Inquiry

Qualitative researchers use rigorous sampling, data collection, analysis, and interpretation to shed light on a particular issue or seek out understandings of phenomena or insiders' experiences (Forman, Creswell, Damschroder, Kowalski, & Krein, 2008). Qualitative researchers typically follow an inductive approach to obtaining information – inferring general ideas and principles from the specifics that are revealed during the course of data collection (Hanson, Balmer, & Giardino, 2011). Qualitative inquiry is often pursued because it offers flexibility that is not typically offered by quantitative research, allowing the researcher to explore unexpected findings more easily and delve deeply and holistically into the chosen topic. Qualitative research projects may change or develop as data is gathered, and

procedures may change or be influenced by the data as it is collected (Creswell & Poth, 2017; Maxwell, 2005).

Qualitative Inquiry and Parent Perspectives

According to Creswell and Poth (2017), qualitative researchers collect data in natural settings that are sensitive to the individual participants, resulting in a final presentation that includes the "voices of participants, the reflexivity of the researcher, and a complex description and interpretation of the problem, and it extends the literature or signals a call for action" (p.37). By this definition, qualitative research methods are well suited to clarify what families experience when seeking a diagnosis for their children who demonstrate characteristics of CVI. Qualitative research methodology has been selected for use in this study because it is particularly useful for gaining an "emic" or insider's perspective. Using qualitative methods allowed me to focus on what meaning the participants assigned to their experiences. Moreover, since this was a relatively understudied topic at the time, qualitative research allowed exploration of multiple facets of the issue.

Since the purpose of this study was to explore parents' perspectives on the process of getting CVI recognized and diagnosed, I selected case study design. My goal was to study multiple cases of this phenomenon, which indicated the need for a multiple-case study format. This methodology required the exploration of multiple cases through in-depth, detailed data collection that included multiple sources of information (such as interviews, documents, and reports). Then, I produced descriptions and themes of said cases, (Creswell & Poth, 2017). This exploration

allowed for theoretical replication of the cases, which strengthened the findings (Yin, 2014).

A case study is one form of qualitative methodology that allows researcher to investigate a phenomenon through the perspective of the participant (Taylor & Thomas-Gregory, 2015). Case studies are particularly useful when asking "how" and "why" questions, to help understand social phenomena (Yin, 2014). Considering case study to be a type of design, or an approach, allows the researcher to collect detailed data from multiple sources, to provide an in-depth description of a case (or cases) within the overall setting of the case (Creswell & Poth, 2017). A multiple case study, wherein more than one case is used within the same study, allows the researcher to analyze more than one source of data regarding each case, and to describe a particular issue within the context of those cases, often considered more compelling due to increased evidence (Creswell & Poth, 2017). This allows the researcher the possibility of replicating, either theoretically or directly, which strengthens the findings further than a single-case study would allow (Yin, 2014).

Focus Group

A focus group was convened in July 2016 to help shape the research questions I wanted to explore in this study. The focus group took place at the American Conference on Pediatric Cortical Visual Impairment and included ten parent volunteers. Each volunteer signed a consent form to allow me to record the focus group discussion and share the results of the study. Each participant had the opportunity to share their experiences with getting a diagnosis of CVI (for their

respective children) with the group, and once everyone had addressed the focus group question ("tell your CVI diagnosis story"), the group was ready to conclude. The participants shared many details during their individual chance to speak, and each individual offered emotional support to the others during the conversation while being respectful of one another's time to speak.

The results, including notes and audio recording taken during the focus group meeting, varied greatly depending on where each family lived, the circumstances of the children's births and early life experiences, and with what professionals each was connected. The information on timing, rural versus urban setting, and circumstances for the child and family were summarized in a table to easily compare each individual's experience (see Appendix I). Families who connected early with knowledgeable professionals received diagnosis as early as seven days into life. Most families, however, encountered professionals who did not know what to do, who instructed the family to wait and see what happens, or who said nothing could be done. For most participants, the diagnosis came months or years after they first mentioned concerns about their child's vision. The results of the session helped guide the development of interview questions for this study. For example, the results from my request that each participant "tell your CVI diagnosis story" confirmed that there is a discrepancy between noted signs of CVI and its diagnosis. However, in the focus group setting, participants told stories quickly and without opportunity to follow-up for clarification due to time constraints. In addition, the focus group included parents of children who were nearer to adulthood. For this study, I limited the age range of the children who had CVI to focus on recent parent experiences. Compared to the

focus group, the multiple case study design used here allowed deeper investigation into specific cases and obtain multiple sources of data about each. This approach also allowed me to collect information about what was helpful and what was not, since there was ample opportunity for expanding upon the participants' diagnosis stories.

Purpose and Questions of Current Study

This study was an exploration of parents' experiences related to their child who had been diagnosed with or who demonstrated characteristics of CVI. The purpose of this research was to explore parents' experiences with CVI and the process of getting it recognized and diagnosed. As such, using a multiple-case study design and collecting "stories" was a way to obtain detailed, personal experiences relayed by those who directly experienced the situation in question. Based on my real-world experiences, informal conversations with parents and other professionals, and discussion during conferences and the focus group described above, the following research questions were developed to guide this research:

1. What are parents' experiences in seeking a diagnosis for their child's suspected vision challenges?
2. What needs do parents recall related to information and supports while seeking a diagnosis for their child's suspected vision challenges?
3. What kind of information is offered or readily available to parents upon diagnosis of CVI?

Procedures

This section describes the study procedures, including participant sampling and setting. A discussion of data collection methods, confidentiality, and data analysis follows.

Recruitment and selection of families. In qualitative research, samples are often purposeful, and participants are selected based on the variables that are relevant to the study (Bogdan & Biklen, 1998). Participants in this study included parent volunteers referred by the Maryland/DC Deaf Blind Project, as well as additional participants who were referred by the first volunteers using snowball sampling techniques. Eleven volunteers were recruited for this study and included participants who had children diagnosed with CVI within the last 5 years and who were between the ages of 6 months and 6 years. The five-year diagnosis requirement allowed parents to have their story in recent memory. In addition, the selection of participants was partially limited to those I could most easily access, within approximately a 250-mile radius of my residence (for in-person interviews) or available for phone interviews if farther away. Initial contact with each family occurred when, after receiving my contact information from a referral source, the volunteers contacted me directly. This initial contact was via email for every participant. My response to each volunteer began with a summary of the purpose of this study, the parameters under which participants would be included, and requested a confirmation of the participant's interest. Once interest was confirmed, a recruitment package containing formal consent forms and detailing the study's purpose, method, participant roles, and

expected benefits to and potential drawbacks of participation was sent to each respondent within a week of initial contact.

The parent participants included 11 mothers and three fathers of children between three and six years of age who had received a diagnosis of CVI. Three of the children's parents chose to participate in the interview as couples. Nine of the families had children in addition to the child with CVI: five with older children, five with younger children, and two who were expecting a baby. Nine of the families lived in suburban areas and two in rural areas.

These parents represent a fairly homogenous group in terms of ethnic backgrounds (including only two parents/couples who self-identified as "black", and the remaining nine parents/couples self-identified as "white"); marital status (one parent self-identified as "single", one as "divorced", and nine as "married"); educational level, which was used as a proxy for income level (7 parents/couples self-identified as having a master's degree or higher, while 4 parents/couples self-identified as having bachelor's); and geographical residence (nine families lived in suburban areas, one in a rural area, and one lived in an urban area). All eleven families reported that they had no other children with documented disabilities living in the home. In terms of age, three parents/couples were between the ages of 30 and 39, seven parents/couples were between the ages of 40 and 49, and one parent was between the ages of 50 and 59. No couples who participated included individual parents who crossed over multiple age ranges. Only one couple had only one child (two, if the family who was pregnant with their second child is counted as having one child), though two other families had only older children, in addition to the child with

CVI, who were not part of the household. Most families were from a limited geographical area, with six of the eleven parent participants residing in Maryland, and two in Pennsylvania (one of whom had just moved from Maryland). The three participants who resided outside of MD and PA were referred via an email recipient list, from the organizer of a weekly, international teleconference for parents of children with CVI. These volunteers represent a sample of parents who have the resources to network with other families and to reach out to state organizations that provide assistance to families. Every parent participant for this study had educated themselves at least a little about CVI – and most had read a lot (including books that were intended for educators and medical professionals) and/or attended workshops intended for education professionals. The present study is very unlikely to have been representative of the overall population of parents whose children have CVI.

All of the children had diagnoses in addition to CVI, which are not specified by family here due to their identifying nature. Four of the children had a hypoxic event close to birth, three of the children had genetic abnormalities (such as mutations), and two of the children were born prematurely with complications. One child had a genetic mutation *and* was born prematurely with complications. Each of the families who participated in this study had means and ability to seek out medical care and services for their child, and to continue advocating for their child when finding answers was difficult.

The children discussed by these informants included four girls and seven boys. In most cases, their parents noticed something was not typical about their vision before the age of one year except for two cases in which parents made this

observation that before age two. The length of time between parents' first concern about vision and the diagnosis of CVI varied greatly, ranging from no time between concerns and diagnosis to five years. Diagnosis occurred at varying points in the children's lives (one at 1 week of age, one at 8 months, five at approximately age 2-3 years, three at approximately age 3-4 years, and one was over the age of five.) Due to the low incidence of several of the children's diagnosed conditions, to maintain confidentiality the information provided for each specific participant is generic, and the parents' and children's names have been changed. The information includes: number of other children in the family, ages parents "first" noticed a difference in their child's vision, ages parents received diagnosis of CVI for their children, and the possible associated condition(s) the children each have. This information is summarized in Table 1.

Table 1: Parent Participant Information

Parent(s)	**Child**	**Other Children (compared by age)**	**Age when *Adult Noticed* CVI Characteristics**	**Age @ *Diagnosis***	**Possible Associated Condition(s)**
Marla	Gwen	1 older 1 younger	2 years (Other Service Provider first noted)	~4 years	Hypoxia
Lisa	Bobby	1 older	<1 month	~2 years	Genetic
Tracey	Phoebe	1 younger 1 on the way	>7 months Teacher noted ~3 years	~3 years	Genetic
Veronica	Brady	4 living in the home, older	Immediately	3-4 years	Hypoxia
Lauren & George	Andrew	1 younger	Immediately	~2 years	Premature & Genetic
Raina & Charles	Harrison	1 younger	6 months; (Teacher noted at ~5)	5 ½ years	Genetic

Nadia & Boyd	Paul	1 on the way	<2 years	~3 years	Hypoxia & Genetic
Bethany	Jonathan	None	Immediately	1 week	Hypoxia
Claire	Dylan	1 older	~1 year	2 ½ years	Very premature
Jennifer	Gail	1 younger	<8 months	~8 months	2 weeks premature
Sharon	Mary	2 much older	1-2 years	2 ½ years	Genetic

A secondary sampling strategy was snowball sampling, which involved identifying cases of interest from participants who knew other people who would be interested in participating as cases themselves (Creswell & Poth, 2017). In other words, cases were supplied based on referrals from other cases. A few additional participants were located through a mass email request, initiated by a participant and sent to the members of a weekly CVI teleconference.

Setting, interview questions, and timing. Qualitative inquiry is traditionally conducted in natural settings (Creswell & Poth, 2017). Face-to-face interviews for this study were conducted in participants' homes, a location of the participant's choosing, so they were likely to be most comfortable to talk openly about their experiences. When face-to-face initial interviews were not possible due to timing or distance, telephone interviews were used. The telephone was also used for follow-up interviews. To focus and guide the interviews and ensure the required data was collected, I used previously-developed open-ended lead questions that were followed with prompts wherever necessary. Lead questions and prompts are found in Appendix F.

Interviewing as a methodology involves orally asking participants questions to facilitate the collection of detailed data, which may be personal in nature, and is often

high in quality and contains deep levels of detail (Block & Erskine, 2012). Eight of the initial interviews for this study were conducted face-to-face and three were by phone, with follow-up interviews all via telephone. In an effort to increase rapport and comfort for the interview participants, I fully introduced myself including my background, interest and history in working with children with CVI, and asked participants if they had any questions before we started each initial interview.

Although interviews were not time limited, I watched for signs of fatigue and distress. The initial family interviews lasted between 31 and 106 minutes. For those interviews that ran longer than 45 minutes, I checked in to ensure the participants wished to continue. All interviews were digitally recorded, with permission from all participants. This allowed me to prepare verbatim transcripts of each interview for purposes of data analysis.

Follow up interviews were scheduled after each initial interview was transcribed and reviewed, so that clarification could be obtained whenever necessary. Specific questions for follow up interviews were based on information from previous interviews, and were primarily used to clarify comments made during the previous interviews and to seek out details of any relevant happenings since each initial interview. No cases required that the second interview be a continuation of the initial interview. In cases where both parents of a child wished to participate, interviews were conducted together and counted as one case. Three cases involved interviews of parents who interviewed as couples.

Follow up interviews were also used as an opportunity to conduct "member checks" to verify that my interpretation of data from initial interviews was accurate.

During this process, I summarized the information I received during the previous interviews and asked participants if I had understood and interpreted the data correctly, and that I had included all of the relevant information from the previous interviews. Two participants had no corrections or additions to request, and they agreed that the salient points I had coded were their most important points. Three participants had additional comments, which expanded upon their original responses to the interview questions, only. Six participants had additional comments as well as new information about things that had occurred since my first meeting with them, such as appointments and meetings. No participants disagreed with the areas coded as important points, but they did expand upon the information they had provided during the initial interview.

Secondary data sources. In an effort to triangulate the data for the study, I sought out additional sources of data including documentation from parents and interviews of professionals in the field of visual impairment. I asked the parents of the five school-aged children who did in-person interviews to provide medical and/or educational records if possible, to corroborate their understanding of educational findings and medical results from appointments, as appropriate. I asked those five families so that I would receive educational records for children who were eligible to receive vision services as documented on an IEP, limited to parents who had met me in person so that we had an established rapport. Since I asked families to provide records directly, it was not necessary to obtain additional HIPPA permissions and connect with each physician's office.

Second, I conducted interviews with two physicians in the MD/DC/VA area, who are qualified to diagnose CVI, in addition to the interviews conducted with parent participants. I recruited these physicians to participate through recommendations from parents, using the recruitment letter found in Appendix C. Since many of the parent participants are from a limited geographic area, they often recommended the same vision-related physicians as potential participants in this study. Both physicians who participated had experience diagnosing CVI, and neither was familiar with me directly. One of the physicians had an urban, hospital-based -, and the other was part of a suburban practice with a number of other eye doctors. Each of these two participants had been diagnosing CVI for more than 15 years, and both were affiliated with universities as well. One of them was a pediatric neuro-ophthalmologist and the other was a pediatric ophthalmologist. Although these physicians were located through parent referrals, they did not have any knowledge of which families participated in the study, so there was no discussion of specific children during the interviews with the physicians.

Third, I asked families to refer me to Teachers of the Visually Impaired (TVIs), with whom I then used the recruitment letter found in Appendix C. Two TVIs were referred by parent informants, and one of them was referred by multiple families. Another had suggested she was interested, but did not respond to several attempts to gain consent via email. Both TVIs who participated knew me distantly due to my involvement with the MD/DC DeafBlind Project, and were school-based and served multiple children with CVI on their caseloads. All professional interviews

took place in-person. Interview questions for these professionals may be found in Appendices G and H.

Because parents who agreed to participate may have had concerns about confidentiality, each parent participant received Statements of Confidentiality when they received the formal consent letters, all of which were approved by the Institutional Review Board (IRB) at the University of Maryland. Each interview began with a brief review of The Statement of Confidentiality and the Letter of Consent, as reassurance that the individual's confidentiality will be protected, and that participants will not be identified in the raw data or write-up of the research.

Interviews were digitally recorded and transcribed, in addition to notes taken during each interview for the purpose of adding details, inserting observer comments into transcripts, and to note probes that I determined valuable while a participant was speaking. I also noted if a participant used a particular phrase or theme repeatedly, or if I recalled that a participant used similar phrasing as a different participant, or if there were changes in inflection, cadence, and tone of voice.

