

# Walden University

## COLLEGE OF EDUCATION

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2008

# ABSTRACT

Assessing Maternal Response to the Impact of Hemangiomas on the Family System

by

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M.S., Russell Sage College, 1984

B.S., College of St. Rose, 1979

Dissertation Submitted in Partial Fulfillment  
of the Requirements for the Degree of  
Doctor of Philosophy  
Education

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## ABSTRACT

For new parents, having an infant can be stressful, but when that infant is diagnosed with a hemangioma, a potentially deforming and problematic tumor, the parents may experience additional stress. A hemangioma is the most common benign tumor of infancy but the least studied. This descriptive mixed methods study sought to add to the literature by exploring and describing self-reported stress levels in mothers of infants diagnosed with a hemangioma. The Hemangioma Reaction Questionnaire (HRQ), a new self-report instrument, and the Impact of Events Scale-Revised (IES-R), were used to collect data. One qualitative question was included in the HRQ. Family systems theory, the biopsychosocial disease theory, and stress-related concepts formed the framework for this study. The convenience sample included 148 mothers recruited from hemangioma informational Web sites. Descriptive analysis was performed and the results were triangulated between the quantitative and qualitative data. Three themes, all present in acute stress, were anchored: intrusion, avoidance, and hyperarousal. Hyperarousal symptoms were present in 48.1% in the EIS-R, 53.0% in the HRQ, and 48% in the qualitative question. Intrusion symptoms were present in 55.5% in the EIS-R, 20.2% in the HRQ, and 46% in the qualitative question. Lastly, avoidance was present in 38.8% in the EIS-R, 29.0% in the HRQ, and 6% in the qualitative question. Future research using a control group and including both parents would provide a stronger assessment. From a social change perspective, this study provides a tool for physicians to use to assess parental stress for potential counseling needs, and offers support for families seeking to establish a medical necessity for treatment.

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## DEDICATION

This work is dedicated to all of the families who have had an infant diagnosed with a hemangioma and have experienced the frustrations, confusion, and extreme stress that is associated with this diagnosis. I pray that this study will help to provide credibility and relief to their effort to get an accurate diagnosis, appropriate treatment, and proper insurance coverage.

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## CHAPTER 1:

### INTRODUCTION TO THE STUDY

On February 7, 1994, my daughter, Christine Mary Shannon entered this world. She was tiny, but she was perfect. When she was 6 weeks old, a small red lesion appeared on her lower lip. Her pediatrician said it was a hemangioma, which according to Batniji, Buckingham, and Williams (2005), is “the most common tumor of infancy and childhood affecting approximately 10% of infants by the age of 1 year” (p. 301). The pediatrician also said that a hemangioma will typically grow for up to 1 year, they are common on the face, and it is unknown as to how large one will become before it stops growing. This prognosis was shocking news for her parents. Shakin-Kunkel, Zager, Hausman, and Rabinowitz (1994) reported that “parents are often shocked by the unexpected hemangioma and its subsequent growth” (p. 524).

The doctor informed Christine’s parents that there was little information in the medical field about these tumors because the topic is scarcely taught in medical school. As a result, treatment information is inconsistent. Additionally, because some hemangiomas resolve over a period of 10 years, insurance companies do not believe treatment is a medical necessity. From the day Christine’s hemangioma was diagnosed, and throughout the following year, this crisis caused acute stress for me. Immediately following the diagnosis, I frequently felt numb and detached because I had no control over the growth of the hemangioma. I often avoided people because of disapproving stares and rude comments. I was unable to sleep soundly, often dreaming of the

hemangioma. At no time was mental health counseling services offered during any of the visits to Christine's doctors.

When Christine was 18 months old, the hemangioma was surgically removed. Some stress was alleviated after the surgery, but the removal was not the end of the stress. The insurance company refused to pay for treatment, stating that treatment was not a medical necessity. The denial was appealed three times before it was overturned. Denial of payment by insurance companies is prevalent today with parents of infants diagnosed with a hemangioma. According to Rozell-Shannon, Marshall, and Waner (1997)

You have health insurance; you expect your benefits to cover the cost of treatment for a vascular lesion. Your insurance or HMO denies coverage. It is not an unusual situation. Many insurance companies still classify the treatment of vascular lesions as cosmetic surgery. (p. 139)

Since little attention was given to study the impact on the family system from having an infant diagnosed with a hemangioma, the National Institute of Health (NIH) convened a special hemangioma conference in 2005 in order to address concerns such as the psychosocial impact on the family. Participants included leading medical specialists and support group leaders concerned with hemangiomas. At this international gathering, Chamlin (2005) stated that "objective clinical outcome measures and subjective quality-of-life (QOL) instruments are lacking for infantile hemangiomas. Such measures are needed to accurately quantify the burden of this disease on children and their families" (p. 396). Thirty years prior to the NIH conference, Drotar, Baskiewicz, Irvin, Kennell, and Klaus (1975) found that "despite the relatively large number of infants with congenital anomalies, our understanding of how parents develop an attachment to a malformed child remains incomplete" (p. 710). In addition, the authors explained,

“Although previous investigations are in agreement that the child’s birth often precipitates major family stress, there have been relatively few descriptions of the process of family adaptation” (p. 710). After 30 years, little progress has been made to quantify the burden of hemangiomas on the family system.

More recently, Weinstein and Chamlin (2005) stated that “compared to other equally or less common congenital defects such as a cleft lip/palate, there is a paucity of data on the quality of life in families and children with vascular anomalies” (p. 256). They also said, “The birth of a child with a malformation has been likened by many authors to a crisis situation” (p. 257).

Is it a stressful event when the parents of an infant are told that their child has a hemangioma, a tumor that could become very large, disfiguring, and potentially problematic? The Impact of Event Scale (IES) could possibly answer that question. The IES, developed by Horowitz, Wilner, and Alvarez (1979), measures current subjective distress related to a specific event. In addition, according to Tak and McCubbin (2002), “Many of the existing studies of family stress and coping hold that the effects of life events and strains contribute to psychological distress” (p. 191). In order to verify whether or not the diagnosis of an infant with a hemangioma is perceived as a stressful event, the IES-R was used to answer this question. If the diagnosis of an infant with a hemangioma is perceived to be stressful, then a process can be proposed to ensure that all parents of children diagnosed with a hemangioma are properly assessed and receive the appropriate mental health referral, if necessary. In addition, the establishment of the clinical diagnosis of Acute Stress Disorder (ASD) or Posttraumatic Stress Disorder

(PTSD) may provide the medical necessity needed by the parents in order to receive insurance approval for treatment of the hemangioma.

### Background

Hemangiomas are the most common tumor of infancy, with most occurring in the head and neck region (60%) and some patients (20%) having more than one hemangioma (Williams et al., 2003, p. 229). These same authors stated that “given these facts and that hemangiomas may be unsightly birthmarks; the psychological stress on the developing child and family cannot be underestimated” (p. 229).

Typically, the family of an infant with a hemangioma will first see a primary care physician who will diagnose the lesion. Because there is little consistent information on the diagnosis and treatment of hemangiomas, the prognosis and treatment options presented to the families vary from physician to physician. Some physicians inform the parents that the hemangioma will eventually involute (regress) so they should just leave it alone. This philosophy is antiquated and damaging to the families, but it persists because the topic is scarcely taught in medical school and infrequently addressed at medical conferences. According to Waner and Suen (1999)

An issue often inadequately dealt with, but an extremely important one nonetheless, concerns the true meaning of the term involution. Unfortunately, a large proportion of parents have been led to believe that the hemangioma will disappear completely and leave no trace. Perhaps the current dogma concerning non-intervention, coupled with the lack of substantial experience on the part of physicians, is partly to blame. (p. 26)

Waner and Suen (1999) further declared, “Only 40% of hemangiomas involute with an acceptable result. Therefore, 60% of all patients with hemangiomas will require

some form of corrective surgery” (p. 27). Williams et al. (2003) also claimed “If these often disfiguring vascular lesions do not involute early, they may have profound psychosocial effects on the child and family” (p. 229).

Tanner, Dechert, and Frieden (1998) pointed out that most hemangiomas develop during the first few weeks of life, often starting as a tiny red dot, grow for up to 1 year, and then begin a long and slow involution cycle. The authors explained that it is uncommon for involution to be completed by 2 or 3 years of age. More likely, only 50% of hemangiomas have completely involuted by 5 years of age, 75% by 7 years of age, and 90% by 9 years of age (p. 446). It is because of this misunderstood perception that hemangiomas will involute, or resolve on their own, that many physicians are reluctant to treat and it is also the reason that many insurance companies do not approve treatment. Based on the outdated information that has been provided to the insurance companies, they operate under the false premise that all hemangiomas regress or involute on their own.

While cleft lip and cleft palate are different from hemangiomas, both are birth defects with an aesthetic aspect. According to Singer and Bergthold (2001)

Cleft lip and cleft palate are common birth defects with both functional and cosmetic dimensions. Treatment often falls into the gray area between reconstructive and cosmetic surgery. As a result, medical necessity determinations involving this condition have proven to be especially problematic. (p. 2162)

Having an infant diagnosed with a hemangioma can be a traumatic and stressful event for most parents. According to an informational brochure produced by the U.S. Department of Health and Human Services in 2003, “a traumatic event is a single event or a series of events that causes moderate to severe stress reactions and is characterized

by a sense of horror, helplessness, serious injury, or the threat of serious injury or death” (U.S. Department of Health and Human Services, 2003, p. 1). A study conducted by Drotar et al. (1975) indicated that shock and a sense of helplessness were experienced by most of the parents interviewed regarding their reaction to the birth of a child with a congenital malformation (p. 710). Nearly 10 years later, a study conducted by Fortier and Wanlass (1984) found that following the diagnosis of a child with a disability, the family members began “working through feelings of grief, anger, guilt, helplessness, and isolation” (p. 13).

Families must deal with the lack of consistent information and uncertainty over whether they will find an adequately trained physician. They struggle to get accurate information, and once they find the right physician, they face the burden of proving to their insurance company that treatment is a medical necessity. According to Glassman, Model, Kahan, Jacobson, and Peabody (1997)

Policymakers, insurers, and health care organizations invoke “medical necessity” to define which services should be offered to patients. However, this term has many meanings and many uses, ranging from general definitions used in health insurance contracts, to specific definitions used to identify which services are “crucial” for individual patients or patient populations. Consequently, “necessity” has an ambiguous meaning; not surprisingly, the manner in which the concept of necessity is applied varies significantly within and across providers, insurers, and states. (p. 152)

As a result, if a medical necessity is not established for the treatment of the hemangioma, the treatment will not be approved and the families will be forced to appeal, live with the deformity, or provide payment for the treatment. According to Williams et al. (2003) in their study to assess the psychological impact of hemangiomas and their treatment on children and their families “This study underscores the importance of evaluating the



psychological role that hemangiomas may have on the entire family unit and the fact that treatment should be tailored to curtail the damaging effects” (p. 233).

The diagnosis of an infant with a hemangioma can result in a myriad of stressful events for the affected family. Brogdon (1999) stated “Facial disfiguration can cause significant psychopathological responses and behaviors in a patient and the patient’s family. Facial disfigurement resulting from any physiological event or pathology is always tragic and produces tragic psychosocial outcomes” (p. 217). Yet, there are no guidelines or disease-specific instruments for assessing the impact of this crisis on the family system. Additionally, there has been no study to establish the diagnosis of an infant with a hemangioma as a stressful or traumatic event. According to Weinstein and Chamlin (2005)

The most important outcome for patients with vascular anomalies and their families is quality of life. Measurement of health-related quality of life is based on the assessment of the individual’s perception of the impact of medical and nonmedical issues regarding physical, mental, and social functioning. Few valid tools exist for this purpose, especially for children with disfigurement and their families. Thus it is important for physicians seeing children with vascular anomalies to anticipate and monitor for signs of psychosocial impairment in both affected children and their parents, and to provide resources to those who might benefit from psychological intervention. (p. 258)

While there is a paucity of studies concerning the impact of hemangiomas on the family system, there have been studies concerning children with other medical conditions. These reports indicate that the diagnosis of a medical condition in an infant or child is a stressful and traumatic event for the entire family system. According to St. John, Pai, Belfer, and Mulliken (2003), “A child with chronic illness, such as a craniofacial deformity, can be an overwhelming burden that destabilizes the whole family

system” (p. 704). They further noted “Significant stress also develops due to the complex long-term nature of habilitating a child with a craniofacial anomaly. The need for multiple hospitalizations and major surgical procedures places a tremendous demand on the ability of these families to cope” (p. 705).

Systems theory teaches us that a family is a type of system that is made up of many parts or units that together create a larger unit referred to as the family. Just as all the parts of a car are important for the car to run efficiently, all the parts of the family are important for the family to be healthy. When one unit, or family member, is affected by sickness or stress, the entire unit is also affected. According to Kazak (1989), “From a systems perspective, a problem in any member of a family has an effect on all other members, and changes in any member of the system affect all others” (p. 26). Jedlicka-Kohler, Gotz, and Eichler (1996) also noted that

The diagnosis of cystic fibrosis in children represents a severe blow to parents, confronting them with traumatic information. A few studies have emphasized high levels of emotional stress during information giving, but none has examined potential cognitive impairment in the parents caused by the trauma of learning the diagnosis. (p. 204)

Additionally, Canam (1993) stated, “The diagnosis of a chronic illness or disability in a child represents a major stressful life event for all family members, yet the diagnosis is but one point in the family’s ongoing experience with the ill child” (p. 46). A recent study examined the prevalence of acute stress disorder in parents of infants hospitalized in the neonatal intensive care unit (Shaw et al., 2006). According to the authors, “researchers have identified posttraumatic stress disorder (PTSD) as a model to describe and explain the psychological reaction of parents to their NICU experience” (p.

206). This prospective longitudinal study was conducted to investigate the posttraumatic stress response of mothers after the birth of a very low birth weight infant (Kersting et al., 2004). The authors concluded “The situation of a mother who has given birth to a very low birth weight infant is a complex, long-term traumatic event that necessitates ongoing emotional support extending beyond the period immediately after the birth” (p. 473).

These are just a few of the studies that support the hypothesis that having an infant diagnosed with a hemangioma is a stressful event for the entire family system.

In an attempt to develop a disease-specific instrument to measure the impact of hemangiomas on the family system, and in order to determine if there is a relationship between the diagnosis of a hemangioma in an infant and the level of stress of the family, an existing instrument, the Stanford Acute Stress Reaction Questionnaire (SASRQ), was modified to the Hemangioma Reaction Questionnaire (HRQ). The SASRQ was originally designed to measure dissociative and anxiety symptoms following a traumatic experience (Cardena, Koopman, Classen, Waelde, & Spiegel, 2000).

Using the 30-item self-report SASRQ as a template, the HRQ was created based on the theory that the diagnosis of an infant with a hemangioma is a stressful or traumatic event that may lead to acute stress and posttraumatic stress symptoms. The SASRQ has been used to measure such traumatic events as earthquakes, motor vehicle accidents, and witnessing a shooting, but has not been used to measure stress as a result of having an infant diagnosed with a hemangioma. If the diagnosis of an infant with a hemangioma is perceived by the parents as a traumatic event, then the HRQ could become the standard for assessing whether the family needs mental health counseling. In addition, because

acute stress disorder or posttraumatic stress disorder symptoms could lead to a clinical diagnosis, this could also provide support for the parents who have had treatment disapproved by their insurance company. Weinstein and Chamlin (2005) reported that a questionnaire distributed to 259 families of patients with a vascular birthmark showed that the majority felt that their life would change dramatically if the birthmark could be eliminated.

### Problem Statement

In a 1975 study (Drotar, Baskiewicz, Irvin, Kennell, & Klaus) and 30 years later at an NIH Conference (Chamlin (2005) it was stated that even though hemangiomas are the most common tumor of infancy, little research has been done on the impact of these lesions on the family system. The current study addressed this gap. In addition, the lack of current educational materials for physicians and insurance companies has confounded this problem, as both populations believe that most hemangiomas will resolve in time to a cosmetically acceptable appearance without intervention (Waner & Suen, 1999). Research over the past 2 decades supports evidence that 60% of all hemangiomas that are not treated will leave a deformity (Waner & Suen). Families receive conflicting information and are forced, because of the denial of treatment, to either live with the deformity for many years, or to provide the funds needed to pay for the treatment.

This descriptive study contributes to the body of knowledge concerning the impact of hemangiomas on the family system. The HRQ, designed to measure the burden of hemangiomas on the family system, was used in this study to assess whether or not the diagnosis of a hemangioma is a stressful or traumatic event that results in acute stress or

posttraumatic stress disorder symptoms. In addition, an existing instrument, the Impact of Event Scale - Revised (IES-R) was used to determine if the diagnosis of a hemangioma is a traumatic event. Together, these two instruments were used to explore and describe whether or not the impact of having an infant diagnosed with a hemangioma is perceived as a traumatic event.

While there is no treatment that would be considered an independent variable in this study, the level of stress of the mothers completing the surveys is an outcome of the diagnosis of the infant with a hemangioma, or a type of dependent variable, as the mother's level of stress may be presumed to be a direct result of the diagnosis. According to Johnson and Christensen (2004), "The primary purpose of descriptive research is to provide an accurate description or picture of the status or characteristics of a situation or phenomenon. The focus is not on how to ferret out cause-and-effect relationships but rather on describing the variables that exist in a given situation" (p. 347).

#### Nature of the Study

This mixed methods descriptive study is what Campbell and Stanley (1963) referred to as the one-shot case study, in which "a single group is studied once, subsequent to some agent or treatment presumed to cause change" (p. 6). The agent or treatment presumed to cause a change in the stress levels of the mothers is the diagnosis of the infant with a hemangioma. This study utilized two Likert scale survey instruments. The HRQ, a modification of the SASRQ, consists of 30 questions in which respondents select the answer that best describes their personal experience following the diagnosis of their infant's hemangioma (1 = *not experienced*, 2 = *very rarely experienced*, 3 = *rarely*

*experienced*, 4 = *sometimes experienced*, 5 = *often experienced*, and 6 = *very often experienced*). The second instrument is the IES-R, which consists of 22 questions in which respondents select the answer that best describes how distressing it has been during the week following the diagnosis of the hemangioma (1 = *not at all*, 2 = *a little bit*, 3 = *moderately*, 4 = *quite a bit*, and 5 = *extremely*). This descriptive study explores and describes the level of stress in mothers of infants diagnosed with a hemangioma. Additionally, there is one question in the HRQ that requires respondents to describe the most profound event that has occurred since their baby was diagnosed with a hemangioma. This question required a qualitative analysis that explored the phenomenon of the impact of having a baby diagnosed with a hemangioma for an increased understanding of the problem. Similar phrases were coded and analyzed in order to generate common themes (Creswell, 2003).

The hypothesis for this descriptive study explored if having an infant diagnosed with a hemangioma was perceived as a stressful or traumatic event that results in acute stress or posttraumatic stress disorder symptoms. Through the creation of the HRQ, and in conjunction with the IES-R, this study described the impact on the mothers completing the surveys. The HRQ, therefore, may be used in the future to explore the impact of hemangiomas on the stress level of the mother or father of the infant diagnosed with a hemangioma. This is significant, because the HRQ can become a tool for physicians to use to refer the the mother or father for mental health counseling, if needed. In addition, if acute stress symptoms are frequently described, this may give the families the leverage needed to appeal the denial of treatment by their insurance company. Lastly, the HRQ

closes a 30-year gap in medical literature that indicated that no instrument exists to measure the burden of hemangiomas on the family system.

The HRQ was pilot tested with 13 mothers of infants who were diagnosed with a hemangioma 1 to 4 years prior to completing the instrument. According to the test-retest analysis of the HRQ instrument, the Pearson  $r$  of each of the factors indicated a range of .70 for depersonalization to .97 for a subjective sense of numbing, detachment, or absence of emotional responsiveness (Rozell-Shannon, 2007). The HRQ measures nine different factors, which include five for dissociative symptoms and one for each of the following symptoms: traumatic event, marked avoidance, anxiety, and impairment in functioning. The Pearson  $r$  measures the strength of the linear relationship between the paired quantitative values in the test-retest study (Triola, 2005). The results of the pilot study indicate that the HRQ is not only a reliable instrument, it also indicates a fairly high correlation to traumatic symptoms, such as .97 for dissociative symptoms, for the mothers completing the survey for the pilot study.

Table 1 depicts the Pearson  $r$  correlation coefficient for the nine measures in the HRQ test-retest pilot study.

Table 1

*HRQ Test Retest Pilot Study Results*

Measure	Pearson $r$ for test-retest reliability
Dissociative symptom (1): Subjective sense of numbing, detachment, or absence of emotional responsiveness	.970
Dissociative symptom (2): A reduction in awareness of one's surroundings	.931
Dissociative symptom (3): Derealization	.701
Dissociative symptom (4): Depersonalization	.705
Dissociative symptom (5): Amnesia	.710
Traumatic event is persistently reexperienced	.943
Marked avoidance of stimuli that arouse recollection of the trauma	.939
Marked symptoms of anxiety or increased arousal	.801
Impairment in functioning	.859

The proposed convenience sample for this study was 169 mothers of infants who were diagnosed with a hemangioma within 1 year of completing the surveys. This number was based on the population size of 300 new mothers of infants diagnosed with a hemangioma who join the various support groups each year. According to Johnson and Christensen (2004) a population ( $N$ ) of 300 has a recommended sample size ( $n$ ) of 169 (p. 218). The criteria for participants included first-time mothers who were between 18 and 40 years of age, who had a high school education or higher, and who were never treated for stress prior to the diagnosis of the hemangioma



### Research Questions

The following research questions and hypotheses were used in this research study:

1. Using the HRQ instrument, what level of stress is reported by mother's who have an infant who is diagnosed with a hemangioma and who complete this survey?

*H<sub>01</sub>*: There is no reported stress level for mothers who have an infant diagnosed with a hemangioma and who completed this survey.

*H<sub>A1</sub>*: There is a reported level of stress for mothers who have an infant diagnosed with a hemangioma and who completed this survey.

2. Using the IES-R, what impact of event measure is reported by mothers of infants diagnosed with a hemangioma and who complete this survey?

*H<sub>02</sub>*: There is no reported measure from the diagnosis of an infant with a hemangioma based on the responses from the mothers who complete this survey.

*H<sub>A2</sub>*: There is a reported measure from having an infant diagnosed with a hemangioma based on the responses from the mothers who complete this survey.

3. For the qualitative question included in the HRQ, what common themes have emerged from the question that asks the mothers to describe the most profound event that has occurred since the hemangioma was diagnosed?

*H<sub>03</sub>*: There are no common themes among the mothers who completed the qualitative question in the HRQ.

*H<sub>A3</sub>*: There are common themes among the mothers who completed the qualitative question in the HRQ. .

### Purpose of the Study

The purpose of this descriptive survey study is to describe and explore the reported levels of stress of mothers of infants diagnosed with a hemangioma to determine if these mothers experienced acute stress disorder or posttraumatic stress disorder symptoms as a result of the diagnosis, and to determine if the diagnosis of an infant with a hemangioma was perceived as a stressful or traumatic event. The hypothesis being tested is that the diagnosis of an infant with a hemangioma is stressful and is perceived as a stressful or traumatic event for the family, particularly the mother of the infant. The qualitative question included in the HRQ explored common themes among the mothers. The rationale for using a mixed study was to use the one qualitative question results to substantiate the findings from the quantitative data results.

This study is important because it closes a 30-year gap in the literature, uses the first disease-specific instrument (HRQ) to examine the levels of stress of mothers of infants diagnosed with a hemangioma, and provides a process for physicians to assess the mental health needs of the affected family. Additionally, the results of this study may be used by parents to support the medical necessity designation required by insurance companies to approve treatment of the hemangioma. Without a medical necessity designation, families will continue to watch their child live with a deformity for many years, or will be faced with the task of providing the necessary funds to pay for treatment. This is the first study that uses a disease-specific instrument to assess the stress levels of

mothers of infants diagnosed with a hemangioma. This instrument may also be modified to assess the stress levels of parents of infants with other specific vascular birthmarks or tumors such as port wine stains, venous malformations, lymphatic malformations or arterio-venous malformations.

### Theoretical Base

The theoretical basis for this study focused on weaving together family systems theory and the biopsychosocial disease theory. Family systems theory and biopsychosocial disease theory have a foundation in general systems theory in that each component that is explored, whether it is the family (family systems theory), or the medical condition, (biopsychosocial disease theory) finds its roots in a larger, more hierarchical system (general systems theory).

#### *Family Systems Theory*

According to Bertalanffy (1968), just about everything can be viewed as a system: medicine, machines, politics, chemicals, air, and even families (p. 4). Forty years ago Bertalanffy stated that:

The concept of systems pervaded all fields of science and penetrated into popular thinking, jargon, and mass media. Systems' thinking plays a dominant role in a wide range of fields from industrial enterprise and armaments to esoteric topics of pure science. Innumerable publications, conferences, symposia and courses are devoted to it. Professions and jobs have appeared in recent years which, unknown a short while ago, go under names such as systems design, systems analysis, systems engineering and others. (p. 3)

Families are a system and when one member, such as an infant, is diagnosed with a medical condition, the entire family system is impacted based on family systems theory. According to Kazak (1989), "From a systems perspective, a problem in any member of a

family has an effect on all other members, and changes in any member of the system affect all others” (p. 26). Additionally, according to Fleming (2007), “Family systems theory has had a significant impact on the study of families and on approaches to working with families. It has guided research into such areas as understanding traumatic events or chronic health issues and their impact on individuals and families” (p. 1). Studies have been done concerning how family systems are impacted by illness of one member, or more specifically by having a child born with a disease or disability, and additional studies address the impact on the family system following a traumatic event, but no such studies have focused on the diagnosis of an infant with a hemangioma. According to Kazak, “The chronic illness in a child is not contained within the child. Rather, the illness has ramifications for all members of the family system. Because one member of the family must make a change, others will be affected” (p. 26). Kazak claimed that there has been a significant amount of writing by family therapists and sociologists regarding family reactions to stressful life events. According to Hill (1966). Kazak stated:

Seminal work on family reactions to stressful life events was conducted in 1966 in the ABCX model. Within this model, a stressor event (A) interacts with family resources (B). The family members provide an interpretation (C) of this event, resulting in an outcome (X). This mode is useful for visualizing adjustment to illness or stressful life events over a period of time. (p. 27)

### *Biopsychosocial Disease Theory*

The biopsychosocial disease theory, on the other hand, offers a look at the biological, psychological, and social components of disease, rather than just looking at medically treating the condition, or more specifically, the hemangioma. This theory has its roots in general systems theory. Rather than looking at disease as merely physical

signs and symptoms, the biopsychosocial disease theory looks at disease as a system comprised of the biological, psychological, and social components. Engel (1962) is responsible for the design of the biopsychosocial theory of disease. This theory evolved in the 1960s out of Engel's personal experience as an internist, psychiatrist, and psychoanalyst and his efforts to formulate a comprehensive understanding concerning human behavior and disease. Engel said, "I have advanced such theoretical frameworks, and in many instances, I am acutely aware of deficiencies" (p. viii). He claimed that this comprehensive view of disease had its deficiencies but it was a step in the right direction toward a systems approach to treating patients. Engel was aware that most physicians had conceptualized human beings in terms of "biologically determined developmental destiny, adapted to and adjusting in a physical, interpersonal, and social environment" (p. 239). However, through the use of the biopsychosocial model, Engel began to introduce many physicians to the "consideration of human beings as a biological, psychological, and social organism. These components encompass the deviations referred to as disease, the proper subject of medicine" (p. 239). It is within this context that the family is examined as a system and therefore any disease that affects one member of the family affects various components of the family functioning, such as the psychological and social components.

Engel (1962) established his theoretical framework for the biopsychosocial model by first defining disease as the "failure or disturbance in the growth, development, functions and adjustments of the organism as a whole or of any of its systems" (p. 240). This broad definition, according to Engel, was an attempt to "get away from the

substantive assumption that disease is a thing in itself, unrelated to the patient, the patient's personality, bodily constitution or mode of life" (p. 240). Engel explained that this definition does not restrict attention to any single system of the body, but rather it permits conceptualization of various breakdowns or failures at all levels, including biochemical, cellular, organic, psychological, interpersonal or social. Most of all, according to Engel, it hopes to permit consideration of the interrelationships of these various levels of organization in the human body. While the diagnosis of a hemangioma is based on clinical findings, there is also a psychological and social component of this often disfiguring tumor.

Most physicians who diagnose a hemangioma neglect to look beyond the biological component of the condition. While families may present with obvious symptoms of distress (psychological) and may be concerned with rude public comments and stares (social) only the hemangioma is addressed. If physicians were to look at the condition in a comprehensive and systems perspective, all components of the disease could be explored and appropriately addressed. In addition, by viewing the impact of the hemangioma on the entire family system, an assessment can be made to insure that the family unit is functional or whether it is in need of mental health counseling services.

The two theories, the family systems theory and the biopsychosocial disease theory, weave together in this study to provide an understanding of how family systems, specifically mothers, are affected by the birth of an infant with a medical condition, such as a hemangioma, and also analyzes the use of the biopsychosocial model theory for understanding how a medical condition, such as a hemangioma, should be treated.

## Definitions

*Acute stress disorder (ASD)*: “Severe dissociative, reexperiencing, avoidant, and hyperarousal symptoms in the immediate aftermath of a traumatic event” (Classen, n.d., p. 401).

*Biopsychosocial model or disease theory*: The biopsychosocial model is the foundation for the biopsychosocial disease theory that basically views disease as comprising the physical (biological), psychological, and social aspects of the disease as opposed to traditionally (Engel, 1962).

*Family systems theory*: Theory based on the study of families and how they work (Fleming, 2007).

*General systems theory*: Theory based on the study of all things being viewed as comprised of component that make up a system or in a systematic view (Bertalanffy, 1968).

*Hemangioma*: Benign vascular tumor of infants (Williams et al., 2003). Also referred to as a vascular anomaly, vascular birthmark, vascular lesion, or congenital anomaly.

*Hemangioma Reaction Questionnaire (HRQ)*: Self-report instrument designed to measure the impact of having an infant diagnosed with a hemangioma. (Rozell-Shannon, 2007).

*Impact of Event Scale – Revised (IES-R)*: A self-report measure designed to assess current subjective distress for any specific life event (Weiss & Marmar, 1997).

*Involution:* Refers to the regression or shrinking of a hemangioma over a period of time ranging from 1 to 10 years (Waner & Suen, 1999).

*Medical Necessity:* “A legal term used in most commercial health plan contracts to establish which interventions a health plan will cover for its members” (Singer & Bergthold, 2001, p. 2162).

*Posttraumatic stress disorder (PTSD):* An “intense sensation of fear, helplessness, or horror in response to a traumatic event. The witness to the event must have at least one reexperiencing symptom, three symptoms indicating persistent avoidance of reminders of the event, and two arousal symptoms.” (Classen, n.d., p. 402).

*Traumatic event:* An event, or series of events, that causes moderate to severe stress reactions and is characterized by a sense of horror, helplessness, serious injury, or the threat of serious injury or death. (U.S. Department of Health and Human Services, 2003).

*Vascular anomaly:* See definition for Hemangioma. There are, however, other vascular anomalies such as port wine stains, venous malformations, lymphatic malformations, and arterio-venous malformations, in addition to hemangiomas.

#### Assumptions

1. It is assumed that the infants of the mothers who completed the surveys were diagnosed with a hemangioma.
2. It is assumed that the mothers completing the surveys understood the concept of stress, trauma, and posttraumatic stress disorder.



3. It is assumed that the reliability test results from the pilot study for the HRQ could be applied to this study.
4. It is assumed that the mothers completing the surveys had no other stress-related illnesses or conditions.
5. It is assumed that the HRQ is an accurate measure of stress from having an infant diagnosed with a hemangioma.
6. It is assumed that the IES-R accurately reflects the measure of a traumatic event.

#### Limitations

1. The study was limited to mothers only. According to Pelchat et al, 1999, “The greater impact of a child’s disability is on mothers” (p. 378). Also, Dellve, Samuelsson, Tallborn, Fasth, and Hallberg (2005) claimed “mothers of children with a rare disability showed more parental stress than fathers” (p. 396). Only using mothers and not fathers was a limitation.
2. The study did not identify other potential confounding variables that could impact the stress levels, such as marital discord, financial problems, death of a loved one, or loss of a job.
3. The participants were not required to be formally diagnosed by a clinician as having either ASD or PTSD.
4. The participants were volunteers who knew that the study was being done to measure stress. This might reveal a bias that would not have been revealed in a study that did not describe the objectives.

5. Participants were solicited from hemangioma -related Web sites. Though this may be perceived as a strict limitation, most families who have an infant diagnosed with a hemangioma go to the various informational Web sites initially in order to locate a treatment specialist because the Internet is the primary source of these specialty listings. Also, many treating physicians routinely refer mothers of infants with a hemangioma to these Web sites for support and informational resources.

#### Scope of the Study

The scope of this study included mothers of an infant diagnosed with a hemangioma during the past year. In most cases, the infant was the firstborn. The participants were between 17 and 41 years of age.

#### Significance of the Study

A research study that describes and explores the relationship between having an infant diagnosed with a hemangioma and the level of stress of the mothers is significant because no such approach, prior to this study, had been taken to measure the burden of this disease on the family system. Additionally, the first disease-specific instrument, the HRQ, was created from an existing instrument, the SASRQ, for the purpose of this study. The IES-R, an existing psychometric instrument, was also used for this study to establish whether or not the participants perceived the diagnosis of an infant with a hemangioma as a stressful or traumatic event. Both instruments used in this study may also provide a process for physicians to use to refer the families for mental health counseling, if needed. This study is also noteworthy because the diagnosis of either ASD or PTSD may provide

support for families who must appeal the insurance companies in order to receive approval for the treatment of their infant's hemangioma, since treatment is often denied on the grounds that it is not medically necessary. According to the U. S. Department of Health and Human Services (2003), insurance companies routinely approve the treatment of stress-related conditions; therefore, establishing that the hemangioma has caused stress-related symptoms in the family may provide justification for treatment.

The creation of the HRE also closes a 30-year gap concerning the lack of an instrument to measure the burden of hemangiomas on the family system. This instrument and this study contribute to the literature on this subject by expanding the understanding of how families are psychologically impacted by having an infant diagnosed with a hemangioma.

### Introduction Summary

In chapter 1, the introduction identified a gap in literature and research concerning the impact of hemangiomas on the family system. The background and nature of this descriptive mixed methods study provided justification for the study and the conceptual framework identified how family systems theory and the biopsychosocial disease theory weave together to provide a theoretical foundation. The problem statement noted that though hemangiomas are the most common tumor of infancy, they are the least studied. It also noted that no disease-specific instrument has been designed to measure the impact of hemangiomas on the family system. The HRQ was identified to address this gap. It is a new, disease-specific instrument that is a modification of the SASRQ, and was created and tested in a pilot study. The IES-R was also identified for this study to be used in

combination with the HRQ. The IES-R is an existing instrument that measures whether or not an event is perceived as traumatic. The two instruments were combined to measure the level of stress of mothers of infants diagnosed with a hemangioma and to determine if they perceived the diagnosis as a stressful or traumatic event. The results of this study may assist parents who have to appeal their insurance company's decision because treatment of the hemangioma has been denied as not medically necessary. It was also referenced in chapter 1 that there is no process for assessing and referring affected families for mental health counseling and that the HRQ could be used to fill this gap. Chapter 1, therefore, established the foundation for the hypothesis that having an infant diagnosed with a hemangioma can cause stress in the mothers of these infants, and the diagnosis may be perceived as a stressful or traumatic event. Definitions of terms, assumptions and limitations of the study were also defined in Chapter 1. The scope established that only mothers of infants diagnosed with a hemangioma would be included in this study. The significance of this study proposed that a gap would be closed in literature, a new instrument would be available for physicians to use for assessing the impact of having an infant diagnosed with a hemangioma on the family system, and a process would be available for these families to use this study as justification to appeal the denial of treatment of the infant with the hemangioma by the insurance companies.

. Chapter 2 reviews related research and literature regarding the impact to the family system from having an infant diagnosed with a medical condition, specifically a hemangioma, and how family systems theory relates to this phenomenon. The literature review also discusses the biopsychosocial model of disease theory and how that model

for understanding disease is appropriate for the diagnosis and treatment of hemangiomas. Additionally, this chapter explores literature regarding trauma and stress as it relates to having a child that is sick or has a medical condition such as a hemangioma. Chapter 3 describes the methodology used in this study that includes the research design, population, sample, implementation procedures, variables, validity, and reliability. It also explores the phenomenon under study for the qualitative portion of this study and categorizes and analyzes the themes and common phrases that emerged from the one qualitative question included in the HRQ instrument.

## CHAPTER 2:

### LITERATURE REVIEW

#### Introduction to the Literature Review

Awaiting the birth of a baby can be extremely stressful, as expecting parents find themselves overjoyed at the prospect of having a baby, but also overwhelmed at the thought of the duties associated with raising a child. For parents of infants born with a birth defect, disability, or medical condition, this overwhelming job called parenting can seem even more consuming (Vacca, 2006, pp. 59-60). According to Ball (2003)

Experts claim that more than fifty percent of marriages crumble when a child is born with or develops medical problems. Sometimes marriages are ruined because the couple's relationship is already weak and the extra stress breaks the bond. And occasionally, healthy relationships break down under the stress of raising a child with special needs. Too much gets stirred up. Emotional highs and lows are the hallmark of a special family, and dealing with those extremes presents its challenges. (p. 15)

The diagnosis of an infant with a birth defect can result in extreme stress for the family. One such birth defect is the hemangioma, the most common benign tumor of infancy, according to Tanner et al. (1998). The estimated prevalence of hemangioma is 10% of all infants by the age of 1 year (Williams et al., 2003). Most hemangiomas occur in the head and neck region (60%) and some patients (20%) have more than one hemangioma. Williams et al. also claimed that "given these facts and that hemangiomas may be unsightly birthmarks, the psychological stress on the developing child and family cannot be underestimated" (p. 229).

The diagnosis, treatment, and impact of hemangiomas on the family system have not been well documented. In an attempt to address this lack of information and in order

to develop uniform standards for the diagnosis and treatment of these lesions, the National Institute of Health (NIH) convened an international hemangioma conference in 2005. Participants included leading medical specialists and worldwide advocacy leaders concerned with the diagnosis and treatment of hemangiomas. At this conference, Chamlin (2005) stated, “Objective clinical outcome measures and subjective quality-of-life (QOL) instruments are lacking for infantile hemangiomas. Such measures are needed to accurately quantify the burden of this disease on children and their families” (p. 396). Thirty years prior to the NIH conference, Drotar et al. (1975) found that “despite the relatively large number of infants with congenital anomalies, our understanding of how parents develop an attachment to a malformed child remains incomplete” (p. 710). In addition, the authors stated that “Although previous investigations are in agreement that the child’s birth often precipitates major family stress, there have been relatively few descriptions of the process of family adaptation” (p. 710). Weinstein and Chamlin (2005) stated that “compared to other equally or less common congenital defects such as a cleft lip or cleft palate, there is a paucity of data on the quality of life in families and children with vascular anomalies” (p. 256). The authors further stated that “The birth of a child with a malformation has been likened by many authors to a crisis situation” (p. 257). Thirty years have passed between Drotar et al. (1975) and Weinstein and Chamlin (2005), and yet little progress has been made to quantify the burden of hemangiomas on the family system.

The following literature review, which spans 30 years, corroborates the hypothesis that having a child diagnosed with a medical condition, such as a

hemangioma, may be a traumatic event that is extremely stressful for the entire family system. It also explores issues such as the medical necessity for treatment of hemangiomas. The diagnosis of acute stress disorder (ASD) and posttraumatic stress disorder (PTSD) symptoms in families that have an infant diagnosed with a medical condition, such as a hemangioma, are also described. The use of the biopsychosocial model for treating such medical conditions as hemangiomas is explored. Additionally, this literature review probes contrary positions to the proposed theory.