Methodological Considerations

Trustworthiness and credibility. To ensure that the data for a qualitative study is collected, analyzed, and understood correctly, the information retrieved must be validated (Creswell & Poth, 2017; Maxwell, 2005). *Triangulation* is a strategy that is used in qualitative research which allows the researcher to use multiple methods of data collection and multiple sources of data about the same phenomenon or issue that is being investigated (Creswell & Poth, 2017; Maxwell, 2005). This process permits

a broader understanding of the issue, and helps address threats to validity in that consistency in data may be determined (Yin, 2014). In this study, data was triangulated across both data sources and methods: specifically through interviews with parents, physicians, teachers, and record review of Individualized Education Plans (IEPs), reports from physicians, and miscellaneous documents provided by the parents.

Another method of verifying the data resulting from interviews is through *member checks*. This is the process by which transcripts and/or analysis and conclusions that were drawn from interactions between researcher and participants is confirmed by the participants as correctly understood (Creswell & Poth, 2017; Maxwell, 2005). I did member checks by sharing my initial conclusions with each participant during follow-up interviews and subsequent communication (brief telephone conversations and/or email to confirm my understanding of ambiguous content) with parents. These member checks helped to ensure that the interviews accurately reflected parent meaning, that points the participants wanted to stress received the appropriate amount of attention, and that any misconceptions were corrected. Providing a summary and my resulting conclusions to each participant before the follow-up interview gave each participant a chance to clarify information and elaborate as needed.

Limitations. As demonstrated by their association with the MD/DC Deaf Blind Project and/or the parent support teleconference, the parents who participated in this study have already accessed various resources about CVI, and may tend to actively seek out resources and support from their community. As a result, this

sample may be biased because these parents are educated about CVI and they have likely networked with other parents and professionals. The results from this study cannot be generalized due to the purposeful selection strategy used, and the qualitative nature of the results. Additionally, for the three cases where couples participated together, their responses were very likely affected by the presence and input of one another.

Positioning of the researcher. As the researcher, I am well versed in CVI and the CVI Range. I was responsible for the Demonstration Classroom for the MD/DC Deaf-Blind Project for five years, and as a result of my work in the field of Deaf Blindness, I was the Onsite Deaf-Blind Project Coordinator at that school. I have served many students with multiple intense needs for over 12 years, and I consult and train about CVI and Instructional Strategies for Students with Deaf-Blindness within schools and at continuing education workshops. I have been granted the Perkins-Roman endorsement in the CVI Range, which some parents and professionals regard as a key indicator of a professional's qualifications to work with children who have CVI. My concern for timely diagnosis is the direct result of my work with students, many of whom demonstrated the characteristics but had no diagnosis of CVI. These experiences may have influenced my interpretation of participant responses, requiring that I take precautions in collecting and interpreting data such as performing member checks and using data sources in addition to parent interviews. I also chose not to interview any parents whose students I was teaching during the data collection phase of the study, to avoid conflicts of interest.

During the investigation, I noticed potential difficulties in keeping my role as the researcher separate from my role as a professional who is well-versed in CVI. Several times I caught myself tempted to provide guidance, information, or opinions about things that parents described. In a few cases, I guided the focus of interviews back to the interview questions and research questions, choosing to answer parents' questions about information that I had after the study/data collection period was completed. As a professional who cares a great deal for this group of children and their families, I felt it was important to provide families with as much information as possible, while trying to maintain a professional role and distance from the participants. I made every effort to not influence the comments or perceptions of the informants during the interview process, while still employing active listening techniques such as establishing rapport, using language to reassure participants that I was listening and understanding, and paraphrasing participants' answers to interview questions back to them.

Although I present myself as a professional in this field, I believe the families that I interviewed felt comfortable enough with me to share their stories. Likewise, the professionals who shared their insights with me seemed to feel comfortable doing so, professional-to-professional. The physicians both thanked me for the work I was doing, and said that they were encouraged that someone was trying to direct attention toward this issue.

Data management. The open-ended interview process resulted in large amounts of raw data. I collected digital interview recordings, notes taken during the interviews, and any additional paperwork and materials that participants wished to

share, including electronic documents. I am the only person who conducted interviews, and the raw digital information was uploaded to a transcription service with guaranteed confidentiality. The raw data was stored in three locations within my home network so that there was a current, working copy on my desktop computer, a backup on an additional hard drive within the same computer, and an additional set of backups that were created and dated each day that changes are made within the data. The dated set of backups were be stored on a portable hard drive, which is kept in my home office. All copies are password protected, and my network administrator and I will be the only individuals who have access. The network administrator has also signed a Statement of Confidentiality, and has access only to the assigned case names, not the participants' identifying information.

Data analysis. In qualitative research, data collection and analysis often overlap so that any questions which emerge during the process of data collection can be pursued in further data collection. Although the research questions in this multiple case analysis were guided by the literature review, specific concepts emerged at times, which required clarification as the data was collected, transcribed, reviewed, coded, and analyzed. By sharing the summaries and resulting conclusions of each interview with the interviewee, member checks were performed and any points of clarification exercised from such exchanges. This feedback was added to the data and handled as such. Continued checks during the interview process itself, including checks into the accuracy of paraphrasing or summarizing, to ensure complete and accurate understanding and interpretation on the part of the researcher was also used. This also served to verify the data for credibility.

As soon as each interview was transcribed, initial coding of each case began. Coding refers to the process of organizing and classifying detailed data to create larger topics and categories, and find patterns. Codes and quotes from initial interviews were then available for use during member checks at the beginning of follow-up interviews. All additional information obtained was also coded, including interviews with professionals and any pertinent data found within records provided by participants. Based on the codes, categories of codes that I considered themes emerged, which were also noted and analyzed as I added and coded additional data such as follow-up interviews. I used a software program called *ATLAS ti* to assist with organizing, data analysis, coding, and categorizing associated documents. As I read through each piece of data, including transcripts, as well as physician reports and IEPs received from parents, I flagged codes and quotes that I believed to be important for later comparison and analyzing. As I interviewed more participants, I was able to code and categorize the subsequent interviews by using the same codes with a high degree of frequency. There were some codes that emerged later in the study, but after the first three or four initial interviews, most new information fit into the existing codes and categories. In some instances, I also merged and divided categories to help ensure the categories were internally consistent.

In addition to within-case analysis, this study involved cross-case analysis, because I expected that the cases collectively would have similarities as well as differences. The cross-case analysis, wherein I examined the entirety of the data collected for all cases, across each question, category, and/or theme, represents much of the information presented in this final report. The process began with coding the

transcriptions from the first several participants' interviews and any documents they provided, noting information that seemed important to the informants. As more informant interview transcripts and documents were added to the project, several themes and ideas emerged as common across cases, and were categorized. After all transcripts had been coded and categories delineated, the categories were organized into related themes. This allowed me to compare and contrast the information from each case to find patterns, similarities and variances. I also present information obtained through interviews with professionals, which may verify or contrast with the information gathered from families.

Data security. Participants were identified by assigned case names, only, within the raw data. Data will be stored in a password-protected digital file identified by a case name, for each participant, which includes interview transcriptions, recordings, notes, and any additional digital media including email communications between participants and me, and scanned documents. I also have a paper-based file for each participant, where the confidentiality forms and other paperwork provided by the participant, as well as hand-written interview notes are stored with a label that matches the digital file. Recorded interviews and transcriptions have been stored on a secure server with a secure backup, as well. All data items have been modified to remove identifiers other than an assigned case name, and the case name identification key is located in a password-protected file. Once the project is completed, all paper items will be shredded and recycled.

Ethical Considerations and Impact of the Study

Additional issues in qualitative inquiry include ethical considerations, personal reflections, and thoughts about potential impact of the study itself. Sensitivity toward the participants, their time, and their stories is vital to the success of the research project. The researcher must always be aware of his or her effect on the study environment, but also the biases, perspectives, and experiences that they bring to the table that effect their interpretations of the data. Another issue for consideration is the potential impact of the study, or its purpose. This impact statement has helped to keep the research focused, while allowing for emergent themes.

Ethical considerations. For the proposed study, clear communication with participants regarding the purpose of the research, consent, and confidentiality was of utmost importance. I established a system to ensure confidentiality and data security for both electronic and paper-based materials. I have encouraged participants to keep an open dialogue with me during the project, and I made certain that they were each aware that they could withdraw from the study at any time. I also shared verbatim transcripts of each interview with the participant for member checking, but not with anyone else who participated in the study.

Summary

I approached this study, aware of my own opinions and biases regarding the diagnosis of CVI in children due to my experiences in working with children who have CVI. I also acknowledge that these biases need not interfere with research practices and processes. Throughout this study, my role was to observe and record

data which would allow me to describe family experiences. I analyzed transcribed interviews, performed member checks, and used triangulation in the form of educational and medical records provided by families to learn about families' experiences. In my position as the researcher, I know that it is important that I did not influence participants during their interviews to strengthen the credibility of the findings.

CHAPTER IV

FINDINGS

This chapter begins with a brief summary of each family who participated in this study and the issues that they were facing. Due to the identifiable nature of such stories, specific demographic information is reported in general terms and names are changed to provide increased confidentiality. Descriptions of service providers (Teachers of the Visually Impaired (TVIs) and eye care physicians), who served as professional participants and other sources of information, are also presented. Following the descriptions of participants, there is a delineation of themes and subthemes that emerged across families, from analysis of the data, to answer the following research questions:

1. What are parents' experiences in seeking a diagnosis for their child's suspected vision challenges?
2. What needs do parents recall related to information and supports while seeking a diagnosis for their child's suspected vision challenges?
3. What kind of information is offered or readily available to parents upon diagnosis of CVI?

Participants

Eighteen parents or sets of parents of children who had been diagnosed with or who demonstrated characteristics of CVI responded to the participant advertisement, which was shared either by the MD/DC DeafBlind Project, by families who had already interviewed for the study, and/or by the facilitator of an international conference call for parents of children with CVI. Of the 18 respondents, five did not

meet the eligibility requirement regarding maximum age of the child, and two did not respond to follow-up emails requesting additional information about their children and to discuss consent. Therefore, the results presented in this chapter are based on communication (in-person, over the phone, and/or via email) with the 11 remaining families. The results also include information obtained through interviews of four professionals – two TVIs and two physicians who are qualified to recognize, provide interventions, and/or diagnose CVI.

Parent participants.

Marla. When I arrived at Marla's home, she was putting her children in bed for the night, so I was briefly able to meet Gwen, who was four years old. Marla's husband was at work, and we were able to sit and talk without much interruption once the children were upstairs. Marla put a great deal of thought into her word choices as we spoke, often hesitating before speaking on a particular subject.

When Gwen was born she seemed to be a typical baby, but she did not feed very well at first, and while still at the hospital, Marla and her husband thought her lips looked blue. When Gwen was placed in the nursery, one of the practitioners noticed her coloring as well, and they placed her on equipment to assist her breathing. Testing revealed that she had aspirated amniotic fluid (without meconium), and she was able to go home within two weeks. Marla said that months later, they noticed that she was not doing the things that their older son had done, in terms of "communication and gross motor". Specifically, she was not making sounds like

their older son had made, she was still crawling at age 2, and at medical checkups she was found to not be reaching milestones.

Gwen had many ear infections as an infant, and had her tonsils and adenoids removed, and PE tubes placed. Her speech was delayed, and they thought it could have been due to hearing issues. It turned out that Gwen experienced delays in many areas of development. Her physical therapists questioned Gwen's vision, but eye care physicians said that her "eyes were fine". The physical therapist pressed the issue and described what she was noticing in sessions with Gwen. Marla explained that, at first, reading the information that the PT sent to the eye doctor, she and her husband felt that the things she had observed were old news. They were unsure about the purpose of rehashing that information, but they went forward with the process to see what would happen. The eye doctor agreed that it sounded like CVI, diagnosed it, and referred Marla to a local TVI who is also a CVI specialist. Gwen was approximately four years old by this time.

During a follow-up interview Marla told me about her thoughts about the school system they were in at the time, and about how they were going to be moving to a different school district that, they had hoped, would better serve Gwen's needs. A number of things happened that affected their decision to move. Within a month of Gwen's CVI diagnosis, the IEP team met to add services to the IEP. The vision teacher who came to the meeting had a visual impairment of her own, and Marla thought that although it was "great that she was helping kids with vision issues", it was going to be difficult for her to see Gwen's visual behaviors. Marla remembered asking the TVI if she had experience working with children who had CVI, and she

said that Gwen would be her first. The TVI then read aloud some information about CVI, which Marla said sounded the same as the information she had received from Gwen's pediatrician. Marla felt that she herself did not know enough to be certain, and that the proposed IEP goals did not seem "useful", in that they did not seem to be challenging and beneficial to Gwen. Feeling that it was not going to be a good fit, she consulted with the CVI specialist, who agreed and she felt validated. At the next IEP meeting they agreed to find a sighted TVI. By then, members of the IEP team seemed to think that Marla was "looking for it [the IEP team's plan] to fail". She recalled an email from the classroom teacher to the TVI where the teacher had said "I don't have any issues. This is all [Marla]". Marla realized it was time to leave the school system when she recognized that no one else on the team seemed concerned.

The new school district placed Gwen in a preschool classroom specifically designed for children with visual impairments, and Marla was very hopeful that Gwen's needs would be well-met there. Marla also provided Gwen's IEP, a report from her eye care physician, and the report from Gwen's school-based functional vision assessment (including the CVI Range). When I last checked in with Marla, Gwen's new placement seemed to be working, and Marla was feeling better about the school-related placement and services.

Lisa. When Lisa's son Bobby was born, he appeared to be a typical baby in most ways. She immediately noticed that he would not make eye contact, but his pediatrician told her that he was curious and trying to look around and take in everything else instead. Later, however, he developed seizures and started to lose the skills that he had gained, such as crawling and walking. The seizure medication had a

known side effect of peripheral vision changes, so Lisa was very observant of his vision after he started taking the medication. One day she watched him crawling across the floor and she said it looked like he was using his hands to feel his way around, and he was not looking at anything. She took him to the doctor immediately and they tested him and said he was fine. Lisa was still concerned, and one of the nurses suggested she contact her early intervention team and inquire about vision services. She was assigned a TVI, who did a vision assessment along with a TVI from the state school for the blind, and they determined that Bobby was showing characteristics of CVI. At that point, Lisa made an appointment with a pediatric neuro-ophthalmologist, who provided the medical diagnosis of CVI when Bobby was approximately 20 months old.

Later testing revealed a genetic mutation that is rare and does not yet have a name other than the name of the specific gene that is affected. Lisa joined a parents' group for those with children who are affected by the same gene, many of whom have CVI, also. Lisa said that she wants other parents to know that if they suspect a vision issue, to request a vision evaluation and get the diagnosis. She also suggested that parents make sure there is a TVI working closely with the early intervention team for any child who has suspected or confirmed CVI.

Lisa and her family had recently moved, and she invited me to her home for the interview. She was having concerns about the new district and classroom where her son, Bobby, would be attending school because the new district did not seem to have CVI-knowledgeable staff. In her experience, school issues had always been difficult and she felt that "they" never met Bobby's needs. In the new district, the

IEP Team had ordered a complete set of assessments, but Lisa questioned the staff's ability to assess Bobby's CVI. The TVI said she had never had a student with CVI before Bobby. Lisa felt that if the CVI was not addressed through accommodations throughout the school day, Bobby would have difficulty in all areas – she said "He's not gonna learn if his visual needs are not met". She was very concerned that his services would then be reduced, but was also hopeful after meeting with the new IEP Team because the new TVI sounded positive.

When I arrived at the family's house, I met Lisa and was also introduced to Lisa's mother, Bobby's private duty nurse, the family dog, and Bobby. Lisa showed me Bobby's bedroom and play spaces, which she had started to modify to reduce visual complexity. Bobby, who was six years old, was playing, and I got on the floor about 3 feet from him and watched quietly. It was not long before Bobby glanced in my direction, then looked a little longer, and after a few minutes he looked right at my face. Lisa mentioned afterward, that she appreciated how I approached Bobby and said that people try to approach him directly, and that sometimes people expect him to give eye contact immediately.

Tracey. Tracey's daughter Phoebe, who was five years old, had been diagnosed with CVI when she was about three years old. Tracey's pregnancy with Phoebe was complex, and after a basic genetic test had abnormal results, she saw many specialists but none were able to say what was "wrong". She spent the last two months of her pregnancy in the hospital, and Phoebe was born at 37 weeks. Phoebe's genetic anomaly was identified as soon as she was born, because her appearance is very typical for Cornelia de Lange Syndrome (CdLS).