The literature review is organized topically and the strategy used for searching literature includes various approaches. An existing library of over 100 peer-reviewed medical journals concerning hemangiomas and other congenital anomalies was used for this literature review. In addition, ProQuest was used to search through Walden University and other university databases for dissertations that focused on family systems, stress, and parenting infants with medical conditions. Other database searches included Medline, Pubmed, and numerous medical university libraries where articles could be obtained for free. EBSCO was the primary resource for journals that dealt with stress and family systems theory. Google was also used for finding journals and articles related to the various noted topics. Lastly, Alibris, an online used book store, was used for purchasing books relevant to the topics in this dissertation. Search terms consisted of various combinations of the following key words: hemangiomas, stress, acute stress, posttraumatic stress disorder, family systems, systems theory, infants with disabilities, biopsychosocial model, and medical necessity.



### Hemangiomas: Facts and Fallacies

Typically, a family of an infant with a hemangioma is first seen by a primary care physician (PCP). PCPs may have little knowledge about vascular birthmarks, or they may be well informed. Since there are no uniform standards concerning the diagnosis and treatment of hemangiomas, the prognosis and treatment options presented to the families vary from physician to physician. Some physicians may refer the family to a pediatric specialist for further evaluation. Others may inform the parents that the hemangioma will eventually involute (regress), so they should just leave it alone. This philosophy of no intervention is antiquated and puzzling for the families, yet it persists because the topic is scarcely taught in medical school and is infrequently addressed at medical conferences or by medical academies. According to Waner and Suen (1999)

An issue often inadequately dealt with, but an extremely important one nonetheless, concerns the true meaning of the term involution. Unfortunately, a large proportion of parents have been led to believe that the hemangioma will disappear completely and leave no trace. Perhaps the current dogma concerning nonintervention, coupled with the lack of substantial experience on the part of physicians, is partly to blame. (p. 26)

Waner and Suen (1999) stated “Only 40% of hemangiomas involute with an acceptable result. Therefore, 60% of all patients with hemangiomas will require some form of corrective surgery” (p. 27). Williams et al. (2003) also noted “If these often disfiguring vascular lesions do not involute early, they may have profound psychosocial effects on the child and the family” (p. 229).

Tanner, Dechert, and Frieden (1998) indicated that most hemangiomas develop during the first few weeks of life, often starting as a tiny red dot, grow for up to 1 year, and then they begin a long and slow involution cycle (p. 446). The authors also claimed

that it is uncommon for involution to be completed by 2 or 3 years of age. More likely, only 50% of hemangiomas have involuted by 5 years of age, 75% by 7 years of age, and 90% by 9 years of age (p. 446). It is because of outdated dogma that postulates that all hemangiomas will involute or resolve on their own that many physicians are reluctant to treat and this is also the reason that many insurance companies are reluctant to approve treatment. From the outdated information provided to the insurance companies, they operate under a false claim that all hemangiomas regress or involute on their own, and therefore, require no intervention.

For decades many well-meaning physicians have touted a no-intervention philosophy. Certain statements have been taken as concrete fact and have been used to refute the more current and appropriate philosophy of early intervention. Such statements include the claim that “for strawberry hemangiomas, the treatment of choice is generally time” (Dieterich-Miller & Safford, 1992, p. 84) and “In the past, the medical community only offered reassurance for the pressing psychosocial needs of these parents and infants, largely because most hemangiomas are not immediately life-threatening and involute over time” (Shakin-Kunkel et al., 1994, p. 524). Tanner et al. (1998) also claimed that “Although the natural history of hemangiomas is one of eventual involution, this is not necessarily good news to the parents of an infant whose physical appearance at birth is unflawed, but quickly becomes abnormal” (p. 446). Another study regarding the treatment of hemangiomas concluded that “Most childhood hemangiomas are managed with a wait-and-see policy and reassurance, since most resolve spontaneously” (Batta et al., 2002, pg. 521). Lastly, according to Simmons and Chiu (2004), “Though many

hemangiomas do not require any form of medical intervention, reassurance with psychological counseling is beneficial” (p. 2).

Some physicians support a wait-and-see approach and others are pushing for early intervention. The outdated philosophy that all hemangiomas will eventually involute and therefore do not require treatment originated long before effective treatment modalities were available. Unfortunately, neither the textbooks nor the primary care physicians have caught up to new treatment. Therefore, outdated literature still persists in medical schools and in the doctor’s office. According to Batniji et al. (2005)

While some authors advocated aggressive management of hemangioams, subsequent articles published in the mid-20<sup>th</sup> century argued that nearly all hemangiomas eventually involuted with no residual deformity. Therefore, a strong opinion developed that the appropriate treatment for hemangiomas was no treatment; this became known as benign neglect. (p. 302)

Additionally, Hochman (2001) compiled 13 articles into a chapter book for physicians concerning the subject of vascular lesions. He made a personal appeal to his medical colleagues to update their knowledge of these lesions:

The teachings that most practicing physicians and those in training rely on with regard to these birthmarks is outdated. The prevailing dogma “leave it alone, it will go away” is no longer universally acceptable in the management of vascular lesions. The emotional and physical consequences are suffered by our patients and their families, some of whom have waited decades for these lesions to “go away.” (p xiii)

Unfortunately, while there is strong evidence that 60% of all hemangiomas do not involute with an acceptable result (Waner & Suen, 1999), many physicians still persist in publishing articles that insist that most hemangiomas will involute without medical treatment. These contrary views are causing many insurance companies to deny treatment because they speculate that the hemangioma will eventually involute. Therefore, they do

not consider treatment a medical necessity. While no one can predict which hemangiomas will involute and look normal and which ones will not, most hemangiomas become lumped into a category classified as not requiring treatment. This issue is one of the many problems facing parents of children with hemangiomas. They struggle to find a doctor who can give them accurate and current information. Once they find the right doctor and receive a treatment plan, they subsequently find out that their insurance company will likely deny approval of treatment. Some physicians report that most of their hemangioma cases are denied treatment. As a result, physicians are reluctant to treat because of the time-consuming paperwork required to appeal the insurance denial. A not-for-profit organization, such as the Vascular Birthmarks Foundation, offer families assistance by providing insurance appeal representatives to assist the families with appealing insurance denials so that their child can receive treatment.

The scenario of having an infant diagnosed with a hemangioma puts the parents on a never-ending roller coaster ride. They are distressed at the thought of their baby becoming deformed from a lesion that is growing and no one can tell them how large it will become. They struggle to find the right physician who can offer them a good prognosis and current treatment options. They rejoice when they find the right doctor, but then they are dismayed when they learn that there is a good chance that the insurance company will not approve treatment. Their options are to raise the resources to pay for the treatment, leave the hemangioma alone and deal with the issues of the deformity, or appeal to the insurance company. According to Canam (1993), "The diagnosis of a chronic illness or disability in a child represents a major stressful life event for all family

members, yet the diagnosis is but one point in the family's ongoing experience with the ill child" (p. 46).

#### Impact of Hemangioma or Other Medical Condition Diagnosis

Canam (1993) stated that, "The diagnosis of a chronic illness or disability in a child represents a major stressful life event for all family members, yet the diagnosis is but one point in the family's ongoing experience with the ill child" (p. 46). In a prospective study that measured the level of stress on the family of an infant undergoing surgery, Majnemer et al. (2006) reported that "Parents of children with chronic health problems are at risk for experiencing ongoing stress, undermining the well-being or overall stability of the families, as well as their ability to cope" (p. 158).

Another study measured the adaptation of parents to their disabled infant. Pelchat et al. (1999) reported "Studies on the reactions of parents to the birth and care of a disabled child have widely demonstrated that such parents experienced more stress and adaptation problems than parents of non-disabled children" (pp. 377-378). Furthermore, Endriga and Speltz (1997) noted in a study regarding infants born with a cleft lip and palate that "Families of infants with cleft lip and cleft palate experience multiple stressors during the infant's first year" (p. 440). An article written about families affected by having an infant with a cleft lip and cleft palate claimed that "cleft parents are often very shocked when they learn their child has a facial disfigurement" (Hodgkinson et al., 2005, p. 10). Additionally, the authors reported that "Parents experience varying degrees of

shock, anger, denial, distress and anxiety. Many parents express a feeling of loss of control” (p. 10).

St. John et al. (2003) argued “A child with chronic illness, such as a craniofacial deformity, can be an overwhelming burden that destabilizes the whole family system” (p. 704). They further noted “Significant stress also develops due to the complex long-term nature of rehabilitating a child with a craniofacial anomaly. The need for multiple hospitalizations and major surgical procedures places a tremendous demand on the ability of these families to cope” (p. 705).

According to Weinstein and Chamlin (2005), “The stigmata of looking different or having a child that looks different can be extremely disabling” (p.256). Furthermore, Tanner et al. (1998) stated “Disfiguring facial hemangiomas were found to be associated with parental reactions of disbelief, fear, and mourning, particularly during the growth phase” (p. 446). Barrio and Drolet (2005) also noted “Hemangiomas often cause extreme parental distress.” (p. 151). The authors further claimed “The effect on parents of having a child with a hemangioma, particularly one on the face, should not be underestimated” (pp. 154-155). Lastly, they stated “A recent study of patients with facial hemangiomas greater than 1 cm in size found that parental reactions of disbelief, fear, and mourning were common and similar in intensity to those found when children had permanent malformations” (p. 155). Williams et al. (2003) noted “If these often disfiguring vascular lesions do not involute early, they may have profound psychosocial effects on the child and family” (p. 229).

Weinstein and Chamlin (2005) further noted that the parents of children with hemangiomas evolve through five stages: “shock, denial, sadness and anger, gradual adaptation, and finally reorganization” (p. 256). According to Barrio and Drolet (2005) “Hemangiomas often cause extreme parental distress.” (p. 151).

Lande, Crawford, and Ramsey (2001), in their article concerning the psychosocial impact of vascular birthmarks on the affected child and the child’s family, suggested that physicians contemplating surgical treatment should conduct a brief inquiry to assess the parents’ initial reaction to the hemangioma. They state:

For some parents, an acute grief reaction accompanies the realization that their child is not perfect. Authors have referred to this response as “the crisis of the birth of a child with congenital abnormalities.” For these parents, shock, denial, and emotional disequilibrium occur. Unfortunately, the grieving process may become chronic, evident psychologically, years after the initial shock. (p. 566)

Other key terms used by parents to describe their reaction to having an infant diagnosed with a hemangioma include fear, anger, disbelief, and helplessness. A study conducted by Drotar et al. (1975) indicated that shock and a sense of helplessness were experienced by most of the parents interviewed regarding their reaction to the birth of a child with a congenital malformation. In another study by Fortier and Wanlass (1984) the results indicated that following the diagnosis of a child with a disability, the family members would begin “working through feelings of grief, anger, guilt, helplessness, and isolation” (p. 13). Additionally, a study by Shakin-Kunkel et al. (1994) intended to assess the well-being of 12 parents of infants with hemangiomas in order to justify the establishment of a self-help group for the parents also included a qualitative study of these parents. The authors interviewed the parents and concluded “there was greater

emotional distress in parents of children with hemangiomas compared with the general population” (p. 528). The authors used existing psychometric instruments to measure parental stress in these parents and compared those findings to normed data.

Symptoms described by parents in these various reports correlate to the definition of a traumatic event. According to an informational brochure produced by the U.S. Department of Health and Human Services in 2003, a traumatic event is a single event or a series of events that causes moderate to severe stress reactions and is characterized by a sense of horror, helplessness, serious injury, or the threat of serious injury or death according to the U.S. Department of Health and Human Services (2003). Though the diagnosis of an infant with a hemangioma correlates to some of the descriptors of a traumatic event, no previous studies have been conducted to support an association between the diagnosis of an infant with a hemangioma and the traumatic reactions by the parents. The interviews conducted by Shakin-Kunkel et al. (1994) reported that the parents were “shocked by the unexpected appearance and growth of the hemangioma” (p. 524). However, the authors did not indicate that the event was traumatic to the parents.

In contrast to the theory proposed in this study that the diagnosis of an infant born with a hemangioma may be perceived as a traumatic event, there have been a small number of studies that indicated the opposite. Barden, Ford, Jensen, Rogers-Salyer, and Salyer (1989) conducted a study to assess the quality of interactions of mothers with their facially deformed infants (p. 819). According to Barden et al.:

Mother-infant interactions were videotaped and rated on discrete and global behavioral measures. Results revealed that mothers of deformed infants rated their parental satisfaction and current life satisfaction more positively than did mothers of normal infants. However, these same mothers were observed to behave in a



consistently less nurturant manner than mothers of normal children. These results suggest that infant facial deformity/unattractiveness may affect the quality of infant-caregiver interactions without parental awareness. (p. 819)

Furthermore, Barden et al. (1989) argued that while the parents of children with a deformity reported being more satisfied than parents of children without a deformity, when they were observed, the parents of children with a deformity “appeared less nurturant” (p. 819). While this study indicated that the parents reported being more satisfied, it also indicated that they were less nurturing and therefore the deformity did have a negative effect on the mother-infant interaction. The authors were looking at parenting infants with a deformity, and were not assessing the parents for various levels of stress or trauma over the diagnosis.

Another study conducted by Bradbury and Hewison (1994) regarding parental adjustment to a child with a deformity indicated that there was more positive adjustment by parents who had support (whether positive or negative) from other family members. The study concluded that parental adjustment was most positive when parental support from other family members was strongest and that parental adjustment was negative when parental support from family members was absent. Similar to the previous study, the authors did not assess the stress levels of the parents, nor did the study indicate how much time had elapsed between the diagnosis of the deformity and the completion of the survey which would have ruled out whether acute stress or posttraumatic stress symptoms could have been present. This study was very narrow in that it focused on the parent-child adjustment phenomenon, rather than assessing the parents for trauma, stress, or any negative impact to the family system.

The impact that hemangiomas have on the family system has not been successfully quantified, nor has any instrument been designed to measure whether or not the impact is a stressful or traumatic event that may result in ASD or PTSD symptoms, until now. Furthermore, the diagnosis of an infant with a hemangioma has been likened to a traumatic event in this literature review. The symptoms described by the parents from several studies correlate to acute stress and posttraumatic stress disorder symptoms, and this has not been previously explored, until now. Other similar studies referenced in this literature review support the theory that having an infant diagnosed with a hemangioma is stressful and can result in acute stress or post traumatic stress disorder symptoms.

#### ASD and PTSD Symptoms and Related Measurement Instruments

According to Ozer, Best, Lipsey, and Weiss (2003) “Roughly 50% to 60% of the U.S. population is exposed to traumatic stress, but only 5% to 10% develop PTSD” (p. 54).

ASD and PTSD symptoms have been described in families that have been impacted by various traumatic events, including events associated with illness or a medical condition of a family member. Some of the symptoms of ASD and PTSD are analogous and some of the symptoms experienced in a traumatic event also correlate to symptoms described in ASD and PTSD. According to Bryant, Harvey, Dang, and Sackville (1998)

Acute stress disorder (ASD) was introduced in the Diagnostic and Statistical Manual for Mental Disorders to describe posttraumatic stress reactions that occur between 2 days and 4 weeks following a trauma. The major difference between the criteria for ASD and PTSD is the formers emphasis on dissociative symptoms. Specifically, to satisfy criteria for a diagnosis of ASD, one must experience a

stressor and respond with fear or helplessness, have at least three of five dissociative symptoms, at least one reexperiencing symptom, marked avoidance, and marked arousal. A major rationale for the introduction of this diagnosis was to identify those individuals who are at risk of developing longer term PTSD. (p. 215)

Self-report instruments developed to measure PTSD have also been used to assess ASD symptoms. For example, the Impact of Events Scale (IES), a self-report measure, has been used to assess symptoms following a traumatic event. Additionally, the Stanford Acute Stress Reaction Questionnaire (SASRQ) has been used to measure acute stress responses.

The psychometric properties of the SASRQ, according to Cardena et al. (2000), “are comparable to those reported for a structured clinical interview” (p. 730). Additionally, the IES has been used in numerous research studies to assess traumatic stress reactions following various traumatic events (Sundin & Horowitz, 2003, p. 870).

The Impact of Event Scale – Revised (IES-R) is an updated version of the IES but it has seven additional items that include hyperarousal symptoms such as anger, irritability, heightened startle response, difficulty concentrating, hypervigilance, and reexperiencing (Weiss and Marmar, 1997). This correlates the IES-R closely to the SASRQ.

The SASRQ was designed to measure dissociative and anxiety symptoms following a traumatic experience. Its measures include: dissociative symptoms, traumatic reenactment, avoidance (of reminders of the trauma), symptoms of anxiety, and impaired functioning. The SASRQ has a high internal consistency for anxiety symptoms (alpha - .91) and dissociative symptoms (alpha .90). The construct validity of the SASRQ was

determined by assessing whether individuals exposed to acute stress or trauma obtained significantly higher test scores than the controls (Cardena et al. 2000). Convergent and discriminant validity was found to correlate to other forms of posttraumatic stress disorder questionnaires than with other forms of pathology, such as depression. Predictive validity indicated that the SASRQ correlated significantly with a PTSD checklist administered at the one-year follow up. According to Cardela et al. (2000), the SASRQ has been shown to have very good reliability, construct validity, discriminant and convergent validity, and predictive validity.

Using the 30-item self-report SASRQ as a template, the Hemangioma Reaction Questionnaire (HRQ) was created based on the theory that the diagnosis of an infant with a hemangioma is a traumatic event that may lead to acute stress or post traumatic stress symptoms. The SASRQ has been modified to measure other condition-specific traumatic events such as earthquakes, motor vehicle accidents, and witnessing a shooting, and measuring the impact of the diagnosis of breast cancer in women, but it had not been modified to measure the impact of having an infant diagnosed with a hemangioma. In a pilot study conducted by Rozell-Shannon (2007) the Hemangioma Reaction Questionnaire (HRQ) was pilot tested in order to determine the reliability of the instrument for measuring this impact of having an infant diagnosed with a hemangioma on the mother. For the pilot study, 13 mothers were surveyed. The pilot focused on validating the instrument rather than on reporting the results of the study. None of the mothers were treated for any stress-related illness. All of the infants were diagnosed within 48 months prior to the completion of the questionnaire. To validate the HRQ, the

test was repeated three to four weeks after completing the initial questionnaire. The results of the study indicated that the test- retest analysis of the HRQ instrument resulted in the Pearson  $r$  of each of the factors ranging from .70 for depersonalization to .97 for a subjective sense of numbing, detachment, or absence of emotional responsiveness (Rozell-Shannon, 2007). Cardena et al. (2000) noted that the issue of test-retest reliability is complex for a scale that evaluates the reactions to a stressful event. In addition, the authors claimed that the correlation in their test-retest study was 69%. These are the same authors who developed the Stanford Acute Stress Reaction Questionnaire (SASRQ). Therefore, the results of the study suggest that the HRQ may be a reliable instrument for measuring the impact of having an infant diagnosed with a hemangioma as a stressful event, and as a possible predictor of ASD or PTSD symptoms (Rozell-Shannon, 2007). This pilot study served as the platform for conducting this descriptive study.

The HRQ and IES-R are self-report instruments that can be readily administered to large numbers of people in a brief amount of time with minimal instruction and are easily scored as a dichotomous or Likert-type measure. According to Elhai, Gray, Kashdan, & Franklin (2005) numerous instruments have been used to assess traumatic event exposure and posttraumatic reactions (p.541). The authors conducted a study to assess which instruments were most widely used by clinicians and researchers to assess traumatic event exposure. The IES-R was listed in the top eight self-report instruments used by clinicians and the SASRQ was listed as a self-report instrument commonly used by researchers.

Helplessness has been noted in the definition of a traumatic event and is also reported as a feeling experienced by the parents of an infant or young child diagnosed with a medical condition. However, no one has used the hypothesis of looking at the diagnosis of a hemangioma as a traumatic event, prior to this study. Nor have any studies examined the symptoms associated with the diagnosis.

To support the theory that having an infant diagnosed with a hemangioma may be traumatic, similar studies of other birth defects or medical conditions in infants were explored. A recent study was done to examine the prevalence of acute stress disorder in parents of infants hospitalized in the neonatal intensive care unit (Shaw et al., p. 206). According to the authors, “researchers have identified posttraumatic stress disorder (PTSD) as a model to describe and explain the psychological reaction of parents to their NICU experience” (p. 206). The authors claimed that ASD symptoms, following a traumatic event, are considered to be a precursor to PTSD. The SASRQ was one of the instruments used by the authors to assess the ASD symptoms in the parents. The study concluded that 28% of the parents in the study met the symptoms criteria for a diagnosis of ASD, “which is consistent with previous findings on the development of ASD after a traumatic event” (p. 209). The symptoms included hyperarousal, flashbacks related to the event of the birth and NICU hospitalization, and avoidance of contact with the NICU. These symptoms are consistent with the diagnosis of a traumatic event and could provide a foundation for the theory that having an infant diagnosed with a hemangioma could result in the same type of traumatic symptoms.

Another similar study was conducted to investigate the posttraumatic stress response of mothers after the birth of a very low birth weight infant (Kersting et al. 2004). The authors concluded “the situation of a mother who has given birth to a very low birth weight infant is a complex, long-term traumatic event that necessitates ongoing emotional support extending beyond the period immediately after the birth” (p. 473). Also noteworthy in this study was the fact that the mothers of the premature infants displayed no “significant reduction in post traumatic symptoms (based on the Impact of Events Scale totals) even 14 months after the birth” (p. 473). Additionally, according to Jedlicka-Kohler et al. (1996), “The diagnosis of cystic fibrosis in children represents a severe blow to parents, confronting them with traumatic information. A few studies have emphasized high levels of emotional stress during information giving, but none has examined potential cognitive impairment in the parents caused by the trauma of learning the diagnosis” (p. 204) (p. 204).

In order to support the theory that having an infant diagnosed with a hemangioma is a stressful or traumatic event that may result in ASD or PTSD symptoms, Table 2 was created. It compares the symptoms described by parents of children with various craniofacial abnormalities, medical conditions, birth defects, or hemangiomas with symptoms defined for an acutely stressful or traumatic event:

Table 2

*Comparative analysis of stressful symptoms reported by parents*

Symptoms defined in various studies as experienced by parents	Key terms corresponding to parental reactions	Symptoms defined as corresponding to a traumatic event, ASD, or PTSD	Key terms corresponding to trauma, ASD, or PTSD definitions
Tanner et al. (1998), state that “Disfiguring facial hemangiomas were found to be associated with parental reactions of disbelief, fear, and mourning	Disbelief Fear Mourning	According to Bryant et al. (1998), for a diagnosis of ASD, one must experience a stressor and respond with fear or helplessness (Criterion A),	Fear Helplessness Reexperiencing
Weinstein and Chamlin (2005) claimed that the parents of children with hemangiomas evolve through five stages: “shock, denial, sadness and anger, gradual adaptation, and finally reorganization” (p. 256)	Shock Denial Sadness Anger Adaptation Reorganization Distress Crisis	have at least three of five dissociative symptoms (Criterion B), at least one reexperiencing symptom (Criterion C), marked avoidance (Criterion D), and (Criterion E) marked arousal (p. 215).	Avoidance Marked Arousal Severe Stress Horror Helplessness Serious Injury Death
According to Barrio and Drolet (2005) “Hemangiomas often cause extreme parental distress” (p. 151).	Acute grief Shock Denial Emotional	An informational brochure produced by the U.S. Department of Health and Human Services, 2003, stated	Numbing Detachment Reduction in Awareness

*Table continues*



Weinstein and Chamlin (2005) claimed that “The birth of a child with a malformation has been likened by many authors to a crisis situation” (p. 257).	Disequilibrium	that a traumatic event is a single event or a series of events that causes moderate to severe stress reactions and is characterized by a sense of horror, helplessness, serious injury, or the threat of serious injury or death. (U.S. Department of Health and Human Services, 2003).	Derealization Depersonalization Dissociative Amnesia
	Helplessness		
	Grief		Avoidance of stimuli
	Anger		Anxiety
Lande et al. (2001) stated, “For some parents, an acute grief reaction accompanies the realization that their child is not perfect. Authors have referred to this response as “the crisis of the birth of a child with congenital abnormalities.” For these parents, shock, denial, and emotional disequilibrium occur” (p. 566).	Guilt		Increased Arousal
	Helplessness	According to Cardena et al. (2000) the SASRQ measures dissociative symptoms such as numbing, detachment, or absence of emotional responsiveness, along with a reduction in awareness of one’s surroundings, derealization, depersonalization, and dissociative amnesia. It also	
	Isolation		Impairment in Functioning
	Shock		
Drotar et al. (1975) argued that shock and a sense of helplessness were experienced by parents	Anger		
	Denial		Same symptoms as above
	Distress		
	Loss of Control		
<i>Table continues</i>	Grief		
	Rage		
	Disappointment		
	Anxiety		
<i>Table continues</i>	Protectiveness	indicates if the event is persistently reexperienced and	Distress
	Guilt	if there is marked avoidance of	Anxiety

interviewed, regarding their	Depression	stimuli that arouse	Nightmares
reaction to the birth of a child with	Significant Stress	recollections of the trauma and	
a congenital malformation (p. 710).		marked symptoms of anxiety or	Numbing
		increased arousal and	
Fortier and Wanlass (1984) noted		impairment in functioning. (pp.	Intrusion
that following the diagnosis of a		719-731).	
child with a disability, the family			Avoidance
members would begin “working		The HRQ was modified from	
through feelings of grief, anger,		the SASRQ and, therefore,	Hyperarousal
guilt, helplessness, and isolation”		measures all of the symptoms	
(p. 13)		indicated for the SASRQ.	Anger
“Parents experience varying		The IES-R assesses current	Irritability
degrees of shock, anger, denial,		subjective distress and anxiety	
distress and anxiety. Many parents		for any specific life event	Difficulty
express a feeling of loss of control”		(Horowitz et al.,1979) The	Concentrating
(Hodgkinson et al., 2005, p.10).		instrument measures responses	
		sets for intrusion symptoms	Hyper vigilance
		such as nightmares, intrusive	
Pope (2000) claimed that parents of		feelings and imagery, and	Reexperiencing
children with special needs		avoidance symptoms such as	
“experience a range of emotions,		numbing of responsiveness,	
including grief, rage,			
disappointment, anxiety,		and avoidance of feelings,	
		situations, or ideas (pp. 209-	
<i>Table continues</i>		218). Weiss and Marmar	

protectiveness, guilt, and depression” (p. 70).

According to St. John et al. 2003), “A child with chronic illness, such as a cranio- facial deformity, can be an overwhelming burden that destabilizes the whole family system” (p. 704). They claimed that “Significant stress also develops due to the complex long-term nature of habilitating a child with a craniofacial anomaly” (p. 705).

(1997) add 7 additional items with their revised IES-R instrument such as anger, irritability, heightened startle response, difficulty concentrating, hypervigilance; and dissociative-like-reexperiencing (p 1).

### Impact on the Family Systems

According to Bertalanffy (1968), just about everything can be viewed as a system: medicine, machines, politics, chemicals, air, and even families (p. 4). Nearly 40 years ago Bertalanffy (1968) stated that:

The concept of systems pervaded all fields of science and penetrated into popular thinking, jargon, and mass media. Systems' thinking plays a dominant role in a wide range of fields from industrial enterprise and armaments to esoteric topics of pure science. Innumerable publications, conferences, symposia and courses are devoted to it. Professions and jobs have appeared in recent years which, unknown a short while ago, go under names such as systems design, systems analysis, systems engineering and others. (p. 3)

Families are a system and when one member, such as an infant, is diagnosed with a medical condition, the entire family system is impacted based on family systems theory. Fortier and Wanlass (1984) stated "The official diagnosis of a handicapped child marks the occurrence of a family crisis" (p. 13). Furthermore, in an article that proposed a model of adaptation and change for families of chronically ill children, Kazak (1989) stated "Having a child with a physical handicap, chronic condition or potentially terminal disorder has long been recognized as stressful for the entire family" (p.27). The author further claimed "It is not uncommon to see mild to moderate levels of distress or psychopathology somewhere within the family system" (p. 25). Additionally, the author stated "From a systems perspective, a problem in any member of a family has an effect on all other members, and changes in any member of the system affect all others" (p. 26).

According to Fleming (2007), “Family systems theory has had a significant impact on the study of families and on approaches to working with families. It has guided research into such areas as understanding traumatic events or chronic health issues and their impact on individuals and families” (p. 1). Additionally, Cox and Paley (1997) noted that the systems approach to understanding families meant that the family needed to be studied as one complete unit, but also studied for their interrelations (p. 246). They further stated that:

Using the perspective of the family as a system highlights the idea that the family is a complex, integrated whole, wherein individual family members are necessarily interdependent, exerting a continuous and reciprocal influence on one another. Thus, any individual family member is inextricably embedded in the larger family system and can never be fully understood independent of the context of that system. (p. 246)

Studies have been conducted regarding how family systems are impacted by the illness of one member, or more specifically by having a child born with a disease or disability. According to Jedlicka-Kohler et al. (1996), “The diagnosis of cystic fibrosis in children represents a severe blow to parents, confronting them with traumatic information. A few studies have emphasized high levels of emotional stress during information giving” (p. 204). Additionally, Canam (1993) stated “The diagnosis of a chronic illness or disability in a child represents a major stressful life event for all family members, yet the diagnosis is but one point in the family’s ongoing experience with the ill child” (p. 46). According to Vacca (2006), “It is imperative that mental health providers become aware of the stressors of raising a child with a disability, and how this affects the family system and their ways of managing their daily life both physically and emotionally” (p. 70).

These are just a few of the studies that liken the diagnosis of a medical condition in an infant or young child to a traumatic event for the entire family system. They are used to provide support to the hypothesis that having an infant diagnosed with a hemangioma is a stressful and traumatic event for the entire family system.

Kazak (1992) presents an interesting cautionary note that is a limitation to family systems theory, as it relates to this application. The author indicated that it can be problematic to view the family as one unit when one or more members of the family may have higher or lower levels of a measurement symptom, such as stress. If a single measurement is representative for the entire family, it can be misleading. The author suggested that any instrument used to measure such things as family stress should be done on an individual basis first, and then as a family unit. The results should then be analyzed and reported separately and then combined. (pp. 122-123). Hodapp, Fidler, and Smith (1998) also claimed that in order to get a good measure of stress and coping in families of children with a rare medical condition, individual self-report measures should be conducted and then measures of the combined family should be done to determine if the stressors are individual or collective (p. 331). Lastly, according to Weinstein and Chamlin (2005)

It is important for physicians seeing children with vascular anomalies to anticipate and monitor for signs or psychosocial impairment in both affected children and their parents, and to provide resources to those who might benefit from psychological intervention. (p. 258)

Having a child with a medical condition, specifically a lesion like a hemangioma, impacts not only the child, but the entire family system. As a result, any treatment plan should also consist of evaluating the entire family in order to determine if a mental health

treatment plan is needed to maintain harmony in the family system. Individual, in addition to family assessments should be considered.

### Biopsychosocial Model – Treating the Hemangioma and the Family

Brogdon (1999) studied families affected by a hemangioma or other vascular birthmark and concluded that “uniqueness and specialness are commonly encountered in these families” (p. 224). He also claimed that:

There is a preponderance of denial present in the medical practitioners treating these families, denial in that many of these practitioners are too conservative or fearful of aggressive, appropriate surgical intervention. This response (or lack of response) frequently results in additional psychosocial trauma to both the child and family. This wait and see conservative attitude can and does result in facial disfigurement in many children. (p. 224)

Brogdon concluded in his study that treatment in and of itself is not a panacea but rather timely and rapid medical intervention interfaced with integration of biomedical and psychological expertise will ensure the best outcome for the entire family, not just the child with the birthmark (p. 228). This comprehensive approach toward treating both the biological and psychological component of a medical condition, such as a vascular birthmark, has its foundation in a theory introduced over 40 years ago, called the Biopsychosocial Model.

Engel (1962) is responsible for the design of the biopsychosocial model. This model evolved in the 1960s out of Engel’s personal experience as an internist, psychiatrist, and psychoanalyst and his efforts to formulate a comprehensive understanding concerning human behavior and disease. He noted that this comprehensive

view of disease had its deficiencies but it was a step in the right direction toward a combined plan of care for patients. Engel was aware that most physicians had conceptualized human beings in terms of “biologically determined developmental destiny, adapted to and adjusting in a physical, interpersonal, and social environment” (p. 239). However, through the biopsychosocial model, Engel introduced physicians to the “consideration of human beings as a biological, psychological, and social organism. These components encompass the deviations referred to as disease, the proper subject of medicine” (p. 239).

More recently, Williams et al. (2003) conducted a study to assess the psychosocial impact of hemangiomas and the treatment of the hemangiomas on the affected child and the child’s family. The authors concluded that “Our survey did not seem to indicate that the children experienced significant emotional trauma from their condition, nevertheless, their families experienced appreciable emotional and psychological distress” (p. 229). In discussing the psychosocial impact of hemangiomas on the family, the authors further claimed that “Rarely do physicians weigh the psychological burden that a disease process carries or, even less frequently, what steps should be taken to avoid the development of such emotional trauma” (p. 233). While the biological component of hemangiomas is addressed, the psychosocial implications on the family system continue to be neglected. Williams et al. (2003) also stated that:

The treatment of vascular lesions has undergone a revolution in thought and practice in the past 10 years. Earlier intervention and advanced therapeutic modalities, such as laser therapy, have permitted the patient and family the opportunity to remove the hemangioma earlier and more effectively and, thereby, to mitigate the psychosocial impact that the hemangioma may otherwise have. (p. 233)



Engel's biopsychosocial model views the entire human system as one general system, thus concurring with Bertalanffy's general systems theory where everything is seen as part of a whole. Engel further views the study of the patient and disease as a general human system composed of the biological, psychological and social subsystems. Engel states that traditional medicine's concept of the patient and disease "is reductionist and tends to restrict attention to only one aspect (unit) of disease, and one which is not necessarily always present" (p. 240). Engel uses grief as an analogy. He says that grief is a process with steps and procedures that ultimately reaches a state of healing. This process can be compared to a burn that has steps and procedures which also reaches a state of final healing. However, Engel made a point that while the burn would be treated because the patient presented with a physical condition, if a patient presented with grief, he or she may not be treated because there would be no visible signs or symptoms to treat (p. 254). Engel further stated that some patients who experience grief present with a physical symptom and that will be treated but the grief will be completely ignored (p. 254). Chamlin (2005) addressed the issue of the psychosocial effects of hemangiomas on the family and also addressed the fact that a subjective way of measuring this impact is lacking. Chamlin stated:

Objective clinical outcome measures and subjective quality-of-life (QOL) instruments are lacking for infantile hemangiomas. Such measures are needed to accurately quantify the burden of this disease on children and their families. Quality-of-life measures are needed because clinical measures and QOL are often not highly correlated. (p. 396)

Chamlin (2005) further stated that “standardized clinical data collection forms need to be developed” (p. 396). Chamlin indicated that though this type of data is difficult to quantify, it can be successfully developed to measure the effects of the hemangioma on the child and the parents (p. 396). Chamlin further suggested that the development of such an instrument should also include issues for parents that focus on loss of control, anxiety, guilt, grief, sadness, concerns about disfigurement and self-esteem, and fear of strangers’ reactions (p. 396). Chamlin emphasized that “the ideal QOL instrument would be disease specific and age specific with comprehensive content and established psychometric properties” (p. 396). According to Chamlin no reliable instrument to achieve these goals exists and that in order to develop one, an extensive literature review must be accomplished and a qualitative interview study should be conducted to collect data needed to draft such an instrument (p. 396).

In order to properly assess the impact of hemangiomas on the family system, an instrument, such as the one suggested by Chamlin (2005), needed to be developed. This instrument could be used by physicians in combination with the physical examination of the hemangioma. As a result, a comprehensive approach to treating the hemangioma and the family with consideration of the biopsychosocial model offers the best hope for a positive outcome for the entire family system.

According to Lande et al. (2001) “The diagnosis and treatment of vascular birthmarks is a unique opportunity for psychiatrists to provide psychosocial guidance, thereby reducing stress and promoting a favorable outcome” (p. 562). Furthermore, according to Pope, Tillman and Snyder (2005), “Parenting stress, if identified as a risk

factor for children with craniofacial anomalies, is easy to assess in a hospital or clinic setting, and appropriate supportive services could then be provided to assist parents at this vulnerable juncture” (pp. 356-357).

### The Medical Necessity of Treatment

While the visible appearance of and medical conditions associated with cleft lip and cleft palate are different from hemangiomas, both conditions are birth defects that affect the appearance of the infant. According to Singer and Bergthold ( 2001)

Cleft lip and cleft palate are common birth defects with functional and cosmetic dimensions. Treatment often falls into the gray area between reconstructure and cosmetic surgery. As a result, medical necessity determinations involving this condition have proven to be especially problematic. (p. 2162)

Additionally, the authors stated that “medical necessity is a legal term used in most commercial health plan contracts to establish which interventions a health plan will cover for its members” (p. 2162).

Families of infants diagnosed with a hemangioma are confronted with the lack of consistent information and an uncertainty over whether they will find an adequately trained physician. They struggle to get accurate information, and once they find the right physician, then they face the task of proving to their insurance company that treatment is a medical necessity. According to Glassman et al. (1997)

Policymakers, insurers, and health care organizations invoke “medical necessity” to define which services should be offered to patients. However, this term has many meanings and many uses, ranging from general definitions used in health insurance contracts, to specific definitions used to identify which services are “crucial” for individual patients or patient populations. Consequently, “necessity” has an ambiguous meaning; not surprisingly, the manner in which the concept of necessity is applied varies significantly within and across providers, insurers, and states. (p. 152)

The authors also stated that in deciding whether a treatment is medically necessary, “insurers and other health care providers have traditionally focused on simple, deterministic decision rules based on the idea that without the service, harm will come to the patient that with the service, a potentially beneficial outcome will result” (pp. 152-156). There is no question, based on the extensive literature review concerning the need for treatment of hemangiomas that treatment will result in a beneficial outcome for the entire family. This argument must be made by each family fighting the denial of treatment. Treatment not only successfully removes the deforming lesion, it also eases the psychosocial pain of the family and reduces the intense stress of living with a child with a birth defect. If a medical necessity is not established and treatment is denied, families will be forced to appeal the denial, live with the deformity, or raise the resources needed to pay for the treatment. In addition, they will linger in a mire of acute stress and psychosocial trauma until the hemangioma is successfully treated.

### Summary

Having an infant is stressful, but having an infant with a birth defect, like a hemangioma, poses an enormous amount of stress on the entire family system. This literature review substantiated that some of the feelings expressed by parents of children with hemangiomas parallel some components of a traumatic event and also align with symptoms of acute and posttraumatic stress disorder. Describing the diagnosis of a hemangioma as a traumatic event which results in acute or post traumatic stress disorder symptoms has never been conducted in a study, until now. This descriptive study is

important to the families affected by a hemangioma because acute stress and posttraumatic stress disorder symptoms are considered medical conditions that require intervention. As such, that diagnosis will support a claim that treatment of the hemangioma is a medical necessity. This claim is needed due to the fact that many requests for treatment are denied because insurance companies consider treatment to be cosmetic or elective. However, this literature review supports the theory that the diagnosis of an infant with a hemangioma may be a traumatic event that may result in acute or posttraumatic stress symptoms, thereby establishing a medical necessity for treatment. If a medical necessity for treatment is not established, the families will have to raise the resources to pay for treatment or leave the hemangioma and deal with the daily struggles involved with having an infant with a deforming vascular tumor. Furthermore, if the hemangioma is treated by using a biopsychosocial approach that addresses the physical, psychological, and social components of this condition, the best outcome for the entire family will be certain.

It is unconscionable that during a time when human organs are transplanted from one person to another, and artificial limbs offer hope to individuals who never thought they would walk or regain the use of a missing limb, that parents of an infant with a benign and often disfiguring vascular tumor, such as a hemangioma, are told that treatment is cosmetic and not medically necessary. It is even more distressing that the psychosocial impact of having an infant diagnosed with a hemangioma has been excluded from the treatment plan.