The foundation working on behalf of children with CdLS has been very helpful, connecting the family with other families that are "going through the same thing". The family was informed in early infancy that Phoebe did not pass her hearing screenings, but it was not presented as a high priority at the time due to other caregiving concerns (feeding Phoebe using a nasogastric tube, for example). They later realized that she genuinely could not hear, and Tracey said that Phoebe started using hearing aids when she was about seven months old. Then, they started looking at her vision and she had surgery to correct ptosis (an ocular condition in which the eyelids droop). Phoebe still did not look at things unless they were extremely close to her face, and after several consults with eye care physicians, Phoebe started wearing glasses. When she was two years old, she went to half-day classes during the week at the school for the deaf. It was there that Phoebe's teacher asked if she could pursue a functional vision assessment, and they concluded that Phoebe demonstrated many of the characteristics of CVI. The TVI who did the assessment understood CVI and provided the family with information about how to adapt things to help Phoebe see them better, where to put things to get her to notice them, and it helped them understand why Phoebe would not look at faces. Tracey said that when they later asked the eye doctor for the diagnosis, he had said that Phoebe was too young and that they should wait. Tracey went elsewhere for a second opinion, and that doctor provided the actual diagnosis, which was helpful when they sought out all of the accommodations for Phoebe's IEP.

Follow-up information about Phoebe and Tracey was obtained via phone, to verify details and my understanding of their story.

Veronica. Veronica and her husband had decided to become foster parents, and eventually adopted Brady when he was six weeks old. Medically, he was not expected to live long enough to see his first birthday, so when Veronica and her husband brought him home, hospice services were already in place. At the time of the interview, Brady was six years old. When Brady was about a year old, his biological mother passed away, and four of his biological siblings joined him in Veronica's care as foster children. Brady had fifteen siblings, total, and some were adults by that time but they were all still connected. Two of his siblings were present for part or all of the interview, which was at their house.

Brady's biological mother had attempted a home birth, which she had done many times before. Unfortunately, labor with Brady resulted in a placental abruption and a prolapsed umbilical cord, so Brady was without oxygen for a length of time before she was taken to a hospital by ambulance for an emergency Cesarean section and Brady received CPR for ten minutes before he was revived. At the time of the interview, Brady had a tracheostomy and was fed via gastrostomy tube, and he was very susceptible to illnesses, particularly respiratory illnesses.

Because he had such a complex medical history, the medical professionals believed Brady's brain was not taking in environmental information such as sound and vision. When Brady was about eighteen months old, it caught her off-guard when the eye doctor said he was "blind". When he was approximately a year old he had gotten glasses, and Veronica said that he had always been able to visually localize on ceiling fans, so she never considered him to be "blind". He also would startle when there were loud sounds, so she believed he could hear sound, as well.

Veronica was unable to show me, but she said that Brady had medical paperwork that she remembered seeing, which indicated that he had CVI.

Veronica was an occupational therapist, and she said that because of her profession, she knew that what doctors say sometimes does not seem to line up with reality. When she was told that Brady was "blind", but she knew that he would visually localize on things (including her face), she did not give it much thought until she attended a conference about deafblindness, where they mentioned CVI. In further discussion about it, Veronica noted that Brady's medical needs were so significant that his vision and hearing needs were set aside, especially when he was an infant. Veronica said that she was cautious about exposing Brady to public spaces for medical and specialist appointments that were not life-preserving, required, or life-altering to limit his exposure to places with germs that could induce illness.

Veronica remarked that being a professional occupational therapist affects her perspective when service providers suggested activities and came to see Brady in their home. For example, she felt that some professionals would visit to work with Brady and they would "ignore the vision". She said that oftentimes, the focus was on tactile stimulation, and when his teacher would ask him what color he wanted for something, she wished the teacher would show him the color options, and she said, "the OT in me wants to facilitate that vision component". She also said when they found that Brady would look at projected and moving light, "The OT in me took off with that and we have tons of different projectors and lights that move, and he enjoys them". We concluded our first interview, and we connected by phone thereafter to

ensure my understanding of their story was correct, and to clarify and add comments as needed.

Lauren and George. Lauren and George, parents of Andrew, decided to participate in this study as a couple. During Lauren's pregnancy with Andrew, they had known at twenty weeks that something was atypical, because ultrasound examination showed abnormal facial formation. A geneticist shared that it could be a wide range of conditions and that he was unable to say if Andrew would survive until delivery, which Lauren described as a "nightmare". Andrew did survive, and he spent his first four months in the neonatal intensive care unit (NICU). Andrew's diagnosed syndrome, Cornelia de Lange Syndrome (CdLS) accounts for his global developmental delays. Along with the diagnosis of CVI, he had small optic nerves that affected his use of vision. In the NICU, Lauren remembered doctors were very concerned about retinopathy of prematurity (ROP). Andrew was tested for ROP and results indicated he did not have it. After leaving the NICU, his ophthalmologist diagnosed nystagmus (an ocular condition where the eyes quiver) and checked to see if Andrew had a tumor, which he did not have. When Andrew's parents questioned the possibility of CVI, which is common in children with CdLS, the ophthalmologist said the he did not have CVI. Lauren and George were specific, however, that the doctor was a general pediatric ophthalmologist and overall they had been pleased with the doctor's observation about nystagmus and that he knew to check Andrew for a tumor. They did feel that he was dismissive with regard to their question about CVI.

Andrew was receiving services from a TVI who was knowledgeable about CVI and believed Andrew did have it, because his difficulties with functional vision were not fully explained by his diagnosed ocular conditions (nystagmus and acuity differences). His parents decided to have him checked by a neuro-ophthalmologist who diagnosed CVI immediately. The physician suggested that the diagnosis may or may not be helpful, "because CVI covers so many things", that interventions may or may not help – but he recommended that they try. Lauren and George suggested that seeing a neuro-ophthalmologist was truly the way to get CVI medically identified, and doing that was the most helpful to them. The assistance they received from the TVI who originally suggested it could be CVI, and the materials she recommended to them for it, were very helpful. She also visited Andrew while other service providers were working with him, so that she could help advise them in ways to encourage him to use his vision during those tasks.

After the interview, we heard Andrew kicking at the camera on his bed, which sent a video signal to a monitor so that his parents could watch him and know whether he was having any difficulties. I heard his voice and asked them if he was vocalizing to himself, and they said that he generally loved bedtime and that he would always play around before going to sleep. Follow-up information for clarification and member checking was accomplished over the phone.

Raina and Charles. Harrison was five and a half years old at the time of the interview with Raina and Charles. Harrison was born breach, and the doctors noticed right away that he had low muscle tone, but it took weeks or months to realize that there were additional concerns. It started with failure to thrive, because there

were many difficulties with feeding, and multiple allergies. Since Harrison was their first child, they did not realize right away that newborns' eyes did not "shake" – Harrison had nystagmus, which is a condition that involves rapid, involuntary eye movements. They started receiving services from Infants and Toddlers when Harrison was 11 months old, and he was unable to lift his head while in prone during a visit to the doctor. Charles said that issues with Harrison's vision were not truly on their "radar", with the other things that were happening.

The ophthalmologist, who saw Harrison when he was 11 months old, ordered an MRI, which showed small optic nerves but no other abnormalities. They saw a geneticist, and found a mutation on the gene that controls the body's making of a specific protein that helps neurons "migrate into place" during prenatal development, which affected Harrison's neurological development. Vision had always been a mystery of sorts; Harrison had difficulty noticing things in his lower visual field, and they had always had questions about how well he could see.

Harrison was placed in an intensive day program when he was three, so that he could receive speech, occupational, and physical therapies as well as vision services. Vision was the only area at the time that did not seem to be improving, and they thought that he had attentional issues. Another TVI observed him in his environment, at home and at his day program, and the team determined that he would likely benefit from a preschool classroom that focused on visual impairments. As he grew, his visual impairment came to be the most apparent, prominent difficulty. At the IEP meeting that took place before my interview with these parents, an

Orientation and Mobility (O&M) specialist who had observed Harrison introduced the family to CVI.

Once the family heard about CVI, they started to educate themselves and suddenly Harrison's visual behaviors started to make sense to them. They realized how CVI impacted Harrison's ability to process his environment and manage his frustrations, for example he got overwhelmed easily in complex environments, during fine motor tasks, and anytime tasks demanded more effort, visually. During my interview with his parents, they discussed some of the concerns and what they would like to see in his next IEP, and they still had many unanswered questions. When I asked about the emotional part of their journey, both parents said that it had been very difficult. Raina mentioned specifically that continuing to hear that he has a low-incidence disability, from professionals in the school system, was frustrating because it felt as though the school system was giving excuses to explain their lack of knowledge about CVI.

I later confirmed that Harrison was 6 months old when they suspected he had a vision issue, and that despite the contact and advice they had received from educational vision specialists, he did not yet have a medical diagnosis of CVI. Although the TVIs in Harrison's school district had been helpful, they were still working on the medical diagnosis. Raina invited me to Harrison's neuro-ophthalmology appointment later that same week, where he was diagnosed with "central visual impairment". The doctor allowed me to audio record the appointment, which was very long and I was unable to stay until the end. Raina also provided me

with a copy of Harrison's IEP, which confirmed many of the things we had discussed during the interview.

Nadia and Boyd. Nadia and Boyd recalled that there was concern about their son, Paul's development in utero starting at 20 weeks' gestation. At the 20 week ultrasound, they found themselves meeting with a geneticist due to several concerns, including Paul's small chin, more amniotic fluid than is typical, and "some problems with the brain" – none of which were definitive nor confirming if anything was going to "be wrong when he came out". At almost 22 weeks, the doctor they met with reminded Boyd and Nadia that it was not too late to terminate the pregnancy, but they decided to go to term.

Paul was born, and the medical team whisked him away to "work on him". Boyd could not clearly remember, during the interview, if he had been able to cut the umbilical cord, but he remembered that there was a sense of urgency for Paul to go to the NICU. Nadia and Boyd wanted to take a picture first, and Boyd said that looking at the photo later, it was clear that Paul was "totally blue." At one point during those moments immediately following birth, Paul flat lined and the doctors were able to revive him. In the months that followed, Paul was unable to tolerate lying in supine because his tongue would fall back and obstruct his airway. He had very low tone and was unable to swallow or keep his tongue forward in his mouth. As a result, he received a gastrostomy tube and tracheostomy tube.

All of these medical issues were, of course, a priority. Two days before Paul was discharged from the hospital, he failed his newborn hearing screening and they discovered he was deaf. This was upsetting for Nadia, and she remembered yelling,

asking what else was wrong with him and if he could see. Because Paul did react to some visual stimulation, Nadia says she thought he could see. Neither had any idea that Paul's visual attraction to lights and things that moved was anything more than the fact that he was a baby. Paul also had Moebius syndrome that included facial paralysis, which made evaluations of his skills more difficult.

Just before Paul turned two, Nadia was talking with other mothers of children with tracheostomies, and one person mentioned CVI, something that she had never heard until then. She pursued it with the host of practitioners that provided early intervention services in the family's home, and the TVI agreed that he fit the criteria. It made Nadia angry that no one had mentioned it or thought of it before that, and Boyd agreed as they both said "we were doing the wrong things". They read Dr. Roman's book about CVI and were heartbroken when they read that it is critical to have CVI identified and interventions in place before children turn two. They made an appointment to see Dr. Roman, and recalled that it was a positive experience and because they learned about what they could do to help Paul use his vision. They reported that as soon as they started presenting information in ways that were adapted for Paul's vision, he started making progress.

In a follow-up interview, I asked if he had received a diagnosis of CVI from a medical doctor, and Nadia said they did not remember having a conversation with a doctor, but that it appeared in his medical paperwork at some point. Nadia and Boyd also confirmed that there was added difficulty in understanding that Paul was not understanding what his eyes took in because of the facial paralysis (he could not blink

his eyes or track things horizontally) and hypotonia. Additional information and confirmation was obtained over the phone.

Bethany. Bethany and her son, Jonathan, lived on the west coast, so our interviews were conducted over the phone. Her son, Jonathan, had a stroke right after he was born, and was diagnosed with "cortical blindness" [which is no longer considered an appropriate term to diagnose children with visual impairment not caused by an ocular condition, due to expectation for vision improvement in CVI (Lehman, 2013)]. Bethany recalled sitting on the floor in the hospital after hearing that, crying, because the diagnosis lead her to believe he was going to be completely blind.

Jonathan was ultimately diagnosed with multiple disabilities, including delayed fine and gross motor skills, delayed speech, feeding difficulties, and seizures, but Bethany felt that his vision had the most impact on his abilities. At the time of the interviews, Jonathan was six years old, and Bethany was working to become a certified TVI. She felt very strongly that, as a parent, her views and opinions at IEP meetings were not considered in the same way that the opinions of professionals were. She thought if she were to become a teacher of the visually impaired, her opinions would be more carefully considered. Bethany was also the author of a blog about her child and their experience with CVI, so this may have increased her ability to put her thoughts on the subject into words during our conversations.

Although Jonathan was diagnosed with CVI almost immediately following birth, Bethany said they had found the transition from early childhood services to school-age services to be most difficult, when services changed from home-based to

school-based. Jonathan found being in school itself to be difficult, and he once said that it was too crowded, there were too many people. Bethany's interpretation was that there was a high level of complexity in his school environment. Bethany said that when she would volunteer in his classroom, it was difficult because when she was finished he would not want her to leave. She said "when I leave, his [visual] access leaves". Jonathan started displaying negative behaviors such as kicking, picking up chairs, and hitting. Bethany said that the additional complexity of a kindergarten classroom caused Jonathan to have anxiety ("fight or flight"). She was working with the school to help the professionals working with Jonathan to make more adaptations for CVI and to recognize the earlier signs of frustration in him, so they could intervene before he reached that "fight or flight" state. She found that school personnel were focused on interventions for the behaviors rather than adjusting the environment to try and prevent them.

Emotionally, Bethany struggled most with having to "constantly explain" her child, and how to explain it easily. She also said that it was particularly difficult that she did not feel "heard" when people did not understand him or his vision despite her attempts to explain things about Jonathan's behavior. Jonathan wore glasses, so people too often assumed that his visual impairment was taken care of with glasses, and Bethany was frustrated that people had "no idea" what she was talking about when she tried to explain. For example, she was very glad when she found out about the weekly teleconference for parents of children with CVI, because support groups for parents of children with visual impairments did not really fit for her situation. She also said that she felt isolated and drained while standing outside the school each day

with other parents, waiting for their children to come out, because her experience, and Jonathan's, was not at all like the other families' experiences. Sometimes even other parents of children with multiple disabilities (including CVI) were difficult to relate to, especially when she had made Jonathan's vision a priority and the other parents had different priorities.

Bethany also had similar feelings when she spoke about meetings with educational professionals – that she was not heard, that she always had to explain Jonathan, and that even the professionals did not understand. She attended a CVI training at Perkins School for the Blind and found that being in a room with so many people who understood CVI was an emotional experience; that it was the only time she had ever been in a room with so many people who "got it," and it was emotionally overwhelming. When she had to teach the professionals how to work with Jonathan's vision it did not increase her confidence in their ability to work with him.

Claire. Claire's son Dylan was born at 23 weeks' gestation, and his twin passed away shortly after they were born. Dylan was in the NICU for nine months after birth, and his family faced many medical complications with him, including cardiac and respiratory concerns. Dylan had also failed his newborn hearing screening, so the family was working on follow-up from that once they were able to bring him home from the hospital. Shortly thereafter, the family noticed that he was not looking at them, that he would stare at light sources, and that feeding him was difficult due to his intense light gazing. The ophthalmologist and neuro-

ophthalmologist had confirmed that the eyes and all parts of the visual system were intact, and they were unable to test how clearly he could see at that age.

Dylan started to attend an infant program at the school for the deaf in the state, where he would go weekly, and the teacher would visit them at home as well. The teacher there noticed that Dylan's visual behaviors were very similar to other students she had who had diagnoses of CVI. The family agreed that someone (a TVI) could come assess him, and that person found that he fit the criteria for CVI, and wrote an assessment report and a recommendation letter for Dylan's eye doctor. This helped the family get a diagnosis of CVI into his medical records. Claire said that the TVI also provided them with a great deal of information at that time, and Dylan was about two and a half years old.