## CHAPTER 3:

### RESEARCH METHOD

#### Introduction to Study Design

The birth of an infant can be extremely stressful for new parents, and when that infant is diagnosed with a birth defect, such as a hemangioma, the joyful birth experience can become even more stressful. No previous studies have measured the levels of stress of parents of infants diagnosed with a hemangioma, and no instrument, prior to this study, has been developed to measure the burden of hemangiomas on the family system. This mixed methods descriptive survey study addressed both of these gaps. The study design included two self administered questionnaires, the Hemangioma Reaction Questionnaire (HRQ) and the Impact of Event Scale-Revised (IES-R). One of the questionnaires, the HRQ was developed by this researcher and included, in addition to the quantitative questions, a personal narrative question for participants to describe (qualitative) the most profound event that occurred following the diagnosis of the hemangioma. The two self-administered questionnaires (quantitative) were used because a larger group could be studied, rather than the small number that is typical for a qualitative interview or a single case study. Survey research is a quantitative research method which, according to Johnson and Christensen (2004), “is a form of nonexperimental research in which questionnaires or interviews are used to gather information, and the goal is to understand the characteristics of a population” (p. 197). One of the goals of this study was to describe whether or not the mothers perceived the diagnosis of the hemangioma as a stressful or traumatic event.

In addition to allowing for the use of a larger group, the two instruments enabled a quantification of the responses of the participants through the descriptive statistics.

There is one open-ended question in the HRQ that asks the mothers to describe the most profound event that occurred since the diagnosis of the hemangioma. Because quantitative research alone does not allow for the exploration of a phenomenon, a combined descriptive and qualitative approach was used in this study in order to investigate this phenomenon. The theory put forth in this study proposed that the diagnosis of the hemangioma in the infant is stressful and is perceived as a traumatic event. The open-ended question enabled themes to emerge. The qualitative component also allowed the personal descriptive experiences of the participants to be correlated to the quantitative responses from the surveys they completed. According to Johnson and Christensen (2004), "The use of multiple perspectives, theories, and research methods is viewed as strength in educational research" (p. 49). The combined approach used for this study included the following steps:

1. The objectives for this mixed methods study were explored and described.
2. The data type for the qualitative portion included themes that emerged from the words used by the mothers to describe their most profound event surrounding the diagnosis of the hemangioma, and the data type for the quantitative portion will relied on frequency distributions using descriptive statistics.
3. The analysis and interpretation of the qualitative portion relied on searching for themes and patterns in the mothers' personal responses, and the

quantitative portion involved statistical analysis (p. 49). The frequency of the responses is depicted in graphs and charts.

Johnson and Christensen (2004) further stated that, by “combining two or more research methods with different strengths and weaknesses in a research study, you can make it less likely that you will make a mistake” (p. 50).

The specific research design for the quantitative portion of this study is referred to as the one-shot case study (Campbell & Stanley, 1963, p. 6). According to the authors, “Much research in education today conforms to a design in which a single group is studied only once, subsequent to some agent or treatment presumed to cause change” (p. 6). None of the other Campbell and Stanley designs were considered since it was not practical to have a control group, nor was it effective to have a type of static group comparison. There was consideration given to a post follow-up of the surveys. This was considered because PTSD symptoms are more enduring than ASD symptoms. However, since the study group included mothers of infants diagnosed with a hemangioma up to 1 year prior to the completion of the survey, this meant that any lingering symptoms would constitute PTSD rather than ASD. According to Classen (n.d.), “ASD symptoms present in the immediate aftermath of a traumatic event. The distress must occur within four weeks of the trauma and persist for at least two days” (p. 401). From that point forward, if symptoms persist, the diagnosis changes to PTSD. According to Classen “ASD was added to the *Diagnostic and Statistical Manual (DSM)* to account for the 1-month gap between the traumatic event and the onset of symptoms, which forms the basis of the definition of PTSD” (p. 420).



This study focused on describing and exploring the self-reported stress levels of the mothers participating in the study, and assessed whether or not they perceived the diagnosis as a traumatic event.

A qualitative research design analysis has been included in this study because there is one open-ended question in the HRQ that required the exploration and identification of similar data. A phenomenological approach was selected because it “allows the researcher to identify the essence of human experiences concerning a phenomenon” (Creswell, 2003, pp. 14-15). An ethnographic approach was not considered since this study did not focus on an intact cultural group in a natural setting over a prolonged period of time (p. 14). Additionally, grounded theory was not selected because this study was focusing on a single group and case study was not selected since the event that would be studied, the diagnosis of the hemangioma, required numerous cases to support the theory that the diagnosis is stressful and is also perceived as a traumatic event. Lastly, a narrative approach was considered but was not selected because the study required more than a few individuals to “provide stories about their lives” (p. 15) with respect to the diagnosis of the hemangioma. Random selection was not considered since it would require hundreds of names of mothers to be collected and the random sample selected from that group. However, there would be no assurance that the group represented the entire population of mothers who have had an infant diagnosed with a hemangioma; instead, a convenience sample was utilized for this study.

With regards to the qualitative analysis portion of the study, the process of data analysis include open coding, where categories were formed by including key terms and

themes used by the mothers to describe the most profound event associated with the diagnosis of the hemangioma.

### Research Questions

1. Using the HRQ instrument, what level of stress is reported by mother's who have an infant that is diagnosed with a hemangioma and who complete this survey?

*H<sub>01</sub>*: There is no reported stress level for mothers who have an infant diagnosed with a hemangioma who completed this survey.

*H<sub>A1</sub>*: There is a reported level of stress for mothers who have an infant diagnosed with a hemangioma who completed this survey.

2. Using the IES-R, what impact of event measure is reported by mothers of infants diagnosed with a hemangioma and who complete this survey?

*H<sub>02</sub>*: There is no reported measure from the diagnosis of an infant with a hemangioma based on the responses from the mothers who complete this survey.

*H<sub>A2</sub>*: There is a reported measure from having an infant diagnosed with a hemangioma based on the responses from the mothers who complete this survey.

3. For the qualitative question included in the HRQ, what common themes have emerged from the question which asks the mothers to describe the most profound event that has occurred since the hemangioma was diagnosed?

*H<sub>03</sub>*: There are no common themes among the mothers who completed the qualitative question in the HRQ.

$H_{A3}$ : There are common themes among the mothers who completed the qualitative question in the HRQ. .

This chapter also includes a description of the target population from which the sample was drawn, details about the instruments used, and data collection and analysis procedures. Lastly, measures taken to protect the rights of the participants were summarized in this chapter.

### Target Population

Participants included mothers of infants with a hemangioma and who were between 17 and 41 years of age. Only mothers who have an infant that was diagnosed with a hemangioma within the year prior to completing the survey were included in this study in order to determine if their symptoms were consistent with ASD or PTSD.

### Sampling Procedure

An announcement was posted on the Vascular Birthmarks Foundation's nine international Web sites and on the MSN Vascular Birthmarks Support Group web site. There are currently over 1,000 members of these Web sites. Members are primarily comprised of mothers of infants diagnosed with hemangiomas. There are 300 new members who join the hemangioma support groups each year. In addition, two physicians referred patients to the web site to consider volunteering for the study. Both physicians conduct monthly clinics. All of the participants in the study were volunteers. A nonrandom convenience sample of volunteers was used to recruit volunteers from the Vascular Birthmarks Foundation Web sites, and the MSN Vascular Birthmark Support

Group Web sites. According to Johnson and Christensen (2004), “a convenience sample consists of people that are available or volunteer or can be easily recruited and are willing to participate in the research study” (pg. 214).

The participants were able to go from a link posted on the hemangioma websites directly to SurveyMonkey to participate in the study. All consents and acknowledgements were included in the on-line study. All respondents were assigned an anonymous number in order to maintain anonymity. It took an average of 10-15 minutes to complete the entire study. All data are maintained on the researcher’s desktop and no information linking the responses to the respondents was or will be shared with anyone.

#### Participant Protection

All participants were volunteers. They were informed of the intent of the study, and their obligations before they participated. Names of the participants were not collected. All participants were assigned a sequential number that corresponded to when they completed the survey so that their responses were anonymous and confidential. Participants were informed that at any time during the study they could withdraw if they felt coerced or threatened in any way. The name and contact information of a psychotherapist was posted on the survey for any participants who felt they needed to talk to a professional at any time during or after participating in the study. This protection was made clear in the IRB package and was detailed in the consent form, letter of intent, web site announcement, and in the confidentiality agreement. Data collected for this study are maintained by the researcher and has not been nor will it ever be disclosed to any other source.

### Sample

The proposed convenience sample for this study was identified as 169 mothers of infants who were diagnosed with a hemangioma within one year of the mothers completing the study. This proposed number was based on the population size of 300 new mothers of infants diagnosed with a hemangioma who join the various support groups each year. According to Johnson and Christensen (2004) a population ( $N$ ) of 300 has a recommended sample size ( $n$ ) of 169 (p. 218). All participants were between 17 and 41 years of age, with an infant diagnosed with a hemangioma within one year prior to the mother's participating in the study.

Many similar studies were done with a much smaller participant size. In a study that was conducted by Jedlicka-Kohle et al. (1996), a survey was administered to 46 parents of children diagnosed with cystic fibrosis in order to study the parents' emotional and cognitive reaction to the diagnosis. The results indicated that parents learning the diagnosis perceived it to be a traumatic and stressful event. Recommendations were made as a result of this study for ongoing educational sessions between the physicians and the parents to reduce the stress and to maximize the learning process for management of cystic fibrosis. (pp. 204-207). Furthermore, a study by Cate-Miller, Cate, Watson, and Geronemus (1999) used 46 parents in their study (p. 190). Lastly, Drotar et al. (1975) observed the parents of 20 children to analyze parental reactions to the birth of a child with a congenital malformation (p. 710). All studies were deemed conclusive in that all of the authors theorized that having a child with a medical condition, facial deformity, or birthmark, is a stressful experience.

This sample was nonrandom as opposed to random because it was not possible to reach an entire population of mothers of infants diagnosed with a hemangioma. Furthermore, most of the mothers who seek information from the Vascular Birthmarks Foundation are given a diagnosis from a vascular birthmark specialist that they find through the foundation web site. Therefore, the hemangioma related websites provided a better pool to solicit from to insure that the mothers who participated in the study had an infant with a hemangioma and not some other type of benign tumor.

#### Study Instrumentation and Materials

There were two self-reporting instruments completed by the participants. The Hemangioma Reaction Questionnaire (HRQ) and the Impact of Event Scale – Revised (IES-R).

##### *The Hemangioma Reaction Questionnaire (HRQ)*

The HRQ consists of 30 questions, an open-ended question which asks the participants to describe the most profound event that occurred following the diagnosis of the hemangioma, and an unnumbered question that asks the participants to indicate how many days they experienced any symptoms of distress.

The HRQ is a modification of the Stanford Acute Stress Reaction Questionnaire (SASRQ). The SASRQ is a valid and reliable instrument designed to measure dissociative and anxiety symptoms following a traumatic experience. It is relevant to the diagnosis of ASD and includes a description of the event, how disturbing it was, and how many days the individual experienced the worst symptoms, according to Cardena et al. (2000, p. 722). It is also used to predict PTSD symptoms in victims of the event being

measured (pp. 720). Its measures include: dissociative symptoms, traumatic reenactment, avoidance of reminders of the trauma, symptoms of anxiety, and impaired functioning. The SASRQ has a high internal consistency for anxiety symptoms ( $\alpha = .91$ ) and dissociative symptoms ( $\alpha = .90$ ). The construct validity of the SASRQ has been measured by assessing whether individuals exposed to acute stress or trauma events obtained significantly higher test scores than the controls, according to Cardena et al., 2000, p. 727). Additionally, several uses of the SASRQ reported high scores for individuals exposed to acute stress or traumatic events such as journalists covering an execution, patients diagnosed with cancer, emergency rescue workers, and Gulf War veterans (p. 727).

Convergent and discriminant validity for the SASRQ were found to correlate to other forms of post traumatic stress disorder questionnaires than with other forms of pathology, such as depression (p. 728). Predictive validity indicated that the SASRQ correlated significantly with a PTSD checklist administered at the one-year follow up. Additionally, according to Cardena et al., there was also some evidence of clinical value of the SASRQ because individuals scoring high on the SASRQ were more likely to engage in dangerous risk-taking than low scores (p. 729). Furthermore, the authors noted that the SASRQ has been shown to have very good reliability, construct validity, discriminant and convergent validity, and predictive validity. Additionally, the SASRQ can be readily administered to large numbers of people in a brief amount of time with minimal instruction and it is easily scored as a dichotomous or Likert-type measure (p. 730).

For all of these reasons, the SASRQ was selected as the instrument that would be modified to a hemangioma-specific instrument in order to measure and describe if the diagnosis of an infant with a hemangioma is perceived by the participants as a stressful and traumatic event that could result in ASD or PTSD symptoms. Since the SASRQ had been modified numerous times to be event specific, it was easily modified to change the event from exposure to an earthquake, or reaction to the diagnosis of cancer, to the reaction to the diagnosis of a hemangioma. When modifying the SASRQ to the HRQ, 14 of the 30 questions remained the same, and 16 were modified to be hemangioma-specific. For example, Question 5 in the SASRQ stated, “I tried to avoid feelings about the stressful event” was modified to, “I tried to avoid feelings about the hemangioma.”

#### *The HRQ Pilot Study*

Once the HRQ was finalized, it was pilot tested to determine instrument validity. Participants included women only, first time mothers, between 18 and 40 years of age, infant’s hemangioma required treatment but had not been previously treated, mother had not been previously treated for stress or PTSD, and the hemangioma was located on the head and neck region and was visible. An announcement was posted on the Vascular Birthmarks Foundation’s nine international Web sites and the MSN Birthmarks Support Group web site. All of the participants were volunteers. They downloaded the questionnaire, consent form, and participant form and submitted them electronically to the researcher. A follow-up survey was sent to all participants three to four weeks after the first survey was completed, in order to test the reliability of the new instrument. All respondents were assigned a letter identifier (A through M) to maintain anonymity. All



data were kept on the researcher's desk top and no information linking the responses to the respondents was shared (Rozell-Shannon, 2007, pp. 9-11). All responses were entered into Excel and analyzed using SPSS, a simple and user-friendly software package that can analyze data.

### *HRQ Test-Retest Results*

Table 3 depicts the Pearson  $r$  correlation coefficient for the nine various measures in the HRQ test-retest pilot study by Shannon (2007):

Table 3

*Pearson r for HRQ Test-Retest Reliability*

Measure	Pearson <i>r</i> for test-retest reliability
Dissociative symptom (1): Subjective sense of numbing, detachment, or absence of emotional responsiveness	.970
Dissociative symptom (2): A reduction in awareness of one's surroundings	.931
Dissociative symptom (3): Derealization	.701
Dissociative symptom (4): Depersonalization	.705
Dissociative symptom (5): Amnesia	.710
Traumatic event is persistently reexperienced	.943
Marked avoidance of stimuli that arouse recollection of the trauma	.939
Marked symptoms of anxiety or increased arousal	.801
Impairment in functioning	.859
.	

Thus, the HRQ is a new instrument that was pilot tested to determine the reliability of the instrument for measuring the burden of having an infant diagnosed with a hemangioma. The results in the preceding table indicate a range of 70% to 97% test-retest reliability. Cardena et al. (2000) stated that the issue of test-retest reliability is complex for a scale that evaluates the reactions to a stressful event (p. 726). In addition, the authors claimed that the correlation in their test-rest study was 69%. These are the same authors who developed the SASRQ. Therefore, the results of the study suggest that

the HRQ may be a reliable instrument for measuring the impact of having an infant diagnosed with a hemangioma as a stressful event, and as a possible predictor of ASD or PTSD symptoms (Rozell-Shannon, 2007). Further study was suggested using a larger sample. This descriptive study serves as that larger sample.

Classen et al. (n.d.) indicated that the diagnosis of ASD includes three dissociative symptoms experienced shortly after a traumatic event. In order for an individual to have a clinical diagnosis for suffering from ASD, the individual must exhibit at least three dissociate symptoms along with at least one intrusion, avoidance, and hyper arousal symptom (p. 620). Additionally, these symptoms must persist for 2 to 28 days beyond the traumatic event. The mothers that completed the self-reporting HRQ in the pilot study were 1 to 3 years post diagnosis of the hemangioma. Clearly any indication of acute stress beyond this point would be symptomatic for PTSD. This extended period of time, 1 to 4 years post diagnosis of the hemangiomas, was selected to determine if symptoms associated with the traumatic event still persisted, even though some of the lesions were treated and were partially, if not completely resolved. Acute stress disorder (ASD) according to Bryant et al. (1998) describes posttraumatic stress reactions that occur between two days and four weeks following a trauma (p. 215). By extending the period of time from the date of diagnosis up to 4 years, the persistence of any ASD or PTSD symptoms would present a strong argument in favor of the diagnosis of an infant with a hemangioma as being a traumatic event with ASD or PTSD symptoms.

### *Impact of Event Scale – Revised (IES-R)*

The second instrument, IES-R, is a self-report measure designed to assess current subjective distress for any specific life event (Weiss & Marmar, 1997, pg. 1). It consists of 22 questions in a Likert-type scale in which respondents select the answer that best describes how distressing it has been during the week following the traumatic event. In this study, the traumatic event refers to the diagnosis of the hemangioma. Participants in this study were asked to rate each item on the following scale: 1 = *not at all*, 2 = *a little bit*, 3 = *moderately*, 4 = *quite a bit*, and 5 = *extremely*.

This descriptive survey based design describes and explores the reported levels of stress in the infant's mother. According to the authors of the IES-R (Weiss and Marmar, 1997), the IES-R was developed to parallel the criteria for PTSD because the original IES instrument did not include this parallel. It was selected for use with the HRQ in order to establish first, that the diagnosis of an infant with a hemangioma is a traumatic event, and second, to correlate to the symptoms of the HRQ which are also measured with the IES-R.

In their report of four different population samples, Weiss and Marmar (1997) reported that “the internal consistency of the IES-R subscales was found to be very high, with intrusion alphas ranging from .87 to .92 (p. 1). The IES-R is an updated version of the IES but it has 7 additional items that include hyperarousal symptoms such as anger, irritability, heightened startle response, difficulty concentrating, hypervigilance, and reexperiencing (Weiss and Marmar, 1997). This correlates the IES-R closely to the SASRQ, and now to the HRQ. The SASRQ measures symptoms that can occur immediately following a traumatic event, during the month of the event, or even during

the month following the event (Bryant et al., 1998, pp. 215-216). The authors also reported that their test-retest data yielded the following correlation co-efficient: Sample 1 ( $n = 429$ ) the subscales: intrusion = .57, avoidance = .51, hyperarousal = .59, and Sample 2 ( $n = 197$ ) the subscales: = .94, avoidance = .89, and hyperarousal = .95. The authors stated that the shorter timeframe between assessments, as well as the impact of the traumatic event for Sample 2, contributed to the higher coefficients for Sample 2 (p. 2).

The reliability of the IES-R has been reported by Weiss and Marmar (1997) as having a very high internal consistency, with intrusion alphas ranging from .87 to .92, avoidance alphas ranging from .84 to .86 and hyperarousal alphas ranging from .79 to .80 (p. 1).

### Data Collection Methodology

Once IRB approval was received for the study, approval number 11-30-07-0320781, an announcement was posted on the Vascular Birthmarks Foundation's Web sites ([www.birthmark.org](http://www.birthmark.org)) and the MSN Vascular Birthmarks Support Group web site at (<http://groups.msn.com/vascularbirthmarksupport/>). All of the participants were volunteers. The participants went to a link to the SurveyMonkey web site where they completed the surveys, consent, acknowledgement, and participant forms. All respondents were assigned a sequential number to maintain anonymity. All data is maintained on the researcher's desk top and no information linking the responses to the respondents will be shared with anyone. All survey responses and data analysis results that are maintained on the SurveyMonkey web site will only be accessed by the researcher.