Claire found the information provided by the TVI to be very helpful, and felt grateful to the teacher of the deaf who made the connection between his visual behaviors and those of other children. Although she was sad about it, Claire found it comforting to read the book that the TVI had recommended, which was about CVI and one family's journey with it. The book helped the family feel hopeful that Dylan's vision could improve, and they felt empowered to work on it. They got lights and low complexity, high contrast items similar to the ones the TVI had brought for assessment, to work with Dylan and help his vision improve. The book recommended by the TVI continued to be a valuable resource, and Claire felt it was like a guide with specific steps she could do with Dylan. At the time of our first interview, Dylan's vision had progressed to the point where he was looking at and recognizing family members and Claire felt that he used his vision a lot. He began to

look at her as if to communicate, and that she felt good about that because it was a way for her to feel more connected to him.

At the end of the initial interview, Claire mentioned that she wished there was some peer-to-peer network for parents of children with CVI noting that it is very lonely and that many parents feel they are alone. She said that most parents do not get it, unless they have a similar situation, and that she felt it would be valuable to have parents connected either in person or by phone. Since I knew there was an existing weekly teleconference, I mentioned that there is such a group of parents, but Claire had never received information about it. After the interview, I contacted a parent whom I knew participated in the weekly call, and I was able to provide Claire with information so that she could make connections with other parents that truly did understand her experiences. I followed up with Claire over the phone to verify my understanding of her story and to thank her for participating. I also wanted to make sure she had gotten the teleconference information and was able to get on the call so that she had more support.

Jennifer. Jennifer and her family lived several states away, therefore the interview was conducted by phone. The eldest of Jennifer's two daughters, Gail, had CVI and was six years old at the time of the first interview. Jennifer said that Gail was typically developing in most ways, other than the visual impairment, by the time of the interview. Gail attended her neighborhood school, and had occupational therapy services at home several times per week. Gail's story was unique in that her diagnosis came early, but interventions did not occur until she was four years old.

Gail was born two weeks early due to Jennifer's preeclampsia, and Gail was low birth weight. Initially, Gail was not nursing well and Jennifer was pumping and feeding her through a small tube attached to Jennifer's finger, which Gail would suck on. This was not giving her enough nutrition, though. Jennifer's mother passed away four months after Gail was born, which was very difficult for the family. Jennifer and her family members all started to notice that there was something atypical with Gail's vision. Jennifer's father-in-law was an ophthalmologist, and he researched and set up an ophthalmology appointment for Gail.

As a result of the appointment when Gail was eight months old, Gail was diagnosed with optic nerve hypoplasia (a condition in which the optic nerves are underdeveloped) and CVI, as well as several other things that Jennifer was not specific about. Because Gail had previously had an MRI, which showed brain damage to the occipital lobe (believed to have been caused by an unknown vascular event in utero), the doctor felt comfortable diagnosing her with CVI. Jennifer felt that if there was no MRI, or if the brain damage did not show on the MRI, they may not have received the CVI diagnosis. However, the doctor focused on the optic nerve hypoplasia, therefore the family did as well.

Jennifer recalled that the eye doctor did not make suggestions about ways to help Gail's vision improve (in terms of CVI), and she wished they had received more information about the CVI. She indicated that when she looked back at the experience, she felt that the eye doctor was great in general, but that he did not seem to know enough about CVI to guide them. Early Intervention professionals began coming to the house, including a TVI, when Gail was ten months old. By the time

Gail was about a year old, she had started to nurse, but the family was concerned that she was not reaching her developmental milestones so they made the choice to have a gastrostomy tube placed. This made a huge difference and she caught up with the motor milestones within three months of the surgery.

Jennifer said that Gail was typically developing in most ways, other than the visual impairment. Gail attended her neighborhood school, and had occupational therapy services at home several times per week. Gail's story was unique in that her diagnosis came early, but interventions did not occur until she was four years old. Jennifer also felt that no one looked at all of Gail's vision diagnoses, however, and CVI interventions were not implemented. When Gail exited Early Intervention, she attended the state school for the visually impaired, for preschool. There were no obvious interventions applied for CVI there, either, and the family also had not tuned into it because everyone thought the other visual diagnoses explained the functional visual impairment. When Gail was four, her ophthalmologist-grandfather wanted to have her retinas examined by a specialist a couple of states away from home. At that appointment, the team of specialists watched Gail in the clinic and also took pictures of her optic nerve – and they suggested she may have optic nerve atrophy, which would be less severe than hypoplasia. As they watched her play in the clinic, one of them mentioned that she was navigating the environment well, and suggested that perhaps the CVI was causing most of the issues she had with her vision.

The family was surprised by this, looked back at her original diagnoses, recalled CVI was there, and that it had been mostly overlooked due to the presence of other conditions. They started to do research, and meet other parents and

professionals who understood CVI, and Jennifer said that it was overwhelming. They set up an appointment with Dr. Roman, which occurred right before Gail turned five, and when school resumed that fall, they came prepared with "informed questions" for the school system to make sure that they were planning to address the CVI.

The school system did not have answers, however. They brought in a consultant to assess Gail, but the results were questionable and the family was able to have it documented such that the IEP Team did not write goals and objectives based on an invalid assessment. The Team did want to implement a "dual media approach" – including print and Braille. The family worked very hard, networking with professionals who understood CVI, to get feedback to the school district that Braille instruction was not appropriate. They succeeded in getting Braille instruction removed from the IEP, despite a great deal of pushback from the school system.

At the time of the interview, Gail was in kindergarten and doing well. She was reading sight words without accommodations and modifications, did not want to use her long cane, and had grown to dislike the color red because when her work assignments were modified, the color was added to help her see the salient features of pictures and words. Gail was six years old, and she did not want to do things differently than her peers. This was difficult for Jennifer, as she said she was not prepared for the impacts of peer pressure so early in Gail's life.

I was able to go back and confirm my understanding of the story during the same phone call with Jennifer. When I asked Jennifer about any advice she had for TVIs or eye doctors, she said that she felt the university system was a huge issue, because the focus is on Braille and ocular impairments, and they typically do not

cover CVI effectively. Jennifer had some interesting ideas for parents, including that families build personal advisory boards – a group of professionals that they can contact when they have questions or need help with something. I later touched base with Jennifer to confirm some minor details and to check in with her by phone.

Sharon. Sharon and her family lived a few states away, so her interview was conducted by telephone. Her daughter, Mary, was the youngest of three children, at five and a half years old, and was almost ten years younger than her middle child. Mary was born with Downs Syndrome, she had epilepsy, and at the time of the interview she did not communicate using speech. Mary depended on a gastrostomy tube for nutrition, and she was learning to stand independently. Sharon said that she had been seizure-free for several months, and that when Mary was able to stay seizure-free for some time, she became more alert and better able to follow simple directions.

Mary was diagnosed with CVI between the ages of two and three. At the time, Sharon and her husband were not sure if she was able to see and hear, just that she would look off to the side and at shiny things (which give the sense that they are moving because the light reflects), and that since Mary did not speak, it was a "complicated challenge". The family started to suspect something was atypical with her vision before Mary was 18 months old, but they had always had concerns because she was not making eye contact. Sharon said that all of the behavioral characteristics of CVI applied to Mary; she would not look at things that were beyond four feet away from her nor would she look and reach for anything. She had been followed by an ophthalmologist from six months of age, and Sharon felt like she was repeatedly

being told to wait (for Mary to grow and for her vision to change), until their ophthalmologist referred them to a specialist, which is the professional who diagnosed it. Sharon said until then, she had never heard of CVI. By that time, Mary had a host of other diagnoses as well, after going through many unpleasant medical tests. Sharon said that it took her awhile to embrace the diagnosis and teach herself how to help Mary learn, but that an occupational therapist was instrumental in helping Sharon through the process.

Sharon recalled that preschool was really difficult for Mary. Even though there was a "vision therapist" there who understood CVI, it took time for the classroom staff and related service providers to grasp the adjustments they needed to make for Mary to be successful, and they did not have some of the tools that would help Mary be able to see (such as a light box). Even after the diagnosis, Sharon had to bring experts from many different disciplines together to develop teaching strategies that would work, but she was also concerned that every time Mary would have to change classrooms in the future– annually, for example – they will go through the process over again, to customize each setting for Mary's success. At the time of the interview, Sharon was considering the option for a home-based program so that the setting would be familiar and conducive to learning. Mary would then be missing the social aspect of school, but the family was willing to find another way to give her that.

Sharon realized that she was "the one" – saying she felt like she had taken crash courses in physical, occupational, and speech therapies, and that people were going to expect her to be the expert as far as Mary was concerned. She felt

responsible not only for understanding all of these things, but also for being able to teach it to others who worked with Mary. Sharon came to understand that every area of life was impacted by Mary's CVI.

Professional Participants

Professionals who participated in this study included teachers of the visually impaired (TVIs) and physicians who primarily work with children with vision impairments. These professionals were referred by other informants in this study, and several of them were referred by multiple participants.

Teachers of the visually impaired. Few parents were able to recommend TVIs that they considered especially skilled or "tuned into" CVI. The criteria for eligibility to participate in this study was that the TVIs were willing and able to speak to the diagnostic process and/or their experiences when working with children with CVI. Two TVIs were referred by parent informants, and one of them was referred by multiple families. Another had suggested she was interested, but did not respond to several attempts to gain consent via email. Both TVIs who participated had met me previously at MD/DC DeafBlind Project events, were school-based, and served multiple children with CVI on their caseloads.

Eye care physicians. Since many of the parent participants are from a limited geographic area, they often recommended the same vision-related physicians as potential participants in this study. Both physicians who participated had experience in diagnosing CVI, and neither was familiar with me directly. One of the physicians had an urban, hospital-based practice, and the other was part of a suburban practice with a number of other eye doctors. Each of these two participants had been

diagnosing CVI for more than 15 years, and were affiliated with universities as well. One of them was a pediatric neuro-ophthalmologist and the other was a pediatric ophthalmologist.

Themes and Subthemes

During the process of analyzing the participants' transcribed interviews, four main themes emerged. The overarching research questions for this study were "What are parents' experiences with the process of getting CVI recognized and diagnosed in their children?", "What needs do parents recall related to information and supports while seeking a diagnosis for their child's suspected vision challenges?", and "What kind of information is offered or readily available to parents upon diagnosis of CVI?". I used software to code each interview, highlighting the points that came out as important to the parents. After several interviews had been coded, I flagged ideas that appeared multiple times and organized these into categories across interviews. The categories were then organized under four main themes. The first theme, "Something was Wrong" reflected a common experience among parents in which they noticed something was different than they expected with regard to their child's vision. It includes the subthemes, "Lack of medical knowledge and understanding", "Delayed diagnosis", and "Seeking clarity". The second major theme, "From Diagnosis to Services", is about the families' journeys from diagnosis to intervention. It includes the subthemes "Information received from professionals", "Lack of educational knowledge and understanding", and "Changing parenting style and learning new skills", which speaks to the parents' adjustments to the new information after their children were diagnosed. The third theme, "From Frustration to Hope" includes

parent reflections on the emotional aspects of the diagnostic experience. It includes subthemes about stress, "It's a struggle", and hope, "These are things we can definitely do". The fourth and final theme, "Advice for Others", is reflective of advice that parents wanted to pass on, including subthemes of "Advice for professionals" working in the field of visual impairment and "Advice for parents". This information is summarized in Table 2 and is described in detail in the following sections.

Table 2: Themes and Subthemes

Themes	**Subthemes**
Something was Wrong	1. Lack of medical knowledge and understanding 2. Delayed diagnosis 3. Seeking clarity
From Diagnosis to Services	1. Information received from professionals 2. Lack of educational knowledge and understanding 3. Changing parenting style and learning new skills
From Frustration to Hope	1. "It's a struggle" 2. "These are things we can definitely do"
Advice for Others	1. Advice for professionals 2. Advice for parents

Something Was Wrong

The first overarching theme was that parents noticed that their children's vision was not developing as they believed it should and that "something was wrong". This theme and its subthemes directly address the first research question "What are parents' experiences in seeking a diagnosis for their child's suspected vision challenges?" in that noticing "something was wrong" lead parents to seek a diagnosis.

Infants use their vision immediately following birth, yet some parents reported that their child did not look at them the way a child without visual impairments would look, from the time their children were young infants. Without this, some parents said they struggled to feel connected with their child. Nadia and Boyd recalled such an experience, when they realized that Paul was not looking, and Nadia said, "There's all sort of emotional things going along with your child not [looking and] smiling at you." For Nadia, not knowing about the existence of CVI early in Paul's life made it difficult to accept and understand what was happening. Nadia recalled that Boyd thought Paul was looking at him, but in hindsight both parents realized that was not the case. Both parents were told that it was not possible to tell what Paul could see, and that they would have to wait for it to become clear, when they mentioned that Paul was not looking at them. Only after they had an appointment with Dr. Roman, did they feel like they understood CVI, stating "she gave us more useful information in that hour and a half than we had ever received. Truly, until that point, everyone was like we'll see what happens when it happens."

In contrast, Lauren and George were less concerned about Andrew's vision when he was an infant, though they knew something was different about it, and Lauren said, "It was pretty clear that he wasn't tracking things the way that he should. For whatever reason, he wasn't visually responding the way that he should. He wasn't making eye contact." Now that he is older, Lauren feels that Andrew's visual impairment, "does make it harder for him. It does exacerbate the other developmental delays". Andrew's parents indicated that since his health stabilized, his visual impairment has had the most impact on his ability to learn. Lauren

explained that they knew he was going to have profound developmental delays, and the vision was a part of that. She noted that if they had received confirmation that Andrew had CVI, they would then have had an opportunity to react sooner.

Like Lauren and George, Raina and Charles recalled that when Harrison was a baby, multiple factors affected his development, but the situation changed as he grew. As more and more of his health and developmental issues resolved, his visual impairment emerged as his primary disability. Rachel said:

> We've always known that vision is a big mystery with him […] we would go back to the ophthalmologist and say that "we're noticing these behaviors, he's not able to attend a task, he can't find his picture on the board," and they kept being like, "Oh maybe it's just attention issues". Nobody ever mentions CVI.

As stated earlier, Marla's daughter, Gwen's, vision difficulty was also considered too soon to diagnose, and Marla was instructed to "just wait and see". These sentiments were also recalled by Lisa, who felt that her son Bobby's CVI – and the fact that he would never look at her face - was explained as infant curiosity (he looked at the ceiling lights, mainly). Phoebe's vision care professional gave her mother the impression that Phoebe was too young for a CVI diagnosis.

Lack of medical knowledge and understanding. Although parents often had the sense that something was wrong, several recalled that some of the physicians that they consulted about their children's vision were unable to answer their questions. Some parents recalled conversations with vision care physicians wherein they were told that it was too soon to act, too soon to diagnose, or too soon to tell if something was wrong. Marla, Lisa, Nadia and Boyd, and Claire were all told all by various

ophthalmologists, that their babies' eyes were fine, and therefore they should be able to see.

Marla described taking Gwen to multiple eye doctors, all of whom said that her eyes were fine. She had started to research visual impairment on her own, and she took Gwen to an eye doctor at the local children's hospital. Despite specifically requesting a conversation about CVI, the doctor focused only on her visual acuity, and he had "nothing to say" about CVI. The doctor implied that it was too early to tell, and suggested not doing anything about it. The physical therapist who was seeing Gwen at school explained her concerns about vision to Gwen's parents and obtained permission to contact the physician directly to inform him about her observations, and only then did the doctor agree that the issue could be CVI.

Lisa encountered a similar obstacle with some physicians, including one who she felt was particularly dismissive. She took Bobby to a specialist who she remembers saying, "He's not blind…He's fine, he's fine. There's nothing wrong." This was after she had taken him to a pediatric ophthalmologist, to obtain a specific medicine that has known side effects related to vision. They waited in the waiting area for hours, where Bobby had been having seizures the entire time. When they were taken back to see the doctor, Lisa recalled, "They finally took us back and the doctor looked at his eyes for 30 seconds and said 'Well, he's remarkably unremarkable,' I'll never forget that."

Likewise, when Claire took Dylan to the neuro-ophthalmologist, as referred by another ophthalmologist, she remembers:

> I didn't know something was wrong with his vision. We didn't catch onto that. Of course he had followed up with ophthalmology and got care and all of our follow-ups here at [a local hospital for children]. He had been referred to a neuro-ophthalmologist who told us the nerves and everything was fine so, 'he can see. We just can't test how much. The nerves are fine so the vision should be fine.'

Several parents who indicated that they had difficulty finding answers, or that they felt dismissed by professionals, had concerns that they had missed points in time where development and learning would have happened if only they had known about CVI and how to help their children. Parents reported that they felt determined to find information and figure out what to do, even though they felt a lack of assistance from physicians. Dr. Valle spoke to the lack of professionals who are knowledgeable about CVI, indicating that pediatric ophthalmology is not a field that many professionals seek out, that it is a "small specialty". He indicated that it requires more coursework and generally pays less, and that few ophthalmologists want to work with children. He said he enjoys it, but that he "can't find enough residents who agree". He also said that pediatric optometry is a growing field, and that may help with some of the difficulty finding a pediatric eye specialist. This lack of professional knowledge, and lack of professionals *with* knowledge may contribute to delays in identification of CVI in children.

Delayed diagnosis. Although their children were diagnosed with conditions that are often associated with CVI, only two of the eleven parents/sets of parents indicated that they did not have a "wait time" between when they suspected a vision difference and when the diagnosis of CVI was made. Some parents had children

whose medical and other developmental needs outweighed the visual needs at first, but they reported that it changed as the children grew. Parents reported difficulty connecting with their children due to lack of eye contact, and that there were emotional aspects of seeking answers about their children's visual delays and not getting those answers. They all agreed that they had needs that went unmet during their search for information and understanding.

The experiences of parents who participated in this study varied, but most indicated that there were delays in getting their children's visual impairments acknowledged, despite that they asked medical professionals for assistance. Some parents had to wait several years after they suspected something was different before it was confirmed by a doctor, while others reported that when they mentioned CVI to ophthalmologists, they were told it was not CVI. For example, Tracey, Andrew, Paul, Mary, and Dylan all had one to two years from the time they started having concerns to the time of diagnosis with CVI. Gwen and Bobby waited approximately two years, while Brady and Harrison had more than three years between suspicion and confirmation. During the time period between when parents notice something is wrong and when they get a diagnosis is often a period of uncertainty and questions.

When asked about delayed diagnoses, Dr. Ross acknowledged that parents and physicians must prioritize. He spoke about a case he was working on at the time, where there was so much going on with an infant on his caseload that the family as well as the professionals had to prioritize. He stated:

> They were managing the ventilator and the super high risk and all the other things that were going on. Well… for this child [they can deal with vision later] because they're still hospitalized but, at some point they do go home and they do get lost. So, I think that pediatricians probably need a refresher on availability of resources for across the disability spectrum.

Dr. Ross suggested that pediatricians be on the lookout for such things, and that it may improve the frequency that children get diagnosed with low incidence disabilities, such as CVI, if pediatricians receive information regularly about disabilities and referrals. Many of the parents who participated in this study felt that medical professionals did not have enough knowledge with regard to CVI in children, and that this resulted in delayed diagnosis and parents' need for clarification around their child's vision differences.

Seeking clarity. There were several reasons that parents in this study had difficulty figuring out what was happening for their child and needed clarification about the nature and extent of their child's visual impairment. Some children showed unusual behavior that initially seemed like attentional issues, while others indicated that there were so many other issues facing their children that they did not know where to start or what to do. In addition to the information related to the first research question, this subtheme also addressed the second research question, "What needs do parents recall related to information and supports while seeking a diagnosis for their child's suspected vision challenges?"

Charles described the difficulty they had understanding some of the characteristics of their son, Harrison:

> But it was all so inconsistent that we could never figure out ... The therapists always ask us, "Well what do you think he can see?" And we never had an answer for it, because every time we'd work with him it seems like he could see different things. It's just sometimes he couldn't see below, sometimes he couldn't see ... We couldn't tell if he could see colors, we couldn't tell if he saw things up close better or far away better […] and every time we tried to sort of pin it down, it was elusive because it was so inconsistent […]

Raina had also recalled the first time she heard the term "CVI" in terms of Harrison's education:

> I don't know, we just started seeing how it really became the most pronounced disability for him and it was at the April IEP meeting that one of the TVI's, the [Orientation and Mobility Teacher of the Visually Impaired] was like, "You know he has CVI." And I'm like, "What's that?" We just kept going through all of the symptoms and behaviors and that's when we started reading about it.

Veronica also indicated that it was difficult for her to cut through everything that was going on with Brady. She first saw that Brady had CVI on some referral paperwork that resulted from an appointment that her husband had attended. Although she did not recall if it was from a doctor appointment or from a meeting with his educational team, she indicated that it was unclear, what action she should take. When I asked Veronica what she remembered from that time, she said:

> They did give me a printout […] I have a list of, I don't know, maybe 10 different [practitioners]. It's literally on my pile of, "these are important papers I want to get to." And it's been there for three years because I don't know where to start. I

don't know anything about any of those different practitioners, I don't know who's gonna do what. I don't know what [Brady] has the capacity to do."

During the time between when parents started seeking a diagnosis for their children's visual impairment and when they found the answers to the questions they had been asking, many of the participants felt that they had to work to find the answers themselves. Several felt that they should have been able to get answers sooner, and that the information should have been readily available when they first mentioned that they thought something was unusual about their children's vision. Dr. Valle agreed that the onus is and has been on parents to keep asking questions. He said that when parents get results that indicate "the eye is fine", if visual behavior is not accounted for and the physician does not know why, parents should then ask if the doctor believes it could be due to brain involvement. While obtaining diagnosis is a critical step for parents affected by CVI in their children, it is the first of multiple steps toward obtaining services to help the children learn.

From Diagnosis to Services

The second major theme found during this study was that participants in this study described interesting and varied experiences in obtaining information and ensuring services *after* their children were diagnosed with CVI. This speaks directly to the third research question, "What kind of information is offered or readily available to parents upon diagnosis of CVI?" Once some parents had the diagnosis, they knew where to look and what to look for, and indicated that from there it was not as difficult to get the information they needed from professionals. This did not apply

to all parents, however, and this dichotomy developed into the first of three subthemes, “Information received from professionals”. Some parents had difficulty obtaining services that they deemed appropriate to help their children make educational progress, which is addressed by the second subtheme, “Lack of educational knowledge and understanding”. The third subtheme, “Changing parenting style and learning new skills”, addresses the shifting actions of parents as they adjusted to the new information they had obtained.

Information received from professionals. Several parents in this study indicated that the information they received at the time of diagnosis, or when services began, was critical in terms of leading them in the right direction when it came to their own research on CVI. For some, it felt important to do their own research and follow through to make sure their children were receiving the *right* services and to know what to do at home to help their children learn, because they did not feel that they received enough from professionals.

Tracey recalled several sources of information that were helpful when her child was diagnosed, including a pamphlet and a video about CVI, provided by the TVI from the state’s school for the blind. The TVI also explained things that she was doing with Phoebe so that Tracey could do them the same way. This was helpful information to have received after Phoebe was diagnosed with CVI. Before the official medical diagnosis, however, the state’s Deaf-Blind Project provided “a lot” of information for Tracey, because the educational vision assessment they had performed led them to suspect CVI. .

Likewise, Claire received information she deemed helpful when Dylan was assessed by a TVI using the CVI Range. Claire also found references in handouts and recommendations to purchase a specific app and a specific book, which was written from a family's perspective. Nadia and Boyd also had a positive experience in terms of the information they received after a CVI Range assessment, after they had driven Paul to Pittsburgh to see Dr. Roman. They said it was the best appointment they had ever had:

> She provided us with the evaluation report that she had done. She then sent the write-up on it later. She gave us tricks for what to do with him, what we need to focus on, what to tell people who came in to give him therapy, what they need to do. She provided that information. We were all over making sure that we shared that with everybody.

In contrast, Veronica felt that the information she received when she found out about Brady's CVI should have been more detailed and focused on how to use particular resources, noting:

> Have an idea of what kinds of things are what, instead of giving me a list of names, tell me 'this [person] would work on responding to stimuli, this [person] would work on computerized programs that tell you whether or not his eyes are activating.' Tell me what the different options are, what the different therapeutic strategies are that each one's gonna offer me, not just a list of names and I have to figure it out.

Similarly, Marla reported that the ophthalmologist did not offer additional information nor resources about CVI, though he did agree to give Gwen the CVI

diagnosis after Gwen's PT discussed her observations with him. When Gwen's first CVI Range was completed, by a TVI, the report that Marla received had included recommendations for interventions and presentation of things to encourage Gwen to use her vision. Seeking even more information, Marla asked Gwen's developmental pediatrician about CVI and the doctor responded that she did not know about it. However, Marla did remember receiving some information from that office, in the mail, at a later time. While Marla appreciated that the doctor or office staff took time to find information and send it, and found it helpful, she was also seeking out a professional who could help them at home.

Although many parents realized that finding information was much easier after they understood what to look for, quite a few continued to struggle to secure intervention services that helped their children make progress. Some parents felt it was important for them to learn as much as they could so that they could help, advise, and/or critique their children's educational teams and educational accommodations, especially in cases where educators did not seem to quite understand how to work with children who had CVI.

Lack of educational knowledge and understanding. Unfortunately, even after diagnosis, parents reported that they often encountered even vision education professionals who did not understand CVI or know what to do. For example, Marla and her family made the decision to move to another school district, at least in part due to Gwen's special needs and the experience they had in her original school district.

Marla mentioned her frustration with Gwen's first preschool experience, and her impression that the educational team thought she was being unreasonable. One of her main concerns was that if she, herself, did not know what accommodations Gwen needed, (including color highlighting of both text and salient features in photographs, reduced visual complexity, backlighting of images, and more) she would not be able to judge the quality of the classroom accommodations, and that she worried about whether her daughter would receive high quality instruction. She concluded that she would have to learn more about what was appropriate for Gwen:

> I started feeling overwhelmed and like I didn't have a handle on what adaptation should be in place… I started to doubt […] the different staff members' ability to implement [Gwen's CVI Range Assessment Recommendations]. […] The vision teacher that they had assigned […] didn't have any CVI experience. So then I felt like, "Oh man, the person that's supposed to be kind of leading the team on vision issues, does not seem to be able to do that."

Marla also preferred to get as many specialists as possible to help her daughter learn and try to overcome some of the developmental delays she has experienced, such as private speech, occupational, and physical therapists. After Gwen was identified as demonstrating characteristics of CVI, trying to find someone to help with Gwen's vision in a similar fashion to those practitioners was not easy. She sought out various physicians and asked them if they knew anyone who could help with Gwen's CVI, and the responses were often that they would need to just wait and see, that there was not really anything she could do, etc. Since then, she has educated herself about CVI and is trying to help accommodate Gwen's environment at home as

well, but she has multiple children and does not always feel that she can devote enough time to it herself.

Lisa had a similar experience with lack of experienced school personnel, but in her case they had recently moved away from a district where she felt Bobby had a knowledgeable TVI and was getting good vision services, though she had not been entirely happy with other components of his educational experiences there. She expressed concern that there seemed to be no one in the new district who truly understood CVI, and that his vision needs were a primary concern if he was expected to learn throughout the school day.

Jennifer's frustration has also been with the educational system. For her daughter, Gail, the school system had written an IEP that included "plenty" of hours with a Teacher of the Visually Impaired, but with the purpose of teaching her Braille. Jennifer had previously attended a conference about CVI, in which several experts had said that braille instruction is usually not appropriate for children with CVI. Jennifer recalled:

> They did let us remove the braille, [but] they did not agree […] and documented that heavily in all the paperwork. And it basically says, you know when 3rd grade testing comes and she's behind then you could always go back to braille. Kind of like a 'we told you so' and we had Dr. Roman on the phone and [a TVI and CVI Mentor]. I think [the CVI Mentor] said, "there's an expectation for change. You have to go into this fully expecting that her vision is going to improve, not that it's *not* going to work."

In contrast, some parents acknowledged that TVIs were helpful in many areas of their children's education. For example, without a knowledgeable TVI, it may be difficult to expect non-vision services professionals, such as Occupational, Physical, and Speech Therapists, to know how to incorporate CVI into their work with children who have this diagnosis. Lauren and George's son had a vision teacher who knew about CVI, and that TVI offered to observe other professionals (e.g. OTs, PTs) during their sessions. Lauren said that having the vision teacher help advise others - the PT, for example, was invaluable. They explained that the physical therapist had been trying to motivate Andrew to move by placing "high value" toys nearby. The consult with the TVI and the PT together helped the PT understand how Andrew's visual impairment impacted his ability to see things like that. Some parents indicated that they had learned a great deal about how to present items to their children, as well.

Changing parenting style and learning new skills. Some parents in this study felt that they needed to become experts themselves because it was difficult to find a teacher who understood and could provide the unique accommodations their children needed to learn. When they found a teacher who truly understood their child and their child's vision needs, they felt that it made a difference.

Several parents noted that many of the things they would customarily do for typically developing children, such as multi-colored blankets, pastel colors, and lots of toys, were not necessarily appropriate, nor helpful, for children with CVI. Lauren and George pointed out that ordinarily they would use brightly colored blankets and offer lots of toys to their child, but that did not work for Andrew. The TVI taught them to use solid, dark-colored blankets and offer just one toy. They said that once

they knew how to offer things, Andrew started playing and interacting in a way he had not done previously.

Nadia and Boyd also experienced the need to change their frame of mind, and expressed frustration that they did not know what to do sooner, "We were doing the wrong things. We had no idea. We did not know that we were supposed to be […] making sure things are easy for him to see…none of that." They also remembered having someone from a local agency for individuals with hearing impairments come out to see Paul, who would sign books to him, but later realizing that Paul could not see the books nor the sign language. Dylan's mother, Claire, had a similar experience in terms of parent knowledge, and she indicated that it helped her to be open to possibilities, summarizing, "You don't know what you don't know." Experiences such as this helped many parents process the emotions that accompanied having a child with a disability that had gone unrecognized by medical and/or educational professionals.

From Frustration to Hope

When describing the experiences of parents, as the first research question for this study was intended to do, it is important to acknowledge the emotional elements of the experience. Parents in this study had mixed feelings about the whole process, from working to get the vision condition recognized and diagnosed, to getting the educational assessment, and acquiring appropriate intervention services which would help their children make progress in school. While many communicated a sense of

relief that they knew what to do from the point of diagnosis onward, they also felt a continued struggle to obtain the appropriate services.

"It's a struggle." The participants in this study were asked about the emotions that they experienced while seeking and after receiving a diagnosis of CVI for their children. Some parents experienced stress – becoming exhausted, angry, or overloaded with the task of caring for their child's everyday needs, going to frequent and lengthy doctor appointments, and doing research to find out what more they could do to help their child learn and function as best they can in the world. Some were frustrated with the process of getting recognition for their child's visual impairment, and that sometimes getting an actual diagnosis is even more difficult. Charles conveyed this frustration:

> Just the fact that we had to wait until a TVI mentioned CVI to us, when Harrison was five years old, or four and a half years old, is crazy. We've been to pediatric ophthalmologists and geneticists and neurologists and […] some of these doctors should have identified CVI as at least a possibility.

Raina continued, describing their experiences at a medical research facility where Harrison was seen multiple times. She said that Harrison had been there all day, three times, for testing. She was frustrated, stating that no one had mentioned that he could have a visual processing issue, medically nor educationally. Raina commented:

> It's a lot. […] Realizing how much needs to be… the interventions that need to be in place and the accommodations that need to be in place for him to make

> progress is really… it's daunting because we're gonna have to really step up our advocacy game to make sure that his educational experience is the best it can be. And we're in a really strong school system, but he's part of a low incidence population and I've started to hear [that it is low incidence] more and more now, and it's making me angry. I'm like, "so?" but that just means there's not as much funding.

Charles agreed, simply stating "It's a struggle".

Raina and Charles were not the only parents to express stress and frustration. Tracey had a similar response when she was asked about the emotional component of the process, saying, "It's a lot. It's a lot for a regular person to take in. It's a lot for a parent who's dealing with everything else to take in." Both Raina and Tracey indicated that there were particularly stressed by the level of difficulty in locating information and services to help their children.