### Data Analysis

All of the survey responses were initially analyzed in the SurveyMonkey web site collection and analysis software. Those results were entered into SPSS for descriptive statistics analysis. SPSS, according to Green and Salkind (2005) is a simple and user friendly statistical software package that can analyze data, calculate basic summaries, prepare charts, evaluate distribution function probabilities, perform simulations, and compare means and proportions. Likert-type scoring was used. The descriptive statistics summarizes frequencies and percentages of the results of each of the two instruments. Analysis focused on exploring and describing the levels of stress of the mothers of the infants diagnosed with a hemangioma and whether or not they perceived the event as traumatic. Score distributions, percentages, and other descriptive statistics were computed for each scale and subscale. Relative frequency graphs were generated to present the distribution of the data. Statistical analysis includes exploring the results of the HRQ and the IES-R surveys.

Both of the survey instruments, consent form, acknowledgement, web site announcement, and participant qualification form are included in the Appendixes section (see Appendix .A, C, F, G, H, and I).

With regards to the qualitative analysis portion of the study, the process of data analysis includes open coding, where categories were formed that includes key terms used by the mothers to describe the disturbing event associated with the diagnosis of the hemangioma. The two primary categories are positive events and negative events. Within each of these two categories, subcategories were formed where similar descriptors were

combined and reported. These descriptors form the qualitative data sets. A table is included in this study that summarizes the categories and subcategories. According to Rubin and Rubin (2005) part of the analysis of a qualitative component of a study involves “combining data units on the same topic” (p. 203). The authors further stated that “To figure out what a specific concept means you look at all the data units where that concept is discussed and then bring together in one file the separate definitions, examples, and refinements” (p. 203). During the analysis of what the mothers indicated was the most profound event since the diagnosis, the data units used were examined and described. The frequency of similar terms used by the mothers to report the most profound event that has occurred since the diagnosis of the hemangioma were combined and reported as percentages. Synonyms were weighted equally. For example, responses by mothers who used words such as *frightened*, *scared*, or *alarmed* were combined. On the other hand, if antonyms were used, such as *reassured*, *comforted*, or *peaceful*, then they were reported as opposing comments. Frequency charts and graphs are used to describe the results of the study.

### Summary

A mixed methods descriptive survey study design has been used with this study. The study design includes two-self administered questionnaires, the Hemangioma Reaction Questionnaire (HRQ) and the Impact of Event Scale-Revised (IES-R). One of the questionnaires, the HRQ, included, in addition to the quantitative questions, a personal narrative question for participants to describe (qualitative) the most profound event that occurred following the diagnosis of the hemangioma. This study focused on

describing and exploring the self-reported levels of stress of mothers of infants diagnosed with a hemangioma and reports whether or not they perceive the diagnosis as a traumatic event. A qualitative research design analysis is included in this study because of the one open-ended question in the HRQ. This required exploration and identification of similar data themes.

The participants were directed to a link on the SurveyMonkey web site where they completed the surveys, consent, acknowledgement, and participant forms. No names were collected and all respondents were assigned a number to maintain anonymity. All data is being maintained on the researcher's desk top and no information linking the responses to the respondents was or will be shared with anyone. All survey responses and data analysis results that are maintained on the SurveyMonkey web site were only accessed by the researcher. Data was entered into SPSS from SurveyMonkey and analyzed for descriptive statistics. Finally, triangulation of the quantitative and qualitative data was performed to demonstrate similarity and convergence of findings.



## CHAPTER 4:

### RESULTS

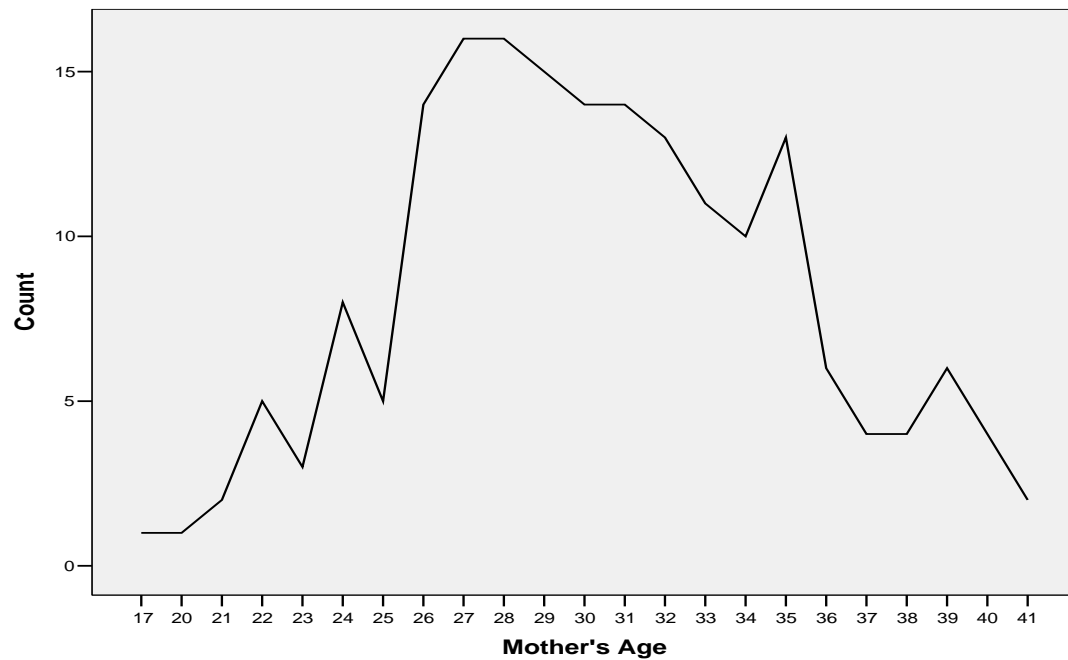
The purpose of this mixed methods descriptive survey study is to describe and explore the reported levels of stress of mothers of infants diagnosed with a hemangioma and to report how these mothers perceived the diagnosis. The hypothesis being tested is that the diagnosis of an infant with a hemangioma is stressful and is perceived as a stressful event for the family, particularly the mother of the infant. Two self-report instruments were used in this study. The HRQ, which is the first instrument used specifically to measure the stress levels of mothers of infants diagnosed with a hemangioma, and the IES-R, an existing instrument, which was used to establish whether or not the participants perceived the diagnosis as a traumatic event. There is one question included in the HRQ that required a qualitative analysis and the remaining data from both instruments relied on quantitative statistical analysis. Both instruments are included in the appendix.

Participants were recruited from the leading hemangioma Web sites and were sent, by means of a link, to the SurveyMonkey web site where they completed the two survey instruments and the required consent, acknowledgement, and participants' forms. All respondents were volunteers. Data from the SurveyMonkey summaries were entered into SPSS. Descriptive statistics and frequency analysis were conducted and tables and graphs were prepared from the data and subscale summaries.

Chapter 4 details the findings of this descriptive study. Participant demographics will be presented first and then the three research questions will be presented with the findings associated with each research question in sequential order.

### Participant Demographics

The proposed convenience sample for this study was identified as 169 mothers of infants that had been diagnosed with a hemangioma within one year prior to completing the surveys, and was based on a population size of 300 (new mothers of infants diagnosed with a hemangioma who joined the various support groups each year). The survey was launched on November 30, 2007 and closed on December 31, 2007. There were 229 mothers who initiated the study. Of that total, 162 completed all study requirements and 148 completed all requirements except for the last instrument (IES). Of the 229 who initiated the study, 220 consented to participate, one declined (and exited the study), and 8 skipped the consent question. Of the 220 who consented, 187 provided demographic information. Figure 1 depicts a single line distribution of the ages of the mothers at the time when the infant with the hemangioma was born. Though the survey instructions indicated that mothers should be between 18 and 40 years of age at the time when the baby was diagnosed with the hemangioma, one mother indicated that she was 17 years old, and one mother indicated that she was 41 years old. Therefore, the age range for the 187 mothers who answered that question was 17 to 41, representing a mean age of 30.34 with a standard deviation of 4.74 years.



*Figure 1. Mother's age range graph*

The 187 mothers who answered the age question also answered the question concerning the country where they live. Over 75% (75.9%) were from the United States and the remaining 24.1% were from numerous regions around the world. Table 4 depicts the countries where the participant's indicated that they live. Over 5% (5.9%) indicated that they live in the Netherlands. The Netherlands has a very large and strong hemangioma support group that explains why that number yielded the highest percentage of respondents outside the United States.

Table 4

*Mother's Country*

Country	Frequency	Percent	Cumulative percent
Australia	7	3.7	3.7
Belgium	2	1.1	4.8
Canada	8	4.3	9.1
China	1	.5	9.6
Egypt	1	.5	10.2
England	2	1.1	11.2
Latvia	1	.5	11.8
Macedonia	1	.5	12.3
Netherlands	11	5.9	18.2
New Zealand	1	.5	18.7
Philippines	1	.5	19.3
Scotland	1	.5	19.8
Sweden	1	.5	20.3
United Kingdom	7	3.7	24.1
USA	142	75.9	100.0
Total	187	100.0	

Of the 187 mothers who answered the questions concerning demographics, over 89% (168) of the mothers (89.8%) indicated that they had never been treated for a stress-related illness, and 58.8% (110) indicated that the baby that was diagnosed with the hemangioma was their first baby.

The following descriptive analysis pertains to the HRQ instrument and is based on the first research question presented in this study.

#### Research Question 1

Using the HRQ instrument, what level of stress was reported by mother's who had an infant diagnosed with a hemangioma?

To answer this question, 162 mothers completed the HRQ, the first disease specific instrument designed to measure the impact to mothers of infants diagnosed with a hemangioma, along with all other required forms. The HRQ is a modification of the SASRQ, an instrument designed to measure dissociative symptoms, reexperiencing of trauma, avoidance, anxiety and hyperarousal, and impairment in functioning (Cardena et al., 2000). According to Cardena et al., the SASRQ instrument could be modified to a specific event. Therefore, the SASRQ was modified to create the HRQ to refer to the specific event (diagnosis of hemangioma on an infant) in order to measure the impact on the infant's mother. The instrument is scored using a Likert-type scale (anchored by 1, *not experienced*, and 6, *very often experienced*) for assessing symptoms. The authors of the SASRQ scored a symptom as present if the respondent marked it as occurring sometimes, often, or very often, which included a response of 4, 5, or 6.

The 30-question Likert-type survey statistics were entered into SPSS software and calculated for individual results and then disaggregated into the nine sub scales that comprise the acute stress reaction. The HRQ, like the SASRQ, is broken down into nine subscales. The first subscale on the SASRQ (Subscale A, traumatic event) is actually the last category on the HRQ. This is the qualitative open-ended question which asks the mother to define the most profound event that has occurred since the diagnosis of the hemangioma. The mothers were also asked to identify how disturbing this event was and for how many days they experienced any of the symptoms indicated in the HRQ. There are nine scales which encompass five subscales for measuring the symptoms of the traumatic event. The first is Subscale B, which comprises five symptoms that reflect a

dissociative diagnosis, Subscale C (reexperiencing of trauma), Subscale D (avoidance), Subscale E (anxiety and hyperarousal); and Subscale F (impairment in functioning). The tables in this section represent the results of each subscale. Some selected individual question results are also included as they are noteworthy. Since the qualitative question, Subscale A, is presented last, the first subscale presented here (B) includes the dissociative symptoms. There are five categories that comprise the dissociative symptoms subscale:

*B. 1.* Subjective sense of numbing, detachment, or absence of emotional responsiveness (includes Questions 20 and 28)

*B. 2.* A reduction in awareness of one's surroundings (includes questions 4 and 24).

*B. 3.* Derealization (includes Questions 3 and 18).

*B. 4.* Depersonalization (includes Questions 3 and 10).

*B. 5.* Dissociative amnesia, ie., inability to recall an important aspect of the trauma (includes Questions 16 and 25).

Subscale C. (Reexperiencing of Trauma) includes Questions 6, 7, 15, 19, 23, and 29). Subscale D. (Avoidance) includes Questions 5, 11, 14, 17, 22, and 30. Subscale E. (Anxiety and Hyperarousal) includes questions 1, 2, 8, 12, 21, and 27. The last subscale F. (Impairment in functioning) includes questions 9 and 26. The one qualitative question which is included in the HRQ (compares to subscale A. in the SASRQ) will be explored with the third research question, following the HRQ quantitative and IES-R quantitative results (see Appendix E).

Table 5 is the first table containing data for describing the results of the HRQ survey study. It contains the descriptive results of the first of two questions included in the first dissociative symptom subscale (B. 1). This symptom is identified as *a subjective sense of numbing, detachment, or absence of emotional responsiveness* (questions 20 and 28). Table 5 subscale results for B. 1 show that 36.4%, or more than 1/3 of the mothers indicated that they experienced a subjective sense of numbing, detachment, or absence of emotional responsiveness as a result of having an infant diagnosed with a hemangioma, and 63.6% did not.

Table 5

*B 1 Subscale: Measuring a Subjective Sense of Numbing, Detachment, or Absence of Emotional Responsiveness.*

		Frequency	Percent	Cumulative percent
Valid	2	58	35.8	35.8
	3	14	8.6	44.4
	4	10	6.2	50.6
	5	12	7.4	58.0
	6	9	5.6	63.6
	7	12	7.4	71.0
	8	22	13.6	84.6
	9	12	7.4	92.0
	10	6	3.7	95.7
	11	5	3.1	98.8
	12	2	1.2	100.0
Total		162	100.0	

Table 6 combines the results of Q. 4 and Q. 24 for subscale B. 2 for dissociative symptoms. Table 6 subscale results show that 22.2% of the mothers indicated that they had a reduction in awareness of one's surroundings as a result of having an infant diagnosed with a hemangioma, and 77.8% did not.

Table 6

*B. 2 Subscale. It measures dissociative symptoms.*

		Frequency	Percent	Cumulative percent
Valid	2	58	35.8	35.8
	3	19	11.7	47.5
	4	16	9.9	57.4
	5	17	10.5	67.9
	6	16	9.9	77.8
	7	15	9.3	87.0
	8	11	6.8	93.8
	9	1	.6	94.4
	10	8	4.9	99.4
	11	1	.6	100.0
	Total	162	100.0	

Tables 7 and 8 include the results of the dissociative symptoms B. 3 subscale for each individual question. The results shown in Table 7 indicate that 43.8% of the respondents to Q 3. experienced symptoms (responses 4, 5, or 6) and 56.2% did not (responses 1, 2, or 3). This is the first question with fairly comparable results. It shows that a high percentage of the mothers indicated that they experienced some sense of timelessness since their infant was diagnosed with a hemangioma.



Table 7

*Question. 3: I felt a sense of timelessness*

		Frequency	Percent	Cumulative percent
Valid	1	55	34.0	34.0
	2	13	8.0	42.0
	3	23	14.2	56.2
	4	28	17.3	73.5
	5	25	15.4	88.9
	6	18	11.1	100.0
	Total	162	100.0	

Table 8 indicates that 25.3% of the respondents to Q. 18 experienced symptoms (responses 4, 5, or 6) and 74.7% did not (responses 1, 2, or 3) which indicates that a quarter of the mothers indicated that things looked different to them from how they knew they really looked as a result of the diagnosis of the hemangioma.

Table 8

*Question. 18: Things I saw looked different to me from how I know they really looked*

		Frequency	Percent	Cumulative percent
Valid	1	90	55.6	55.6
	2	17	10.5	66.0
	3	14	8.6	74.7
	4	28	17.3	92.0
	5	8	4.9	96.9
	6	5	3.1	100.0
	Total	162	100.0	

Table 9 combines the results of Q. 3 and Q. 18 for subscale B. 3. Table 9 subscale results show that 31.4% of the mothers indicated that they experienced derealization as a result of having an infant diagnosed with a hemangioma, and 68.5% did not. It is worth noting that Q. 3 results (I felt a sense of timelessness) were fairly even with 43.8% did and 56.2% did not experience timelessness but when combined with Question 18 (Things I saw looked different to me from how I know they really looked) were 3:1 (74.7% did and 25.3% did not). When combined, this brought the subscale results to 31.4% did and 68.5% did not. This is why it is important to report the results individually as well as using the combined subscales so that an accurate portrayal of the data is presented and discrepancies are noted where there are considerable variances.

Table 9

*B. 3 Subscale. It measures derealization.*

		Frequency	Percent	Cumulative percent
Valid	2	46	28.4	28.4
	3	5	3.1	31.5
	4	14	8.6	40.1
	5	24	14.8	54.9
	6	22	13.6	68.5
	7	20	12.3	80.9
	8	8	4.9	85.8
	9	13	8.0	93.8
	10	4	2.5	96.3
	11	5	3.1	99.4
	12	1	.6	100.0
Total		162	100.0	

Table 10 combines the results of Q. 10 and Q. 13 for subscale B. 4. Table 10 subscale results show that 26.6% of the mothers indicated that they experienced depersonalization as a result of having an infant diagnosed with a hemangioma, and 73.2 % did not. The subscale for these two questions is more realistic than some of the other combinations where there are extremes. Therefore, this subscale could be fairly reflective of this dissociative symptom, though no cause and effect relationship can be concluded from this descriptive and exploratory study. This study does not correlate data but rather describes the results of the statistical analysis of the survey responses.

Table 10

*B. 4 Subscale. It measures depersonalization.*

		Frequency	Percent	Cumulative percent
Valid	2	67	41.4	41.4
	3	12	7.4	48.8
	4	16	9.9	58.6
	5	11	6.8	65.4
	6	13	8.0	73.5
	7	13	8.0	81.5
	8	12	7.4	88.9
	9	4	2.5	91.4
	10	7	4.3	95.7
	11	4	2.5	98.1
	12	3	1.9	100.0
Total		162	100.0	

Table 11 combines the results of Q. 16 and Q. 25 for subscale B. 5. This table shows that only 3.7% of the mothers indicated that they experienced dissociative amnesia, the inability to recall an important aspect of the trauma, and 96.3% did not.

Table 11

*B. 5 Subscale. It measures dissociative amnesia or the ability to recall an important aspect of the trauma.*

		Frequency	Percent	Cumulative percent
Valid	2	100	61.7	61.7
	3	24	14.8	76.5
	4	14	8.6	85.2
	5	9	5.6	90.7
	6	9	5.6	96.3
	7	4	2.5	98.8
	9	1	.6	99.4
	10	1	.6	100.0
Total		162	100.0	

Subscale C comprises the second grouping of questions for measuring traumatic symptoms. This subscale indicates that the traumatic event is persistently reexperienced, and includes Q. 6, Q. 7, Q. 15, Q. 19, Q. 23, and Q. 29.

Table 12 subscale results show that 20.2% of the mothers indicated that the traumatic event is persistently reexperienced, and 79.7% did not. For this subscale, there are six questions and the range of symptoms experienced by the mothers is as low as 13.0% for Q. 29 and as high as 42.6% for Q. 7. The results shown in Table 12 indicates a large distribution of responses. This prompts the reader to question whether a subscale that indicates 20.2% of the mothers had symptoms for this subscale but 79.7%

did not is reflective, since in one of the questions that comprise this subscale, 42.6% experienced symptoms. A weighted system that will equalize the results should be considered.

Table 12

*Subscale C. It measures if the traumatic event is persistently reexperienced*

		Frequency	Percent	Cumulative percent
Valid	6	35	21.6	21.6
	7	10	6.2	27.8
	8	10	6.2	34.0
	9	6	3.7	37.7
	10	11	6.8	44.4
	11	11	6.8	51.2
	12	8	4.9	56.2
	13	4	2.5	58.6
	14	7	4.3	63.0
	15	9	5.6	68.5
	16	7	4.3	72.8
	17	6	3.7	76.5
	18	5	3.1	79.6
	19	8	4.9	84.6
	20	1	.6	85.2
	21	2	1.2	86.4
	22	4	2.5	88.9
	23	5	3.1	92.0
	24	2	1.2	93.2
	25	1	.6	93.8
	26	3	1.9	95.7
	27	2	1.2	96.9
	28	1	.6	97.5
	29	1	.6	98.1
	30	1	.6	98.8
	31	1	.6	99.4
	33	1	.6	100.0
	Total	162	100.0	

Subscale D comprises questions for measuring marked avoidance of stimuli arouse recollections of the trauma, and includes Q. 5, Q. 11, Q. 14, Q. 17, Q. 22, and Q. 30. The results shown in Table 13 for Q. 5 indicate that 54.9% of the mothers experienced symptoms and 45.1% did not. This is the first question in this entire study where the symptoms experienced were a greater percentage than those not experienced.