Because CVI has not been well-recognized, the added stressor of having to teach or train professionals, and struggle to get accommodations, weighs on some parents. Bethany explained some of the sources of her own frustration, "I think the emotional piece is really hard, and they're like 'I have never heard of that'. […] I think having to constantly explain your child […] you get more than anything, 'well what does he see?'" She also mentioned that when well-meaning people suggested perhaps getting together with other families of children with visual impairments would help her, she felt that she could not relate to them, and that she felt alone at times, "it's very, very different. The ease with which they get things that we can't, we can only dream of… the supports that they have that we don't have, maybe don't even

exist for our kids." Bethany said that she often thinks, "You have no idea what I'm talking about. Literally none."

The stress that parents of children with disabilities experience is not a newly introduced concept. However, the uniqueness of parenting a child who has a disability that is not well-known, even among professionals, leads to additional stressors such as having to explain the disability and struggling to find information about how to help the child, leaving parents feeling frustrated and, sometimes, alone. Even so, many parent participants of this study were able to find hope in what they learned.

"These are things we can definitely do." A few parents in this study described their experiences with diagnosis and intervention as hopeful. CVI is a diagnosis that has known strategies for improving the visual processing in children who have it, so while a diagnosis may confirm the type of visual impairment, it also provides an opportunity to take action to improve it.

After wondering for so long what Gwen needed, Marla referred to the overall identification and assessment process as, "a good thing that we're learning about CVI and [Gwen's] ability to process vision." Marla said that she felt like the CVI Expert who assessed Gwen truly understood, and that "everyone [at Gwen's school] felt good about it" when the CVI Assessment Report came back, because it made more sense than some other evaluations. Marla felt hopeful after the assessment was completed, because it was a relief to get the results and have it confirmed that "there was some issue going on." She recalled that the experience of talking with the TVI

who assessed Gwen was positive, even though at the time she did not understand CVI:

> It was like after everyone at her school that worked with her, had read [the report…] they all wrote back to me, like 'wow this is so great'. After I sent them the report, they were so excited in a way, and some of them used that word, were so excited, like just to know that these recommendations are so helpful, these are things we can definitely do. There was this real kind of almost celebration.

Hope was also evident in some of the things Lisa said about her son, Bobby, and his experiences with CVI. Bobby had been crawling, pulling to stand, and walking, when he started having seizures that affected his skills. He had to relearn those motor skills when his seizures were under control, but he struggled to do so because of his visual impairment. Lisa remembered the positivity when she was first meeting Bobby's new TVI,

> I hope that his vision teacher now… she made a comment when I first met her that "He sounds like he's gonna be a lot of fun." And I liked that comment because he is fun and he's really smart in so many ways. So I hope she means that and she feels as good as I do when I watch how he's learning how to adapt.

Other parents also said that they continued to be hopeful for the future of their children. Lauren and George felt relieved when they received the diagnosis of CVI for their son, Andrew, who has history of very complex health issues. When asked what kinds of emotions they felt when Andrew was diagnosed, Lauren said, "I was thrilled. [...] First, it explained some stuff that was otherwise unexplained. […] Things were very frustrating to us. So it was very exciting to get a reason for [the

visual behaviors]." They remembered thinking that a CVI diagnosis was a good thing, because it meant that Andrew would qualify for additional services and that his visual behaviors would be addressed.

In the same way, Jennifer felt hopeful as well, because she had noticed improvement in Gail's vision – Gail was recognizing things and had increased visual curiosity - since getting the CVI diagnosis and implementing interventions. Furthermore, Claire said that she remembers reading the "Little Bear Sees" book and that she found it comforting to read the parents' perspective that it provided. She felt hopeful after reading that there was something she could do to help her son use his vision better. She said:

> Visually, there's definitely been growth […] He recognizes us for sure... and he uses his vision, a lot. So I feel good because I see the difference in him. […] I still hold him and rock him ... and I'm like 'what are you trying to say to me?' It's almost communication but he looks dead on and it's that connection. It's wonderful.

This is in stark contrast to parents' experiences and feelings when their infants are not able to make eye contact with them.

Some parents reported feeling hope after receiving confirmation of the CVI diagnosis, in part due to the additional information they had also received. Nadia and Boyd remembered that after they drove to Pennsylvania to see Dr. Roman, they felt hopeful because she had provided so much information.

In addition to feeling hopeful after their children received a CVI diagnosis, some parents also felt relieved that they understood more after getting the diagnosis than they did before. Veronica thought back to a conference she had attended, as a professional, after the family had been told that Brady was "blind." It was at the conference that she realized that since Brady still had vision that he used, "blind" may not have been the right word:

> It was only when I went to the [state's] Blind Deaf Conference and […] they had this whole big speaker talking about Cortical Vision Impairment […] I didn't bother to figure out what that meant until I was at that conference […] I've studied it on my own because of that conference and learning about it […] I gathered the information that he very may well see everything that's around him and his brain just isn't able to filter it, understand it, remember it, process it, or do anything with it.

Interestingly, Raina and Charles observed their son's difficulty coping with stressful situations, and noted that the CVI diagnosis explained a great deal of his behaviors that had been a mystery. Harrison had a history of low muscle tone, feeding difficulties, and allergies. It was difficult for them to get answers about why he demonstrated difficulty attending to tasks and why he would have "meltdowns", and Charles said:

> [We read about] CVI meltdowns…and I was like ...that's him. […] [He] has a pretty highly irritable frustrated anxious baseline already because he can't see the visual world. […] It's just a lot of hitting and frustration and acting out in

> the classroom and we've just been totally confused by this because it seemed like a discipline issue and we just tried a million different things and nothing made it any better until [Raina] came across the article on CVI meltdowns and […] that's exactly what it is.

Parent participants who found the answers they sought, and who felt that they were headed in a desirable direction, expressed positive feelings such as hope for the future of their children. The often frustrating process they had undertaken to get CVI diagnosed, followed by efforts to locate and secure services that were appropriate for their children, did not remove the hopefulness they were able to feel, despite the anticipated likelihood that they would have to continue to work hard to ensure their children continued to receive those services. Participants in this study were willing to share some words of wisdom with other parents, and with professionals.

Advice for Others

The parent participants of this study had all educated themselves about CVI in some way – some attended workshops, others read about CVI independently, and some sought out information by any means they could. Parents have an important perspective when it comes to the medical treatment for their children and interaction with medical professionals, therefore, the parents who were interviewed were asked if they had any advice for professionals in the field of visual impairment, as well as other parents whose children demonstrate characteristics of CVI. Although this does not address a research question directly, it does speak to their experiences, what they took from those experiences, and what they would like others to know.

Advice for professionals. Nadia and Boyd had a strong reaction when asked about their advice for professionals, and Boyd did not hesitate:

> I mean, diagnosing it and not being afraid to have a conversation with the parents about it is the first thing they could do better. Learn about what it is ... 'cause too many people have no idea what it is and what it entails. That's a big thing.

Nadia agreed, saying:

> [Professionals should] figure out what questions to ask when you have a severely disabled child – to be able to identify whether or not CVI is on the table, and if CVI is on the table and you are not the best person to evaluate, immediately refer the parent to a person who can evaluate. [If] someone would've checked him out when he was an infant and been like 'pretty sure this is a thing', that would've made a huge difference.

When asked for her recommendations for professionals, Bethany had a difficult time putting it into words because she felt there were so many things to improve. In the quote below she references people who are "CVI endorsed", referring to a professional endorsement that may be obtained from Perkins School for the Blind, which may be an indicator of the professional's knowledge of CVI. Most of her thoughts were concerning TVIs, and she said,

> I feel like there's so little support through the TVIs, or let's say the traditional ones, and the people who know CVI and are CVI endorsed are exceptional. But

> they need to know so much more […] I guess just maybe to learn as much as they can ... they need to always be learning about CVI.

Alternatively, Claire's thoughts were focused on physicians and what they need to do to improve the diagnosis process for children with suspected CVI. Claire works at a local hospital, helping parents of children with disabilities or medical complications navigate the healthcare system. She said:

> This is something that exists out here that you need to know about. You have patients that could come through your doors and parents need to know. And the sooner the better, right? Know the signs, educate yourself, and listen to parents [about] what they're seeing. […] Educate yourself.

Interestingly, Dr. Ross, a neuro-ophthalmologist who diagnoses CVI and receives regular referrals from TVIs who believe they recognize CVI in some of the children they work with, had a suggestion when asked about families' access to CVI diagnosis and services:

> For some families, they at least know but, the patients who are … in the lower end of our socioeconomic spectrum, [it may be more difficult to access service providers and supports] because they're not ... They're [living] day to day, and can't spend the time to say, "I'm going to make fourteen phone calls." It's almost like [the state] needs a single contact phone number for child disability call. Then, they sort of can help triage out to the right agency. [It should be noted that the Child Find system *is* a single-contact resource for children with suspected disabilities in the state where Dr. Ross practices.]

Parents who have been through the process of identifying an existing issue, seeking diagnosis, and securing services are uniquely qualified to make suggestions to professionals about how they can better help and serve other families. As such, I also asked for their perspectives and if they had any advice for other parents who face similar challenges.

Advice for parents. When asked what advice she had for parents, Marla said that she would encourage them to make adaptations and accommodations right away, regardless of the age of the child. She suggests that if parents believe something is not right, they continue to seek help and guidance, even after they are given a "normal eye exam". She said:

> Wherever you are in catching it, really try to make an effort to learn about CVI and find ways to incorporate the accommodations into your daily routine, and not ... dwell too much on that you didn't catch it until now.

Nadia and Boyd suggested that parents have their children evaluated by someone knowledgeable, as soon as possible, which is the consensus of parents and professionals when speaking on early intervention. They acknowledged that it may be difficult to seek out additional diagnoses, but that it is worth the additional effort because identifying CVI and making adjustments can only help the child learn. Lisa and Claire both agree with that sentiment, and suggest that if parents suspect a vision issue and need help, they contact their early intervention team right away to get services started. Claire said:

> If you do suspect there's something, then you don't stop. If you're not getting the answers there, then you talk to people. You try to go beyond them because

there are people out there that do have the answers and to recognize it. You may not know what it is you're looking for, but if you know that there's something going on - because [parents] all know when something is not right - visually something is going on, you don't stop until you get an answer … Some families, it can be overwhelming. It's just one more thing. But just be open. Be open to hearing it and learning and educating yourself about whatever. The CVI, the condition, because there are things and steps that you can do to improve your child's vision. Just be open as a parent. We have to be our kid's advocate, we all are. Don't stop.

Jennifer also had a few suggestions for parents. One thing she said that was different than many of the parents' responses to the question about advising other parents, was that over the course of time, parents may want to build their own personal advisory board. She also suggested that parents request a Perkins-Roman CVI Range-endorsed professional be on the team of service providers.

When I asked Dr. Valle what he would suggest to parents in terms of getting the right diagnosis, to lead them to the right interventions, he said that parents need to be persistent, and know what questions to ask the eye care professionals, "Is my child's vision okay? If it isn't, is it the eyes? If it isn't, what is it and where do we go?"

Some of the participants in this study recommended that families also seek out other types of support, besides professionals who can help them find answers. Lisa belonged to at least one large group of parents who advocate for children with specific genetic mutations. Similarly, Nadia mentioned that she sought out support

and advice from other families of children with tracheostomies. Claire also highly recommends that parents network with other parents who are in similar situations:

> Unless you live it, you just don't know […] a peer-to-peer support group I think would be wonderful ... [professionals are] looking and searching but how great would it be if [they can] say, 'I'll come out and see you but there's this group of parents and they meet regularly...' Or even if it's a phone thing, whatever.

After the interviews with Claire had concluded, I sent her information about a weekly conference call for parents of children with CVI, and I also told her about other groups that existed for parents online. She strongly recommended parent support groups, but was not aware of these options at the time. When I asked Dr. Ross about his thoughts for parent support, he suggested that the state's Child Find system is not completely filling the need in terms of finding children with disabilities. He seemed particularly concerned about families with lower socioeconomic status, as indicated in the quote from him earlier in this chapter. Parents and professionals all contributed thoughtful advice and concern for families of children who have CVI.

Summary

The participants in this study agreed to openly discuss their experiences in getting a CVI diagnosis and services that were appropriate for their children. The themes that emerged from the cross-case analysis of parent interviews include parents' views about delayed diagnosis, lack of professional knowledge, and how parents sought to clearly understand what was happening, as in some cases CVI looked like other things and there was a lot going on with some of the children. Additional themes arose around post-diagnosis information sharing between parents

and professionals, and interventions that children did or did not receive. Parents also found that they had to learn new ways to parent and new approaches to use with their children, to help them learn. Finally, themes that centered on emotions, including stress and hope, were presented in this chapter, as is parents' advice for other parents as well as for professionals. The following chapter will summarize the importance of this information in the context of theory and related research, and define some potential next steps in the area of CVI diagnosis and intervention.

CHAPTER V

DISCUSSION

Cortical Visual Impairment is the leading cause of visual impairment in children in developed countries (Good et al., 1994; Groenveld, 2003; Hyvärinen, 2005; Khetpal & Donahue, 2007). There has been an increase in the population of children with conditions associated with CVI, due to increased survival rates of infants born with these conditions (Good et al., 1994; Khetpal & Donahue, 2007; Murphy and Carbone, 2011), and a reduction in the number of children with permanent ocular conditions (Good et al., 1994; Groenveld, 2003; Hyvärinen, 2005; Khetpal & Donahue, 2007). Nevertheless, there are disparities in the level of awareness, professional knowledge, and receipt of appropriate and timely services provided to children with CVI. The purpose of this study was to describe the experiences of parents as they sought to obtain accurate diagnoses for their children. In this chapter, I summarize the findings of this study and discuss their significance in the context of previous literature focused on CVI, parent perspectives about diagnosis of disabilities in their children, neuroplasticity and sensitive periods, and Bronfenbrenner's Bioecological Systems Theory. I also discuss implications of the current study and recommendations for future research.

Summary of Findings

During the process of analyzing the participants' interviews, several themes and subthemes emerged. These themes addressed the research questions that were developed before the study began. "Something was Wrong" speaks to the question

"What are parents' experiences in seeking a diagnosis for their child's suspected vision challenges?" because perceiving that "something" was different about their children's vision guided parents to seek medical advice. The theme "From Diagnosis to Services" includes information about "What kind of information is offered or readily available to parents upon diagnosis of CVI?" which was the third research question that was developed for this study. To acknowledge the emotional aspect of parents' experiences, the second research question was, "What needs do parents recall related to information and supports while seeking a diagnosis for their child's suspected vision challenges?" which is addressed by the theme "From Frustration to Hope," which emerged from the interview data. Lastly, the theme "Advice for Others" resulted from interview questions that were asked to find out what the parents wanted to pass along as "words of advice" for other parents as well as professionals.

The parents of the nine children who had delayed diagnosis said that they wished they had known about CVI sooner and that they had received more information about it both before and after their child was diagnosed. Many were frustrated that they had to see multiple healthcare providers to get a diagnosis and an explanation for what was affecting their children's vision. Parents also said that even after diagnosis, they still had difficulty obtaining and keeping appropriate services for CVI. At the same time, in hindsight, some parents said they felt relief and hope about their children's vision once they understood what was happening.

A major related finding was that most parents expressed that professionals often demonstrated lack of understanding, knowledge, and training about CVI. The results of the Jackel et al. (2010) study indicated that the participants in their survey

experienced encounters with professionals that lacked understanding, knowledge, and training about CVI, and that this lack of training made it more difficult for the parents to secure appropriate accommodations for their children. The results of this study are consistent with those findings, in that the lack of professionals who understand CVI affected how much information parents received and how quickly they received it, as well as the rate of diagnosis. In both studies, some parents were told that there was no intervention that would help. Several participants in the current study described having to go to several doctors and specialists to receive a diagnosis, after hearing from other doctors to "wait and see" or the child was "too young to tell".

An additional finding was that parents who did not have difficulty in getting a diagnosis for their children, still had difficulty getting and maintaining appropriate services, as did their counterparts who did have delayed diagnosis. Many parents had difficulties with receiving the diagnosis, securing appropriate services, and keeping those services. Parents continued to experience stress and frustration related to the diagnosis of CVI because of the difficulty they had obtaining and keeping services that were appropriate for their children over time. The parent who did not describe frustration as a primary experience was a mother who had a child that was so medically complex, his vision was not a top priority for the family – though she did express that she wished she had time to pursue it. Most of the parent informants had ongoing concerns about whether their children were receiving and would continue to receive appropriate services. Vision is something that individuals use all day, every day. Newcomb (2009) suggests that, since children with CVI typically are served by multiple providers from multiple disciplines, intervention training and/or training to

enhance understanding of CVI should extend beyond just physicians and TVIs, to include related service providers such as special educators, occupational therapists, physical therapists, speech-language pathologists, teachers of the deaf and hard-of-hearing, audiologists, and various medical professionals. A few parents worried that they would have to revisit these concerns at annual IEP meetings at least, to ensure the interventions and accommodations were in place, that the IEP team understood CVI, and that instruction was appropriate. They described stress in the form of grief, anxiety, and exhaustion about this issue during the interview process.