Table 13

*Question. 5: I tried to avoid feelings about the hemangioma*

		Frequency	Percent	Cumulative percent
Valid	1	33	20.4	20.4
	2	25	15.4	35.8
	3	15	9.3	45.1
	4	42	25.9	71.0
	5	31	19.1	90.1
	6	16	9.9	100.0
	Total	162	100.0	

The subscale results for category D represents *marked avoidance of stimuli arouse recollections of the trauma*, and includes Q. 5, Q. 11, Q. 14, Q. 17, Q. 22, and Q. 30. Half of the questions that comprise the results shown in Table 33 for subscale D had fairly high and notable individual scores (Q. 5, Q. 14, and Q. 17). All three questions claimed that the mothers tried to avoid feelings, conversations, or thoughts about the hemangioma. Alternatively, the results of Q. 11 are much lower, and concerns avoiding *activities* that remind the mother of the hemangioma. The two lowest scores represent *avoiding places or people who reminded the mothers of the hemangioma*. Thus, one might surmise that a good percentage of mothers wanted to avoid feelings, conversations, or thoughts about the hemangioma but not as many wanted to avoid activities, places, or people that reminded them of the hemangioma. Table 14 for subscale D indicates that 29% of the mothers experienced symptoms and 70.9% did not, but because of the nature of subscales it does not describe the results of the individual questions. In Q. 5, more mothers had symptoms than those who did not (54.9% did and 45.1% did not). Two of the six questions had such low symptom results for the mothers (Q. 22 had 8% and Q. 30 had 11%) that when all of the data is combined it diminishes the important results of Q. 5 which indicates that the majority of the mothers tried to avoid feelings about the hemangioma. This information could be significant for physicians so that they can focus some of the informational dialogue on explaining to the mothers that they should openly discuss their feelings about the hemangioma. These results could also be used to refer mothers to support groups or mental health counseling, if necessary. While the subscales are used to determine stress and trauma symptoms, the results from each individual

question should also be considered when determining if the individual completing the survey is in need of counseling.

Table 14

*Subscale D. It measures marked avoidance of stimuli that arouse recollections of the trauma*

		Frequency	Percent	Cumulative percent
Valid	6	14	8.6	8.6
	7	9	5.6	14.2
	8	11	6.8	21.0
	9	15	9.3	30.2
	10	8	4.9	35.2
	11	6	3.7	38.9
	12	10	6.2	45.1
	13	10	6.2	51.2
	14	8	4.9	56.2
	15	6	3.7	59.9
	16	8	4.9	64.8
	17	8	4.9	69.8
	18	2	1.2	71.0
	19	8	4.9	75.9
	20	4	2.5	78.4
	21	5	3.1	81.5
	22	4	2.5	84.0
	23	7	4.3	88.3
	24	4	2.5	90.7
	25	7	4.3	95.1
	26	4	2.5	97.5
	28	1	.6	98.1
	29	1	.6	98.8
	31	1	.6	99.4
	35	1	.6	100.0
	Total	162	100.0	



Of the nine subscales, the next category, subscale E, presents the two highest results for mothers who indicated that they experienced symptoms (Q. 1 with 59.3%, and Q. 2 with 63%). What's even more interesting is that this is the only subscale where the combined results (53%) indicate that more mothers indicated a symptom is present (a symptom is considered present if the respondent marked it as occurring sometimes, often, or very often, which includes a response of 4, 5, or 6.) than those who did not (47%). The reason that this may be more representative of the individual scores is because five of the questions had symptoms results that ranged between 43% and 63% with only one question (Q. 8) having a low score of 17.3%.

Table 15 shows the results for subscale E which indicate that 53% of the mothers experienced marked symptoms of anxiety or increased arousal and 47% did not.

Table 15

*Subscale E: It measures marked symptoms of anxiety or increased arousal*

		Frequency	Percent	Cumulative percent
Valid	6	17	10.5	10.5
	7	3	1.9	12.3
	8	4	2.5	14.8
	9	2	1.2	16.0
	10	6	3.7	19.8
	11	3	1.9	21.6
	12	3	1.9	23.5
	13	9	5.6	29.0
	14	6	3.7	32.7
	15	6	3.7	36.4
	16	7	4.3	40.7
	17	4	2.5	43.2
	18	6	3.7	46.9
	19	6	3.7	50.6
	20	5	3.1	53.7
	21	7	4.3	58.0
	22	8	4.9	63.0
	23	9	5.6	68.5
	24	11	6.8	75.3
	25	8	4.9	80.2
	26	4	2.5	82.7
	27	5	3.1	85.8
	28	8	4.9	90.7
	29	5	3.1	93.8
	30	1	.6	94.4
	31	1	.6	95.1
	32	2	1.2	96.3
	33	4	2.5	98.8
	34	1	.6	99.4
	35	1	.6	100.0
Total		162	100.0	

Subscale F is the last of the nine subscales. It indicates a measure for impairment of functioning and includes the individual results for Q. 9 and Q. 26. The results shown in Table 16 indicate that 49.4% of the mothers experienced symptoms and 50.6% did not. This is the question which was closest in response between “did and did not experience symptoms”. Therefore, nearly half of the mothers indicated that they experienced thoughts of the hemangioma that made it difficult to perform work or other routine tasks that they needed to do.

Table 16

*Question. 9: Thoughts of the hemangioma made it difficult for me to perform work or other things I needed to do*

		Frequency	Percent	Cumulative percent
Valid	1	41	25.3	25.3
	2	24	14.8	40.1
	3	17	10.5	50.6
	4	36	22.2	72.8
	5	21	13.0	85.8
	6	23	14.2	100.0
	Total	162	100.0	

The results shown in Table 17 indicate that 32% of the mothers experienced symptoms and 68% did not. This is interesting since the previous question was fairly equal with regards to difficulties performing tasks but it was not as equal when it came to causing problems with relationships.

Table 17

*Question. 26: The hemangioma caused problems in my relationships with other people*

		Frequency	Percent	Cumulative percent
Valid	1	73	45.1	45.1
	2	22	13.6	58.6
	3	15	9.3	67.9
	4	33	20.4	88.3
	5	13	8.0	96.3
	6	6	3.7	100.0
	Total	162	100.0	

Table 18 combines the results for subscale F which includes Q. 9 and Q. 26. The results shown in Table 18 indicate that 39.6% of the mothers experienced symptoms and 60.4% did not. Unfortunately, once again combining question results into a subscale does not depict important individual question results. For example, Q. 9 had an almost even division between the number of mothers who experienced symptoms and those who did not (49.4% did and 50.6% did not) but this is not reflected in the subscale results which show that only 39.6% of the mothers experienced symptoms for this subscale.

Table 18

*Subscale F. It measures impairment in functioning*

		Frequency	Percent	Cumulative percent
Valid	2	37	22.8	22.8
	3	12	7.4	30.2
	4	11	6.8	37.0
	5	14	8.6	45.7
	6	24	14.8	60.5
	7	18	11.1	71.6
	8	15	9.3	80.9
	9	12	7.4	88.3
	10	12	7.4	95.7
	11	4	2.5	98.1
	12	3	1.9	100.0
Total		162	100.0	

This concludes the descriptive presentation of the data collected from the HRQ study. In summary, while the subscales do not reflect individual question results, which can be important in assessing if a mother needs counseling, they have been used extensively with the SASRQ instrument for assessing the presence of stressful and traumatic symptoms. The results for subscale E indicated that more than half of the mothers (53%) experienced marked symptoms of anxiety or increased arousal (hyperarousal) which was the highest response rate of all three subscales.

Lastly, the following two tables include a description of the total number of days that mothers indicated that they experienced the symptoms expressed in the HRQ. Tables 19 and 20 show the results of the descriptive statistical analysis of the number of days indicated by the mothers. The range include 0 = *none*, 1 = *1 day*, 2 = *2 days*, 3 = *3 days*, 4

= 4 days, and 5 = 5 or more days. Table 19 indicates the mean or arithmetic average number of days, the mode indicates the day or days most often selected by mothers. Table 19 indicates that the average number of days that the mothers indicated the symptoms persisted was 3.37 and the most frequently selected number of days was 5.

Table 19

*Number of Days Symptoms Persisted*

N	Valid	162
	Missing	0
Mean		3.37
Median		5.00
Mode		5
Std. Deviation		2.021
Range		5

Table 20 depicts the frequency of distribution in the total number of days that the mothers indicated that they experienced symptoms. As previously stated, the mode for the number of days was 5, indicating that 5 or more days was the most frequently selected choice for that question (54.3%). The second most frequently selected choice was not 4 or 3 days, as may be surmised, but rather 0 days. The frequency for 0 days was 29 for a total of 17.9% of all responses for that question. The remaining choices were fairly evenly distributed.

Table 20

*Distribution of Days Symptoms Persisted*

		Frequency	Percent	Cumulative percent
Valid	0	29	17.9	17.9
	1	12	7.4	25.3
	2	12	7.4	32.7
	3	14	8.6	41.4
	4	7	4.3	45.7
	5	88	54.3	100.0
	Total	162	100.0	

The importance of all of the previously stated data revolves around the notion that the HRQ is intended to measure acute stress disorder symptoms, similar to the SASRQ. According to Cardena et al. (2000) it was noted that high scores on the SASRQ (for which the HRQ was modified) may be helpful for determining if the respondent needs mental health treatment and may be useful in tracking response to that treatment.

However, they also noted that in the management of patients, further research is needed to evaluate the scores on the SASRQ (the HRQ for this study) and their clinical significance. However, this is the first instrument designed to measure the impact on the caregiver (mother for this study), therefore, it closes a gap where an instrument has been needed for decades to measure the impact of hemangiomas on the family system.

The first research question asked what level of stress is reported by mother's who have an infant who is diagnosed with a hemangioma who completed the HRQ. This study describes the percentage of mothers who indicated that they did or did not experience a certain level of symptoms as a result of the diagnosis of the hemangioma. The highest percentage of symptoms for a subscale that was indicated by the mothers was for Subscale E (anxiety and hyperarousal) with 53% indicating that they experienced these symptoms as a result of having an infant diagnosed with a hemangioma. However, according to Cardena et al. (2000) for the diagnosis of acute stress disorder to be made, there needs to be a direct exposure or witnessing to a trauma involving intense negative emotions, at least three dissociative symptoms (numbering, depersonalization, derealization, being in a daze, and amnesia), reexperiencing or intrusion of the traumatic event, marked avoidance of stimuli related to trauma, marked anxiety or hyperarousal, and clinically significant distress or impairment. These symptoms must last for a minimum of 2 days and a maximum of 4 weeks, and they must not be directly caused by a psychoactive substance or a general medical condition (p. 721). Based on the data collected from this study, no determination can be made as to whether or not the study participants met the criteria for acute stress disorder. However, the first question can be



answered as stated for the alternative hypothesis which is that there is a reported level of stress symptoms for mothers who completed the HRQ. The highest of these levels was reported for Subscale E (anxiety and hyperarousal) with 53% indicating that they experienced these symptoms as a result of having an infant diagnosed with a hemangioma. The mothers reported measurable levels of stress for all nine subscales, one was very low (Subscale B5 = 3.7%) and one was very high (53.0%). The remaining seven subscales ranged from 20% to 39%. Additionally, 54.3% of the mothers indicated that symptoms persisted for 5 or more days.

#### Research Question 2

Using the IES-R instrument, the second research question asks what impact of event measure is reported by mothers of infants diagnosed with a hemangioma and who complete this survey?

To answer this question, 148 mothers completed the IES-R survey. IES-R is a self-report measure designed to assess current subjective distress for any specific life event (Weiss & Marmar, 1997, pg. 1). It consists of 22 questions in a Likert-type scale in which respondents select the answer that best describes how distressing they felt during the week following the traumatic event. In this study, the traumatic event refers to the diagnosis of the hemangioma. Participants in this study were asked to rate each item on the following scale: 1 = *not at all*, 2 = *a little bit*, 3 = *moderately*, 4 = *quite a bit*, and 5 = *extremely*.

According to the authors of the IES-R (Weiss and Marmar, 1997), the survey was developed to parallel the criteria for PTSD because the original IES instrument did not

include this parallel. The EIS-R was selected for use with the HRQ in order to establish that the diagnosis of an infant with a hemangioma is a stressful and traumatic event, and to show a relationship to the symptoms of the HRQ.

The IES-R subscales include *Intrusion* (questions 1, 2, 3, 6, 9, 14, 16 and 20), *Avoidance* (questions 5, 7, 8, 11, 12, 13, 17, and 22), and *Hyperarousal* (4, 10, 15, 18, 19, and 21). The IES-R scales are typically based on a score range of 0 to 4 but SurveyMonkey does not possess the ability to use a score of 0, so the scale was adjusted to a range of 1 to 5, instead of 0 to 4. The timeframe for the use of this instrument has been modified from the standard 7 days to up to one year. No reliability or validity exists for this new, non-standard measure. For this study only, the percentage of responses for each category are presented. The three subscales represent the combined scores for 148 respondents. The use of this instrument is not to determine that the diagnosis of a infant with a hemangioma is a stressful and traumatic event but to describe and report the perceptions of the mothers who completed this survey. It is also used to explore any similarities between the scores in the IES-R and those from the HRQ. Tables 21, 22, and 23 include the descriptive statistics for the three subscales. *Intrusion* is the first subscale to be described and is comprised of eight questions that comprise this subscale (1, 2, 3, 6, 9, 14, 16, and 20). The IES-R appears in Appendix D and the scoring instructions appears in Appendix E. Since there are 8 questions in this subscale and a range of answers from 1 to 5, the total possible range is 8 to 40. The EIS-R scoring sheet does not suggest a cut off score, like the SASRQ or HRQ which makes the results difficult to interpret because

without a cut off score for indicating the presence of symptoms, the scores simply represent the percentage of responses to each answer.

The *Intrusion* subscale results presented in Table 21 indicate that for choice 1 (not at all) 2.7% of the mothers selected this answer; for answer 2 (a little bit) 42% of the mothers selected this answer; for answer 3 (moderately) 39.2% of the mothers selected this answer; for answer 4 (quite a bit) 14.9% of the mothers selected this answer; and for answer 5 (extremely) 1.4% of the mothers selected this answer. *Intrusion* refers to the degree of being overwhelmed by thoughts and feelings about the traumatic event. (Janssens et al. 2006). In this case, the intrusion would refer to the diagnosis of the hemangioma.

Table 21

*Intrusion Subscale*

		Frequency	Percent	Cumulative percent
Valid	6	3	2.0	2.0
	8	1	.7	2.7
	9	6	4.1	6.8
	10	6	4.1	10.8
	11	7	4.7	15.5
	12	11	7.4	23.0
	13	7	4.7	27.7
	14	6	4.1	31.8
	15	14	9.5	41.2
	16	5	3.4	44.6
	17	7	4.7	49.3
	18	10	6.8	56.1
	19	9	6.1	62.2
	20	8	5.4	67.6
	21	11	7.4	75.0
	22	7	4.7	79.7
	23	1	.7	80.4
	24	5	3.4	83.8
	25	3	2.0	85.8
	26	7	4.7	90.5
	27	3	2.0	92.6
	28	5	3.4	95.9
	29	2	1.4	97.3
	30	1	.7	98.0
	32	1	.7	98.6
	33	1	.7	99.3
	34	1	.7	100.0
Total		148	100.0	

The *Avoidance* subscale results presented in Table 22 indicate that for choice 1 (not at all) 10.1% of the mothers selected this answer; for answer 2 (a little bit) 51.4% of the mothers selected this answer; for answer 3 (moderately) 27.2% of the mothers selected this answer; for answer 4 (quite a bit) 10.9% of the mothers selected this answer; and for answer 5 (extremely) 0.7% of the mothers selected this answer. *Avoidance* refers to the tendency to keep off thoughts of the trauma (Janssens et al. 2006). In this case, avoidance would refer to keeping off thoughts of the hemangioma.

Table 22

*Avoidance Subscale*

		Frequency	Percent	Cumulative percent
Valid	8	15	10.1	10.1
	9	13	8.8	18.9
	10	12	8.1	27.0
	11	11	7.4	34.5
	12	9	6.1	40.5
	13	11	7.4	48.0
	14	6	4.1	52.0
	15	10	6.8	58.8
	16	4	2.7	61.5
	17	5	3.4	64.9
	18	5	3.4	68.2
	19	6	4.1	72.3
	20	8	5.4	77.7
	21	5	3.4	81.1
	22	5	3.4	84.5
	23	5	3.4	87.8
	24	1	.7	88.5
	25	1	.7	89.2
	26	4	2.7	91.9
	27	3	2.0	93.9
	28	2	1.4	95.3
	29	1	.7	95.9
	30	1	.7	96.6
	31	1	.7	97.3
	32	3	2.0	99.3
	35	1	.7	100.0
	Total	148	100.0	

The *Hyperarousal* subscale results presented in Table 23 indicate that for choice 1 (not at all) 12.8% of the mothers selected this answer; for answer 2 (a little bit) 39.2% of the mothers selected this answer; for answer 3 (moderately) 32.4% of the mothers selected this answer; for answer 4 (quite a bit) 12.2% of the mothers selected this answer; and for answer 5 (extremely) 3.5% of the mothers selected this answer. Hyperarousal refers to anxiety, sleep, or concentration difficulties that occur as a result of a trauma (APA,1994). In this case, hyperarousal would refer to anxiety, sleep, or concentration difficulties related to the hemangioma.

Table 23

*Hyperarousal*

		Frequency	Percent	Cumulative percent
Valid	6	19	12.8	12.8
	7	13	8.8	21.6
	8	13	8.8	30.4
	9	6	4.1	34.5
	10	12	8.1	42.6
	11	7	4.7	47.3
	12	7	4.7	52.0
	13	3	2.0	54.1
	14	12	8.1	62.2
	15	9	6.1	68.2
	16	8	5.4	73.6
	17	8	5.4	79.1
	18	8	5.4	84.5
	19	3	2.0	86.5
	20	7	4.7	91.2
	21	1	.7	91.9
	22	6	4.1	95.9
	24	1	.7	96.6
	25	1	.7	97.3
	26	1	.7	98.0
	27	1	.7	98.6
	28	2	1.4	100.0
	Total	148	100.0	

Comparing the results from the HRQ and IES-R is difficult since the creators of the IES-R do not recommend anchoring any particular score to mean the presence of a symptom. However, for the purpose of this study only, the three responses for the HRQ which are considered present for a symptom: (4) *sometimes experienced*, (5) *often*



*experienced*, and (6) *very often experienced*, are likened to the responses in the IES-R for presence of a symptom and include: (3) *moderately*, (4) *quite a bit* and (5) *extremely*. The results shown in Table 24 indicate that hyperarousal is the symptom that shows the closest relationship for the two surveys. For the HRQ, mothers who indicated that they experienced hyperarousal symptoms for subscale E (marked symptoms of anxiety or increased arousal) was 53% as compared to 48.1% for the same symptom in the IES-R survey subscale for hyperarousal. While this study is strictly descriptive, it is interesting that both subscales for the same symptom were fairly close in finding the same percentage of mothers experiencing that symptom. The avoidance symptom was also fairly close with 29% from the HRQ indicating they did experience those symptoms as compared with 38.8% from the IES-R. The HRQ for intrusion yielded 20.2% for Subscale C (the traumatic event is persistently reexperienced) and 55.5% for the IES-R survey instrument. This could be explained by the fact that the questions pertaining to intrusion in both instruments are not as close in description as the questions pertaining to avoidance and hyperarousal for both surveys.

Table 24 presents a comparison of the results of like categories for symptoms that mothers indicated that they did experience:

Table 24

*Comparison of HRQ to IES-R*

EIS-R	% indicated symptoms	HRQ	% indicated symptoms
Intrusion	55.5%	Subscale C	20.2%
Avoidance	38.8%	Subscale D	29.0%
Hyperarousal	48.1%	Subscale E	53.0%

Therefore, the second research question which asked what impact of event measure is reported by mothers of infants diagnosed with a hemangioma and who complete this survey is also answered for the alternative because the mothers did report a measure of impact from the event for all three subscales.