Several parents in the current study also reported that eventually they had experienced positive feelings, such as relief and hope. They had the knowledge and empowerment to advocate for their children, and created relationships by networking with other parents who understood what they had experienced. Five of the families interviewed specifically indicated that they had educated themselves about CVI through their own research and social networks. The parents who specifically expressed feeling hopeful also mentioned that they understood that their children's vision could improve with the right interventions.

Parents also contributed their thoughts for increasing professional attention toward CVI, and ways that physicians and teachers can help parents. Such advice included that professionals (including TVIs, pediatricians, pediatric eye care physicians, and pediatric neurologists) need to know that CVI exists, should be open minded about the possibility of a CVI diagnosis, should be willing to discuss CVI

with families, and need to be willing to refer families to other practitioners if they do not themselves possess the knowledge to make a diagnosis and recommendations.

Connection to previous studies. The findings here are particularly important in light of previous research, which has found that delays in diagnosing CVI create lost opportunities for learning, and that earlier detection and diagnosis may improve outcomes for these children (Baird et al., 2000; Davies et al., 2003; Dutton et al., 1996; Huo et al., 1999; Khetpal & Donahue, 2007). Timely diagnosis and intervention for children with disabilities, in general, has been identified as an important factor in reducing the impact of disabilities on development (Addison, 2003; Davies, et al., 2003; Halfon et al., 2004; Peters, 2010). The concepts of sensitive periods and neuroplasticity suggest that earlier identification and intervention are key to reducing the impact of CVI on the child and family (Cohen-Maitre & Haerich, 2005; Good et al., 1994; Good, Jan, Burden, Skoczenski, & Candy, 2001; Hoyt, 2003; Huo, Burden, Hoyt, & Good, 1999; Khetpal & Donahue, 2007; Lam, Lovett, & Dutton, 2010; Lueck, 2010; Malkowicz, Myers, & Leisman, 2006).

Jackel, Wilson, & Hartmann's study in 2010 was survey-based, and the questions included four basic categories: parents' suspicion of a vision issue, diagnosis and information provided to parents, etiology, and services the participants' children were receiving. The first two categories in particular inspired the research questions upon which the interview questions were based for my study. I sought to explore these questions in a qualitative manner and compare the experiences of

participants for my study and those of the survey participants in 2010. Like the study described in this work, the 2010 study also demonstrated that parents would like to be provided with more information about how to help their children, and parents feel they have to do research on their own to find out more information about CVI when it is diagnosed (Jackel et al., 2010). This is consistent with the sentiments of the participants in the current study, who stated that there is a deficiency of information provided to them regarding diagnosis, awareness, and appropriate services for children with CVI.

Results of the current study indicate that parents believe if professionals had followed through with investigation of children's vision difficulties based on parental concerns, the children may have been diagnosed earlier. Parents' concerns were especially significant when considering previous research about diagnosing CVI in children, and were consistent with those obtained in previous studies of parents of children with other types of disabilities, as well (Baird et al., 2000; Howie-Davies and McKenzie, 2007). Under the lens of Bronfenbrenner's *proximal* processes, parents' experiences and concerns should be taken seriously, as they are the primary means by which a young child learns (Rosa & Tudge, 2013). Explicitly, parents want their concerns to be taken seriously, do not always agree with a "wait and see" approach, and sometimes they feel that professionals do not understand the parent's perspective (Addison, 2003; Davies, et al., 2003; Klein et al., 2011; Peters, 2010).

Parental stress and frustration reflected another theme identified in this study, which is consistent with the findings of other researchers who examined disability

and parent perceptions. Parents in this study expressed frustration with getting diagnosis (if diagnosis was delayed), as well as obtaining and keeping interventions in place for their children. As reported in previous studies of parent stress and diagnosis of disabilities, delayed diagnosis results in additional stress for parents (Davies et al., 2003; Graungaard & Skov, 2006; Moh & Magiati, 2012). The additional experiences of parents of children with CVI as they try to get information and navigate medical and educational institutions that do not have professionals who understand CVI, in terms of Bronfenbrenner's Bioecological Systems Theory, results in stress. This stress affects and is affected by the child's immediate environments (*proximal processes* and *microsystem*) including the family home, the child's classroom, and the relationships between the *microsystems* (*mesosystem*) such as the relationship between the school service providers and the family (Bronfenbrenner, 2001, Rosa & Tudge, 2013).

Another important point with regard to parents' experiences with having a child who has an unidentified, undiagnosed, and/or uncertain diagnosis is the total impact on families, aside from the direct impact of the condition itself. Research on parental uncertainty has revealed negative effects such as increased stress and risk of anxiety, depression, and health consequences (Ablon, 2000; Lenhard et al., 2005; Rosenthal, Biesecker & Biesecker, 2001). Based on parent reports for this study, one may also conclude that the parents who must do additional research (such as finding a qualified professional to provide services, appropriate interventions, and being the "expert" that must train service providers) are likely to devote a great amount of time to these activities. These potential issues may add to those that parent participants

revealed for this study, creating situations where parents have multiple atypical stressors. Bronfenbrenner's Bioecological Systems Theory may be applied to these issues, as well.

Lack of early identification of CVI, leading to the absence of appropriate early intervention and the potential need for longer-term assistance overall, also is impacted by (and may impact) governmental policy (*exosystem*), in that school-age children who are not diagnosed with visual impairments are not typically supported by teachers of the visually impaired, which then affects how federal quota funds for children affected by visual impairments are allocated (American Printing House for the Blind, no date *b*). Furthermore, children who have undiagnosed CVI are likely to require some school services, be it special education or other, due to learning struggles resulting from the reduced ability to understand visual information, and these services may be less effective when CVI goes unrecognized because the educational professionals do not then know how to present educational materials. Additionally, delays in diagnosis can risk the closing of sensitive periods of visual development, during which neuroplasticity is higher and learning to use one's vision may occur most efficiently. Finally, delayed diagnosis is also highly likely to affect (and be affected by) the *chronosystem*, in that the effects at each level also affect one another over time and within the time in history it occurs.

Likewise, the lack of professionals who understand CVI – physicians as well as TVIs – affects multiple *systems* levels. Parents in this study expressed a need to learn as much as possible about the vision impairment issues their children were

experiencing, both so they could do more with their children at home (*microsystem*), and also to be able to critique school-based interventions (*mesosystem*). This additional knowledge can affect parents' behavior in their interactions with the child – emotionally, but also in terms of how the parents approach learning and play interactions with their children. The time parents spend to learn as much as they can may also impact the *exosystem* (e.g., parents' place(s) of work and parents' social relationships and activities). Since these systems are nested and bidirectional, the effects of these issues can spread to other *exosystems* and *microsystems*.

Impact of the Study

While research has expanded in the arena of CVI assessment and diagnosis, there has not been research conducted with regard to parent perception of the assessment and diagnostic process, especially regarding disclosure of the diagnosis itself. Parents in this study expressed having felt relief when their children received a CVI diagnosis. This can presumably be a relief because they have "finally" found an answer, which itself comes with approaches to help improve a child's vision. Unfortunately for many of the participants, having the diagnosis does bring the diagnosis journey to an end, but it usually also takes parents to the next phase of seeking and securing intervention professionals who are well-versed in CVI.

One key result of this study is to provide a starting point for future research. Questions such as "Is it truly difficult to get a CVI diagnosis? If yes, why?" and "What do parents think about this?" are vital to guide research that will directly influence practice. Having a basic understanding of what the participating families

have experienced in terms of what they did to get a diagnosis (if they did), what gives parents hope, and what is frustrating during the process will shed light on the experiences of families and the process itself. This study aimed at yielding information that could be used to influence professional development and increase awareness and knowledge in professionals who can diagnose CVI.

Limitations of this Research

Efforts were made to verify the data obtained from participants in this study, but like the other studies cited in this summary, the present study had limitations which may be addressed with further study. These limitations can be divided into three categories: participants, methodology, and researcher.

Participants. The parents who participated in this study were strictly volunteers, who were recruited via networking with the MD/DC DeafBlind Project and through participant referrals. The highly educated nature of the participants in this study further limits the degree to which the results can be compared to the general population of parents of children with CVI, and therefore leaves additional questions about other parents' experiences. Additionally, parent participants included three fathers and eleven mothers for eleven cases. The addition of fathers during the interviews for those three cases may have affected the content of responses to interview questions. The lack of fathers as participants for the other cases may represent a gap in that data.

Not all medical professionals agree that CVI is difficult to diagnose. For example, those affected can be identified and managed, if professionals are alert to

the possibility that it may be present, and by careful behaviors that are indicative of CVI (Dutton, 2013; Philip & Dutton, 2014). In terms of professional participants from this study, both TVIs and both ophthalmologists that were interviewed had significant pre-existing knowledge of CVI, and are not likely to be representative of the greater population of these specialists. One of the TVIs had studied under Dr. Roman, and therefore had a good foundation to understand CVI. It may be difficult to locate specialists that do not understand CVI and who would be willing to participate in a study about CVI.

Methodology. Interviews for this study were primarily conducted face-to-face, but due to the travel limitations of the researcher, three were conducted by phone, which limited the geographical span of the study. This is important because states vary in terms of available, knowledgeable professionals and means for identification of CVI. Although participants who were interviewed over the phone were responsive and forthcoming, it is important to note that the phone interviews eliminated face-to-face contact. This may have altered the information shared by participants and interfered with my ability to note non-verbal cues such as facial expression and body language, including any physical indications of discomfort such as posture, furrowed brows, shifting in one's seat, pacing, and tears. Lack of non-verbal indicators meant that I had to listen for similar types of clues, using participants' voices, only, so such indicators of discomfort may have been imperceptible. Of note, the only two participants who had children who were diagnosed with CVI immediately were interviewed over the phone. Even so, they both expressed the same difficulties with obtaining and maintaining services as the

other respondents. Additionally, three cases included interviews of couples, rather than of only one parent. This possibly influenced the results because they may have, and likely did, affect one another's responses to the interview questions. Although no parents openly disagreed about the answers to the interview questions, specifically, during the interviews, it is possible that the conversation continued when I was no longer present.

Researcher. In qualitative research, the researcher serves as the "instrument" for data collection and observation (Creswell & Poth, 2017). As a result, the perceptions and biases of the researcher may influence the data collection and analysis. As the researcher, I knew some of the participants in advance of the study, although distantly. When I was recruiting participants, I did not request nor allow parents of students at my school to volunteer because I felt I knew too much about them to be objective. Six of the informants, representing four families, however, were participants in multiple events that were hosted by the MD/DC DeafBlind Project and at which I served as a volunteer, so we were acquainted in advance of the study. My role during these events was to assist with childcare while the parents participated in the event, so my direct interaction with each parent was limited. These informants also had toured the school where I was employed, and I had participated in "Pre-Admission" meetings at which the children's educational needs were discussed to determine if placement in my classroom was appropriate. This may have affected parents' willingness to participate in the study, as well as their comfort level in terms of sharing their experiences. The other eight informants, representing seven families, were not familiar with me before the recruitment advertisement was

distributed. The recruitment letter that the MD/DC DeafBlind Project sent on my behalf was extremely helpful in helping to locate volunteers, and may also have increased parents' comfort level with participating in the study.

In terms of my own bias, my experiences with CVI were primarily work-centered in that I originally started to learn about CVI so that I could better serve my students. My students at that time were not diagnosed with CVI, but all of them had multiple disabilities and had demonstrated characteristics of CVI. Once I learned how to educationally assess for CVI, I realized that it was pervasive within my school. Therefore, I had also taught other professionals at the school about CVI and recommended instructional strategies and interventions for the students. I was bewildered that the students were not receiving vision services from their home school districts because they did not have medical diagnoses of CVI and were therefore not considered by the school systems as having visual impairments. I worked to change this situation for the students that I could, by talking with families to help them understand and to help them to pursue diagnosis if they chose to do so, therefore they would qualify for services. Although I was providing them with the interventions they needed, I was always concerned that those interventions would stop if I was no longer their teacher. These experiences with children and CVI may have influenced the research questions I pursued, and the way I used interview questions with parents and other informants for this study. It may also have influenced my perception and interpretation of their responses. I attempted to mitigate the effects of my own bias, by developing the basic interview questions as a guide, and performing member checks to ensure derived meanings were accurate.

Since this study was qualitative in nature, parents often led the conversation after I asked the initial question. The first interview question was very broad, by design, and often the participants shared enough information that some of the follow-up prompts were unnecessary. I also sought clarification and validation by participants during follow-up interviews.

I documented my reflections during and after the interviews. I wrote that I felt "generously welcomed" into families' homes, and that conversation generally was "very comfortable, almost casual". Some families asked if their quotes could be edited, due to passionate language that some parents used, and I noted that it was satisfying to have parents feel comfortable enough to use colorful language during the interview process. I responded that they need not worry about the words themselves, that I could edit if needed, and that they should use whatever language they felt best communicated their message. I was also aware that the participants were very generous with their time, and seemed to make effort to make me as comfortable as possible, when my priority was to have the participants feel comfortable. One of the professional participants gave up part of his lunch or paperwork time to speak with me, as he fit me into his day with very short notice and parents had relayed that it takes months to get an appointment with him.

Some parents asked me for my opinion, or asked for advice about CVI-related things during or after their interviews. For example, "have you seen that in children you've worked with?" and "are other parents experiencing this?" At times it was difficult to refrain from offering advice, I was concerned about validating parents

without influencing them, and often I waited until after the interviews were complete, and then asked the parents if there was anything they wanted to ask, or if they needed more information about something. Knowing that I am a professional who works with children who have CVI, a few times parents asked what more they could be doing at home. In one instance, the parents had said they wished there were peer supports for parents, and after the initial interview I sent her information about the weekly CVI conference call and online parent groups. I also noted that I felt it was very important to carefully validate when participants shared emotional and personal experiences, making sure that the validation would increase the comfort level of the parent without affecting the story they were telling.

Conclusions

Delays in diagnosis of CVI, from the parents' perspective, can lead to frustration, stress, and missed opportunities for intervention, creating a situation of delayed opportunity for improvement. Additionally, families may experience heightened stress when they find that the delay may have caused them to miss the sensitive period of visual development when neuroplasticity is greater and progress may occur at an increased rate. In terms of Bronfenbrenner's Bioecological Systems Theory, delayed diagnosis affects multiple levels of systems, which distributes the effects bidirectionally to all levels through *proximal processes* within the *microsystem*, and also between the *microsystems* (*mesosystem*), into the *exosystem*, and affecting (and affected by) the *chronosystem*. Likewise, the lack of professionals who understand CVI – physicians as well as TVIs – affects multiple *systems* levels.

The most obvious of examples is the additional stress experienced by parents of children with CVI as they try to get information and navigate a system that does not have professionals who understand CVI, and the effects this parental stress then has on the *microsystem*, *mesosystem*, and *exosystem*.

Recommendations and Future Research

The existing body of literature about family experiences with CVI and its diagnosis in children is very limited. However, the research that existed prior to the initiation of this study demonstrated that parents were experiencing frustration and delays in diagnosis of CVI (Jackel et al., 2010). The results of both studies also indicated that many parents are not receiving helpful information at the time of diagnosis, and that many professionals do not seem to have enough knowledge about CVI to help families feel comfortable with the diagnosis and services. The current study is not representative of parent experiences nationwide nor worldwide. I was not able to reach families for this study, for example, if they had a child with unidentified CVI, including families that did not have the resources to actively pursue a diagnosis.

An interesting question is, how families with fewer resources are coping, and whether those children been identified as having CVI. Interview studies with families of children who have multiple disabilities including visual impairments, but no diagnosis of CVI, represent a gap in the research. However, that sample may be difficult to reach. Additionally, research that examines the records of the population of children in school with multiple disabilities and questionable visual skills, and whether or not those children have diagnosed CVI, is a possible way to access a wider

sample to unveil the population of children with potentially undiagnosed CVI. Further research with larger samples may have varying results, or may yield results similar to the study by Bernadette Jackel (Jackel et al., 2010). Another question for future research may be related to how to best educate professionals about CVI, to improve rates of diagnosis and knowledgeable interventions. Since CVI is often observed in children with specific, associated conditions, a diagnosis of any of those conditions should automatically trigger an assessment for CVI.

BONDING ISSUES WITH PARENTS

These parents found answers – but what of those that have not?

In discussion – address issue of how CVI is different than other disabilities

Implications for Practice. The participants in this study mentioned several helpful resources and processes. These resources should be shared with any family who has a child with suspected CVI, as recommended reading. The primary resource that was mentioned by the informants from this study as helpful was Christine Roman's book, *Cortical Visual Impairment: An Approach to Assessment and Intervention* (Roman-Lantzy, 2018). Additionally, *Little Bear Sees: How Children with Cortical Visual Impairment can Learn to See* was mentioned as a helpful resource, and it was written in parent-friendly language (Tallent, Tallent, & Bush, 2012). These books are helpful to parents who are looking for information about CVI and what they can do to increase positive outcomes for their children, in terms of vision.

In the most general terms, parents who wish to increase their child's visual access in the home environment may start by considering reducing visual and environmental clutter (complexity, including sensory input that may interfere with

visual processing). Using color(s) that their child seems to react positively toward, and using light to help guide their child to look at an item or in a particular direction may help.

Interventions for children with CVI are very individualized. Ideally, a child who has suspected CVI should be assessed by an educator who is well versed in CVI and the CVI Range; preferably, one who has the Perkins-Roman endorsement from Perkins School for the Blind and therefore has demonstrated skill in using the assessment tool. Specific, individualized interventions can then be designed and implemented based on the results of the assessment. Parents may find a multitude of intervention ideas based on a simple online search for CVI interventions, however they should be cautioned that every child with CVI is unique, and therefore, what works well for one child may not work well for their own child.

Practical implications. The primary finding of this study is that increased education of professionals – physicians and TVIs, specifically – is needed. This is evidenced by parents' expressed frustration in their attempts to locate practitioners who understand CVI, the length of time between parent concerns and diagnosis of CVI in their children, and their perceived need to educate themselves so that they are able to help and monitor the professionals. Although the data resulting from this study cannot be generalized to other parents of children with CVI, it provides a glance at the difficulties experienced by such parents. According to Dutton (2013), diagnosis of CVI does not need to be difficult. It is a diagnosis that involves recognizing some sort of damage to the brain, coupled with visual behaviors that are not explainable by the conditions of the eyes alone. Furthermore, early intervention

can improve outcomes for children with CVI (Huo et al., 1999), which could potentially increase independence and decrease their needs in terms of later intervention services. Therefore, it is widely beneficial to have children with CVI diagnosed so that they can receive intervention, as early in life as possible.

The combined concepts of sensitive periods, neuroplasticity, and Bioecological Systems Theory help emphasize the significance of partnerships between parents and professionals in child development. Neuroplasticity and sensitive periods affect the overall development of human beings, affecting all *systems* within Bioecological Systems Theory. Without a timely diagnosis to sufficiently explain a child's vision characteristics, as in CVI, it may impact all other systems, which are then subject to further impact due to missed sensitive periods and reduced neuroplasticity. Considering the *chronosystem* and regarding CVI diagnosis, the impact of delayed diagnosis over time impacts the child's ability to visually observe the environment and further delays conceptual development.

We, as professionals, have a responsibility to these children and to these families. The words of parents are telling, and parents are counting on us to identify CVI where it exists, and help them to help their children. In the words of one parent,

> [The doctors] just looked at me. And at the time in my head I was like "of course he can see." Because he can see some things. That's another reason this is so complicated, is like ... if you're a parent and you don't know that this exists and this is a thing ... you assume your kid either sees or he sees black.

Appendices

Appendix A

Summary of Studies Reviewed

Author(s)	**Topic/ Key Points as Related to this Study**
Addison (2003)	Child development, diagnosis delays Professionals do not see the delays in development as the parents see them
Baird, McConachie, & Scrutton (2000)	Child development, CP, disclosure of diagnosis, parents are sometimes dismissed or disregarded when they express concerns, only to find later that their suspicions were founded
Davies, Davis, & Sibert (2003)	Child development, jargon vs. parent-friendly language, when diagnoses are delayed, some parents report that the delay causes more stress and frustration than the diagnosis itself
Dutton et al. (1996)	CVI, prevalence earlier identification and intervention are key to reducing the impact of CVI on the child and family focus on prevalence, characteristics, and diagnosis of CVI
Halfon et al. (2004)	Child development, diagnosis delays, consequences delay in starting Early Intervention Services, resulting in loss of developmental potential
Hatton et al. (2007)	CVI, associated conditions leading cause of visual impairment in developed countries focus on prevalence, characteristics, and diagnosis of CVI
Huo et al. (1999)	CVI, Improvement is greater when diagnosed younger ability and potential, given the appropriate interventions, to improve their functional vision skills and use their vision more productively and functionally, due to neuroplasticity focus on prevalence, characteristics, and diagnosis of CVI
Jackel, Wilson, & Hartmann, (2010)	CVI, Difficulty getting services, lack of professional training, knowledge, and understanding
Khetpal & Donahue (2007)	CVI: ability and potential, given the appropriate interventions, to improve their functional vision skills and use their vision more productively and functionally, due to neuroplasticity focus on prevalence, characteristics, and diagnosis of CVI
Klein et al. (2011)	Child development, disconnect between parent and professional perceptions
Peters (2010)	Child development, diagnosis delays Timely diagnosis and intervention for children with disabilities, in general, has been identified as an important factor in reducing the impact of disabilities on development
Tallent, Tallent, & Bush (2012)	CVI, lack of professional knowledge. Book mentioned as helpful by a participant.

Appendix B

Advertisement for Participant Recruitment

Parents Wanted for Cortical Visual Impairment Research

Parents who have a child diagnosed with Cortical Visual Impairment (CVI) have an opportunity to participate in research aimed at improving knowledge about the state of education regarding CVI in the medical field.

The research is being conducted by a student researcher and doctoral candidate at the University of Maryland. She is also a teacher of students with special needs.

The research will explore the experiences of families in obtaining assistance for their children who have CVI. Information will be collected via interviews with families.

To learn more about this study, or to learn about participating in the interviews, please email Sara Kempler at _____ or call xxx-xxx-xxxx.

Thank you.

Appendix C

Advertisement for Participant Recruitment

Physicians and TVIs Wanted for Cortical Visual Impairment Research

Physicians and Teachers of the Visually Impaired (TVIs) who work with children diagnosed with or demonstrating characteristics of Cortical Visual Impairment (CVI) have an opportunity to participate in research aimed at improving knowledge about the state of education regarding CVI in the medical field.

The research is being conducted by a student researcher and doctoral candidate at the University of Maryland. She is also a teacher of students with special needs.

The research will explore the experiences of families in obtaining assistance for their children who have CVI. Information will be collected via interviews with families and professionals.

To learn more about this study, or to learn about participating in the interviews, please email Sara Kempler at _____ or call xxx-xxx-xxxx.

Thank you.

Appendix D

Letter of Introduction –

My Name is Sara Kempler. I am a doctoral student at the University of Maryland, and I also teach at a small school for children with special needs. I have had a great deal of training in the area of CVI Assessment and Intervention.

The population of students that I teach has a rather large percentage of students with undiagnosed vision difficulties, which lead me to question the experiences of families in trying to get medical and professional attention given to their children with similar vision issues, specifically CVI. The purpose of my study, a requirement for my doctoral program, is to describe the experiences of families of children who have been diagnosed with CVI.

The study will involve a set of interviews (2-3, at most) of at least 30-45 minutes each. Identifying information gathered about families will be kept strictly confidential, and the written results of the study will refer to each family by a case number, only.

Appendix E

Informed Letter of Consent –

Initials ____ Date ____

Informed Letter of Consent

Identification of Project
Parent Perspectives on Diagnosis of CVI in their Children: A Qualitative Analysis

Statement of Age of Subject
I state that I am over 18 years of age and wish to participate in research conducted by Sara K. Kempler, a doctoral candidate in the Department of Special Education at the University of Maryland, College Park, MD 20742. The study is being conducted under the supervision of Dr. Paula Beckman of the University of Maryland.

Purpose
The purpose of the research is to examine CVI and the impact this diagnosis has on families. I understand that the student investigator wants to learn about my perspective related to this issue so that more information is available to help professionals who work with children with atypical vision that is not related to an ocular visual impairment.

Procedures
The procedures involve at least two interviews with the student investigator, Sara K. Kempler. The purpose of the interviews is to learn about my experiences and perspectives related to the diagnosis of CVI in my child, as well as my opinions about the information and services my child has received for the condition.

The interviews will be conducted in-person, with follow-up options in-person or via telephone. The interviews will also be audiotaped for transcription, and will last approximately two hours, total.

Confidentiality
All information will be kept confidential to the extent permitted by law. The names of any family members or professionals who I mention during the study will not be identified at any time. The information I provide will be grouped with information that others provide for reporting and presentation. I understand that tapes and transcripts will be stored in a locked file

cabinet in the home of Sara K. Kempler, investigator, and destroyed at the completion of the study.

I further understand that all computer files will be password protected.

___ I agree to be audiotaped during my interviews for this study.

___ I do not agree to be audiotaped during my interviews.

Risks

There are no known risks to my participation.

Benefits, freedom to ask questions, and withdraw information

I understand that the information I provide is not designed to help me personally. The information that I provide will be used to improve understanding of the experience of families who have a child diagnosed with or with suspected CVI, so that professionals in education, health, and disability organizations can provide appropriate support and resources to these families. I understand that I can ask questions about the study and can decline to answer specific questions that are a part of the interview process. I am free to withdraw from participation at any time without penalty.

Sara K. Kempler, M.Ed.
<address>
<phone>

Printed Name of Participant_____

Signature of Participant_____

Date _____

Appendix F

Initial Interview Questions for Parents

Lead Question I: Tell me a little about your family.

Potential Prompts/Followup questions

a. How many children do you have?
b. Who is the oldest?

Lead Question II: Tell me about <CHILD's NAME>

Potential Prompts/Followup questions

a. How old was <child's name> when you first suspected something was different about his/her vision?
b. What steps did you take to get medical attention for his/her vision?
c. What obstacles did you face during the process of getting a medical diagnosis for his/her vision differences?
d. What did you find that was helpful during the process of getting a diagnosis?

Lead Question III: Thinking back to the time when you were seeking a diagnosis, tell me what the process of getting the diagnosis was like.

Potential Prompts/Followup questions

a. Was there something specific that made you feel that way?
b. How do you feel about it now?
c. What has happened to make you feel that way?

Lead Question IV: When <child's name> was diagnosed with CVI, what kinds of information did you receive?

Potential Prompts/Followup questions

a. What did you find helpful about the information?
b. What was not helpful?
c. Are there any suggestions you might offer to vision professionals (such as doctors and teachers of the visually impaired) that would assist other families in their journeys to get their children diagnosed?
d. Is there anything you would like to share with other parents who have children with suspected CVI?

Appendix G

Interview Questions for Professionals (TVIs)

Lead Question I: Have you ever assessed CVI in a child?

Potential Prompts/Followup questions:

a. If not, can you tell me if you see many children that demonstrate characteristics of CVI?

Lead Question II: Do you feel comfortable assessing CVI in children?

Potential Prompts/Followup questions:

a. Why or why not?

Lead Question III: Have you received any education about Cortical Visual Impairment?

Potential Prompts/Followup questions:

a. If so, what sort of information did you receive?
b. If not, have you read about it or learned in other ways?

Lead Question IV: What do you recommend to parents of children that demonstrate characteristics of CVI?

Potential Prompts/Followup questions:

a. What have you known or observed families to have to do to get a diagnosis of CVI for a child?
b. Do you feel there is enough information available to parents?
c. Can you think of anything that could make the process easier for families?

Appendix H

Interview Questions for Professionals (Physicians)

Lead Question I: Have you ever diagnosed or assessed CVI in a child?

Potential Prompts/Followup questions:

a. If not, can you tell me if you have any children in your practice that demonstrate characteristics of CVI?

Lead Question II: Do you feel comfortable diagnosing/assessing CVI in children?

Potential Prompts/Followup questions:

a. Why or why not?

Lead Question III: Have you received any education about Cortical Visual Impairment?

Potential Prompts/Followup questions:

a. If so, what sort of information was provided?
b. If not, have you read about it or learned in other ways?

Lead Question IV: What do you recommend to parents of children that demonstrate characteristics of CVI?

Potential Prompts/Followup questions:

a. What have you known or observed families to have to do to get a diagnosis of CVI for a child?
b. Do you feel there is enough information available to parents?
c. Can you think of anything that could make the process easier for families?

Appendix I

Results from Focus Group, July 2016

Participant ID	Sex of Child	Age of child when signs noted	Age of child at Diagnosis	Age of child when services began	Geographical Type (Urban or Rural)	Additional Info
I1a	M	5	7 years	12	U	GI Issues, Seizures
A1a	F	<1week	7 days	7 days	U	Born at West Penn, connected with C.R. early
C1a	M	Early	3.5 months	4	R	Diagnosed at 3.5 months but told to wait and see. Moved to another state, vision services discontinued
M1a	M	4 years	4.5 years	4.5 years		Adopted from Bulgaria; heavily medicated and living in crib for first 4 years of life. School did not know what to do, services hard fought
H1a	F	6 weeks	5 months	10 months	R	Pursued dx in part because mom is a NICU nurse
E1a	F	2.5 months	9 months		R	"If CVI, nothing you can do" "Have access to services but they don't know what to do"
M2b	M	4months	> 6 months		U	Parents told "God made a mistake" and "Nothing we can do". Says biggest problem is getting qualified TVIs.
L1a	M	~6 months	~6 months	~6 months	U	SBS at 6months, drugged for 3 months, but already had EI because was a twin and born early. Lucky to have found TVI trained by C.R. and live close to Perkins
J1a	M	7 months?	8 months?	14 months?	U	Raw emotion, story a bit confusing

R1a	F	“early”	“early”	No one knew what to do	U	“Delayed Visual Maturation, don’t buy trouble” “Nothing you can do, might resolve. Come back in a year”. Parent looked up CVI on her own and saw C.R. in a hotel rom.

Appendix J

Table 1 – Parent Participant Information

Table 1: Parent Participant Information

Parent(s)	**Child**	**Other Children (compared by age)**	**Age when *Adult Noticed* CVI Characteristics**	**Age @ *Diagnosis***	**Possible Associated Condition(s)**
Marla	Gwen	1 older 1 younger	2 years (Other Service Provider first noted)	~4 years	Hypoxia
Lisa	Bobby	1 older	<1 month	~2 years	Genetic
Tracey	Phoebe	1 younger 1 on the way	>7 months Teacher noted ~3 years	~3 years	Genetic
Veronica	Brady	4 living in the home, older	Immediately	3-4 years	Hypoxia
Lauren & George	Andrew	1 younger	Immediately	~2 years	Premature & Genetic
Raina & Charles	Harrison	1 younger	6 months; (Teacher noted at ~5)	5 ½ years	Genetic
Nadia & Boyd	Paul	1 on the way	<2 years	~3 years	Hypoxia & Genetic
Bethany	Jonathan	None	Immediately	1 week	Hypoxia
Claire	Dylan	1 older	~1 year	2 ½ years	Very premature
Jennifer	Gail	1 younger	<8 months	~8 months	2 weeks premature
Sharon	Mary	2 much older	1-2 years	2 ½ years	Genetic

Appendix K

Table 2: Themes and Subthemes

Themes	**Subthemes**
Something Was Wrong	4. Lack of Medical Knowledge and Understanding 5. Delayed Diagnosis 6. Seeking Clarity
From Diagnosis to Services	4. Information Received from Professionals 5. Lack of Medical Knowledge and Understanding 6. Changing Parenting Style and Learning New Skills
From Frustration to Hope	3. "It's a Struggle" 4. "These are things we can definitely do"
Advice for Others	3. Advice for Professionals 4. Advice for Parents

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