## Research Question 3

For the qualitative question included in the HRQ, what common themes have emerged from the question which asks the mothers to describe the most profound event that has occurred since the hemangioma was diagnosed?

This question is answered from the mothers' response to the one qualitative question included in the HRQ. The mothers were asked to briefly describe the most profound event that occurred since their baby was diagnosed with a hemangioma. In the HRQ, the word *profound* was selected because it is a neutral word that can have a

negative or positive response. In the SASRQ, the word *disturbing* is used which could be interpreted as a negative response. The responses were all put into one of four like categories and then entered into SPSS for descriptive statistical analysis. Table 25 details a key that was used for the analysis.

Table 25

*Qualitative Question Key*

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P	=	Positive response. Mother indicated that the profound event was a positive experience.
N	=	Negative response. Mother indicated that the profound event was a negative experience.
O	=	None or no response. Mother indicated that there was no profound event that occurred since the baby was diagnosed with a hemangioma.
A	=	Neutral response – Mother indicated a response that could be perceived as neutral – neither negative nor positive.

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Table 26 indicates the results of the analysis of the qualitative question which was answered by 162 mothers. The results indicate that 80.9% of the mothers expressed a negative response to the question, only 5.6% indicated a positive response, and an equal amount, 5.6%, was indicated for a neutral response where the answer could be perceived as neither negative nor positive. One example is that of a mother who indicated that the most profound event for her was searching on the Internet. That response could mean that it disturbed her to see photos of infants with hemangiomas when she was searching, or it could mean that it was a positive event because the mother found a support group and

treatment information. The choice of *none or no response* was selected by 8% of the mothers.

Table 26

*Qualitative Question Response Frequency Distribution*

		Frequency	Percent	Cumulative percent
Valid	A	9	5.6	5.6
	N	131	80.9	86.4
	O	13	8.0	94.4
	P	9	5.6	100.0
	Total	162	100.0	

The four categories that were selected for sorting all of the responses from the mothers were broken down into similar responses per category. Table 27 depicts the responses for the *Negative* category since this was the largest (80.9% of the responses). The most common negative comment was *people making rude comments or negative statements*. More than one third (37%) of the mothers gave this response. The next most profound event noted by the mothers (15%) was the surgery or other treatment of the hemangioma. The remaining like categories had similar percentages of responses. The word *rude* was used by 70% of the mothers included in the first category (where 37% of the total responses were included). Clearly having people make a negative or rude comment about the hemangioma is perceived as a profoundly negative experience for many (49) of the 162 mothers. This information may assist physicians or even support group coordinators in preparing to address this matter by providing parents with a coping mechanism so that such events are not perceived so profoundly.

Table 27

*Negative response categories*

Negative Responses = N	Frequency	Percent
People making rude comments or negative statements	49	37%
Surgery/Treatment of the hemangioma	20	15%
People staring at the baby's hemangioma	12	9%
Growth of the hemangioma was startling	12	9%
Worrying about the baby because of the hemangioma	10	8%
Hemangioma bled or ulcerated	7	5%
Crying over the hemangioma	6	5%
Diagnosis of the hemangioma	5	4%
Stress from the hemangioma	4	3%
Doctors not knowing how to diagnose or treat the hemangioma	3	2%
Family problems	2	2%
Could not work on PhD because of so many doctor visits	1	1%
Totals	131	100%

Table 28 depicts *none or no response* (8% of the responses). The mothers who responded to this question simply stated that nothing profound happened or that there was no profound event.

Table 28

*None or no response categories*

None or No Response = O	Frequency	Percent
Nothing profound	8	62%
None	5	38%
Totals	13	100%

Table 29 depicts the *Neutral* (5.6%) response. Mothers who responded to this question noted events that could neither be labeled as positive or negative without knowing what the mother meant. Therefore, responses that fell into this category were neutral. Four mothers indicated treatment was the most profound neutral event and an equal number indicated researching the issue on line was the most profound event.

Table 29

Neutral response

Neutral Responses = A	Frequencies	
Treatment	4	44%
Researched issues on line	4	44%
Not sure	1	12%
Totals	9	100%

Table 30 depicts the positive responses by the mothers when they described the most profound event since the diagnosis of the hemangioma. There was only a 5.6% response for the positive category and the most frequently stated positive event was the treatment or removal of the hemangioma. The second most positive profound event was when the mother noticed the hemangioma was shrinking or going away.

Table 30

*Positive response*

Positive Responses = P	Frequencies	
Treatment or removal of the hemangioma	3	33%
Hemangioma began to shrink or go away	2	23%
Didn't see any new growth	1	11%
Birth of another child without a hemangioma	1	11%
Baby had a birthday	1	11%
Someone actually said our baby was beautiful	1	11%
Totals	9	100%

## Triangulation of Data

In an attempt to triangulate the quantitative results from the two surveys and the qualitative results from the one narrative question in the HRQ, the three themes: *Intrusion*, *Avoidance*, and *Hyperarousal* are used as anchors. Table 31 shows the negative responses of the mothers as they relate to the three themes in this study. Two of the themes were almost even (48% for *Hyperarousal* and 46% for *Intrusion*). The third theme, *Avoidance*, represented only 6% of the responses. One possible explanation that the theme *Avoidance* was so low could be explained because in studies where the traumatic event is something negative such as an earthquake or combat, the trauma would



logically be avoided. However, when the traumatic event is the diagnosis of an infant with a hemangioma, the hemangioma cannot be avoided as it is part of the infant.

Table 31

*Negative response categories*

Negative Responses = N	Themes	Frequency	%
People making rude comments or negative statements	I	49	37%
Surgery/Treatment of the hemangioma	H	20	15%
People staring at the baby's hemangioma	I	12	9%
Growth of the hemangioma was startling	H	12	9%
Worrying about the baby because of the hemangioma	H	10	8%
Hemangioma bled or ulcerated	H	7	5%
Crying over the hemangioma	H	6	5%
Diagnosis of the hemangioma	A	5	4%
Stress from the hemangioma	H	4	3%
Doctors not knowing how to diagnose hemangioma	H	3	2%
Family problems	A	2	2%
Could not work on PhD	H	1	1%
Totals		131	100%

This third research question asked what common themes have emerged from the question which asks the mothers to describe the most profound event that has occurred since the hemangioma was diagnosed. The results also found for the alternative as there were numerous common themes, over 80.9% were negative, and the most common negative comment was *people making rude comments or negative statements* (37%). This comment was combined with a very similar comment *people staring at the baby's hemangioma* for a total of 46% which relates to the *Intrusive* subscale theme. The other responses were also put into categories that relate to the other two themes (*Avoidance and Hyperarousal*). Table 32 triangulates the data from the EIS-R, HRQ, and the Qualitative question. Symptoms for *Intrusion*, *Hyperarousal*, and *Avoidance* (the three anchors) were present in all three components of the study.

Table 32

*Triangulation of EIS-R, HRQ, and Qualitative Question*

EIS-R	% indicated symptoms	HRQ	% indicated symptoms	Qualitative question	Present or not
Intrusion	55.5%	Subscale C	20.2%	Intrusion	46%
Avoidance	38.8%	Subscale D	29.0%	Avoidance	6%
Hyperarousal	48.1%	Subscale E	53.0%	Hyperarousal	48%

### Summary

Chapter 4 has presented the descriptive results of the two quantitative surveys and the one qualitative question included in the HRQ instrument. The results were triangulated to strengthen the study.

The data analysis for this study indicates that the results for one of the anchors, *Hyperarousal*, have the closest relationship between the two surveys and the one qualitative question (EIS-R at 48.1%, HRQ at 53.0%, and the Qualitative question at 48%). This suggests that the symptom of hyperarousal may be present in mothers of infants diagnosed with a hemangioma. While this study is strictly descriptive, and therefore does not imply any correlations in the data, it is interesting that results for the same theme (*Hyperarousal*) were fairly close for two different instruments and one narrative question. The *Intrusion* theme was at 55.5% for the EIS-R and a close 46% for the qualitative question but only 20.2% in the HRQ. The narrative question, therefore, more strongly related to the IES-R survey than the HRQ. The last theme, *Avoidance* had the 38.8% for the EIS-R and 29.0% for the HRQ. Only 6% of the qualitative question results indicated the symptom of *Avoidance*. This could be because the mothers were asked to explain the most profound event that occurred since the hemangioma was diagnosed. All descriptors that would relate to the definition of avoidance would not directly relate to a description of a profound event.

The quantitative and qualitative components of this study have been triangulated and all of the research questions have been found for the alternative hypothesis. The

triangulation of the data collected for this descriptive study may support future use of the HRQ instrument for assessing the impact of hemangiomas on the family system.

## CHAPTER 5:

### SUMMARY, CONCLUSION, AND RECOMMENDATIONS

#### Overview

The purpose of this descriptive survey study was to describe and explore the self-reported symptoms of stress of mothers of infants diagnosed with a hemangioma and to report if the mothers perceive that diagnosis as a traumatic event. The study included two self report instruments, the HRQ and the IES-R. There was one qualitative question included in the HRQ which explored common themes among the mothers. There were 148 mothers who completed all components of the study and 162 who completed all consent forms but only one of the two surveys. The participants were volunteers who were recruited from international hemangioma informational websites. All participants were mothers of infants who had been diagnosed with a hemangioma in the past year. Their ages ranged from 17 to 41 years and the mean age was 30 years. Over 75% of the participants were from the United States. Slightly over 89% of the mothers indicated that they had never been treated for a stress-related illness, and 58.8% indicated that the baby diagnosed with the hemangioma was their first child.

Because this was a descriptive study, no direct correlations have been made between the survey results and a diagnosis of acute stress or of a traumatic event. However, there were some interesting results which may suggest a distinct relationship.

Three main themes anchored this study: *Intrusion*, *Avoidance*, and *Hyperarousal*. Literature on ASD and PTSD indicates that all three of these symptoms should be present for a diagnosis of ASD or PTSD to be established and for the event to be considered

traumatic. Using these three themes as anchors, Table 32 presented in Chapter 4, indicate that symptoms were present in EIS-R and the HRQ instruments, as well as in the qualitative question included in this study. The results for one of the anchors, *Hyperarousal*, have the closest relationship between the two surveys and the one qualitative question (EIS-R at 48.1%, HRQ at 53.0%, and the qualitative question at 48%). This suggests that the symptom of hyperarousal may be present in mothers of infants diagnosed with a hemangioma. While this study was strictly descriptive, and therefore does not imply any direct correlations in the data, it is interesting to note that the results for *Hyperarousal* were fairly close for the results of the two different instruments and one narrative question. The *Intrusion* theme results show 55.5% symptoms present for the EIS-R and a close 46% for the qualitative question but only 20.2% in the HRQ. The narrative question, therefore, more strongly related to the EIS-R survey than the HRQ. The results for the last theme, *Avoidance* showed 38.8% for the EIS-R and 29.0% for the HRQ, and only 6% for the qualitative question.

The presence of these symptoms is significant because it suggests that parents of infants diagnosed with a hemangioma should be assessed for mental health counseling needs. According to Winston et al. (2002)

Within the first month after a traumatic event, individuals may display reexperiencing avoidance, and hyperarousal symptoms as well as dissociation (feelings of unreality or emotional numbing). The presence of these responses, collectively known as acute stress disorder (ASD), alerts providers to those who may be at risk for ongoing difficulties. (p. 1)

This study is important because it closes a 30-year gap and utilizes the first hemangioma specific instrument (HRQ) for a study which focused on measuring the

impact of the diagnosis of a hemangioma on maternal stress. It also provides a process for physicians to assess the mental health needs of the caregiver through the use of the HRQ instrument. Additionally, the results of this study supports the theory that having an infant diagnosed with a hemangioma is a stressful and traumatic event, which may be used by parents who are denied medical treatment by insurance companies who do not consider treatment a medical necessity. Without a medical necessity designation, families will continue to watch their child live with a deformity for many years, or will be faced with the task of providing the necessary funds to pay for treatment.

There were three research questions included in this study. The alternative was found for all three research questions and the results were triangulated.

This chapter also provides a discussion of the interpretation of findings along with a discussion of the research questions, limitations of the study, and implications for social change. Conclusions and recommendations for future research concerning the information presented in this study are also presented in this chapter.

### Interpretation of Findings

The following conclusions were derived from analysis of the data collected for each of the three research questions presented in this study. These findings are presented in consecutive order and begin with the applicable research question.

#### Research Question 1

Using the HRQ instrument, what level of stress is reported by mother's who have an infant who is diagnosed with a hemangioma and who completed this survey?

The data from this question was collected from the HRQ, and summarizes the percentage of mothers who indicated that they did or did not experience symptoms as a result of the diagnosis of the hemangioma. Using the three themes anchored for this study, *Hyperarousal*, *Avoidance*, and *Intrusion*, the highest percentage of symptoms was reported for Subscale E (*Hyperarousal*) with 53% indicating that they experienced symptoms; Subscale C, which relates to *Intrusion*, 20.2%; and for Subscale D, which relates to *Avoidance*, 29.0% indicated the presence of symptoms. Based on the data collected from the HRQ, no determination can be made as to whether or not the study participants meet the criteria for acute stress disorder. However, this question was answered as stated for the alternative hypothesis because there was a reported level of stress symptoms for mothers who completed the HRQ.

#### Research Question 2

The second research question explored what impact of event measure was reported by mothers of infants diagnosed with a hemangioma who completed the IES-R. This instrument was selected for use with the HRQ in order to establish first, that the diagnosis of an infant with a hemangioma may be a traumatic event, and second, to show a relationship to the symptoms of the HRQ. The IES-R data analysis results for the three subscales reports the presence of symptoms of *Intrusion* (55.5%), *Avoidance* (38.8%), and *Hyperarousal* (48.1%). which suggest that the mothers report that they perceive the event to be traumatic. Additionally, the results of the HRQ and the EIS-R have some similarities. The *Hyperarousal* subscale for the EIS-R was 48.1% as compared to 53.0% for the HRQ. The *Avoidance* subscale for the EIS-R was 38.8% as compared to 29.0% for



the HRQ. The subscale with the least similarity was the *Intrusion* subscale which had 55% for the EIS-R and 20.2% for the HRQ. Because this was a descriptive study, no direct correlations can be made between the survey results and the determination that the hemangioma diagnosis is a traumatic event. However, the results suggest a distinct relationship and that the mothers self-reported that they perceive the event to be traumatic. Therefore, the results indicate that there was a level of impact reported by the mothers and there is a relationship between the symptoms reported for the HRQ and the IES-R, also stated for the alternative hypothesis.

### Research Question 3

For the qualitative question included in the HRQ, what common themes have emerged from the question which asks the mothers to describe the most profound event that has occurred since the hemangioma was diagnosed?

Regarding the qualitative portion of this mixed study, one narrative question was analyzed for coding into similar categories. This question asked the mothers what the most profound event was since the hemangioma was diagnosed. The narrative responses were split into negative and positive answers since the word profound could be interpreted both ways. Over 80% of the responses were negative. Of those 80%, the leading negative response was that *people were making rude comments or negative statements*. Another similar response *people staring at the baby's hemangioma* was reported by 9% of the mothers. These two similar responses were combined to equal 46% and were related to one of the three subscale themes, *Intrusion*. The remaining responses were also related to the other themes. For the symptom *Avoidance*, only 6% of the

mothers reported a symptom but for *Hyperarousal*, 48% reported the presence of the symptom through their descriptive comments. Therefore, common themes were reported in the narrative question by the mothers of infants diagnosed with a hemangioma and these common themes were triangulated with the results of the two quantitative instruments. This question was also, therefore, found for the alternative hypothesis.

Combined, all of the data analysis results reported by the mothers indicated that the diagnosis of the infant with the hemangioma was stressful and perceived as a traumatic event. However, this was a descriptive study and no direct correlations can be made from the reported results.

### Conclusion

This study described and explored the results of two self-report survey instruments completed by mothers who had an infant diagnosed with a hemangioma in addition to the results of the one qualitative question included in the HRQ. Some of the symptoms that must be present for acute stress disorder to be diagnosed have been reported by mothers in this study, though no correlations have been made. In addition, the symptoms reported by the mothers from the EIS-R suggest that they perceive the diagnosis as a traumatic event, though once again no direct correlation can be made. However, in triangulating the results of the two instruments with the one qualitative question, there is a strong indication that the diagnosis of an infant with a hemangioma was considered acutely stressful, and was perceived as a traumatic event by the mothers who participated in this study.

### Recommendation

This study can help physicians who treat infants with hemangiomas to begin to explore the mental health of the parents. It may also validate the use of the HRQ for assessing the level of stress of the parents. However, the study was very limited, therefore, more research should be done with a control group, both mothers and fathers, and with repeated assessments at various intervals following the diagnosis.

This was the first study to assess the theory that having an infant diagnosed with a hemangioma may be acutely stressful and perceived as a traumatic event. The data may not be conclusive, because it was a descriptive study, but the results are significant enough that they may raise awareness about the impact of the diagnosis on the family system. The results may also provide documentation for parents who must appeal the denial of treatment of the hemangioma by their insurance company because treatment is not considered to be medically necessary.

### Post script

This letter was received in February 2008 by this researcher. It eloquently sums up the journey of many families who have a child diagnosed with a hemangioma. It is reprinted with approval from the mother.

### A Letter From a Mother of a Child With a Hemangioma

Dear Vascular Birthmarks Foundation:

Four years ago, I had been literally devastated and so very sad when I discovered that my beautiful daughter was born and shortly thereafter developed a hemangioma on her mouth and upper lip. My dreams were shattered and a heavy

depression took over my life. This journey was not what I had expected. How could this have happened to me? What did I eat? Was I exposed to something dangerous when I was pregnant? I had been so prepared to meet her and I couldn't wait to show her off and share her with the world. I was in awe when those dreams were shattered and I couldn't believe that what should have been the happiest days of my life were riddled with tears, sadness, confusion, and guilt. The first year of her life was a combination of the absolute happiest and saddest days of my whole existence on this planet. How horrible! How can a decent mother even write or think those thoughts that I was having? I wanted to hide, go away and stay home. I avoided people, places and groups. When I did put on my brave face and venture out, it usually resulted in stares, ogles, and rude comments, therefore fueling my desire to stay home and cry. Nobody around me understood my journey, and so my loneliness with this set in. In many ways over the past 4 years, this journey has gotten a bit easier for me to bear, because she has grown, and it has faded a bit, not to mention I've gotten a tougher skin because it was important to me that she see me deal with this in a positive way. But it still hurts and I still cry sometimes, because I don't want her to feel bad about her appearance. I have been told by the only surgeon who does this in our province that we will have to wait until she is 10 or 11 before he will touch it. We used to visit him every year with hopes that he would change his mind, but he hasn't. He thinks we are being vain and we are doing this for US, and not HER. He says we have to be careful not to remove (excise) the hemangioma on her lip as we may deflate the lip and it will impact the rest of her life. What do you think? Should we just wait another 5 years for it to finish involuting? And let her "deal with this" on her own,

or should we keep trying to pursue this and have it removed? We gave up trying after the surgeon told us we could ruin her lip for life....but we want to know your opinion!

PLEASE! Any advice at all will be extremely helpful. Signed, N.P. (2/22/08).

Update: The child is scheduled to meet with a surgeon in New York who has informed the mother, after reviewing current photos, that he can safely remove the hemangioma and that her daughter will look normal after one outpatient surgical procedure. The surgery is not covered by the parent's insurance but they have opted to have it done and agreed to pay the projected cost (over \$10,000). This is a typical scenario that this researcher has frequently encountered over the past 14 years and it is the reason that this study has been undertaken.

## REFERENCES

- American Psychiatric Association. *Diagnostic and statistical manual of mental disorders*, (4<sup>th</sup> ed.). Washington, DC: American Psychiatric Association.
- Ball, K. F. (Ed.). (2003). *Sturge-Weber Syndrome*. Mt. Freedom, NJ: Sturge-Weber Foundation.
- Barden, C. R., Ford, M. E., Jensen, A. G. Rogers-Salyer, M., & Salyer, K. E. (1989). Effects of craniofacial deformity in infancy on the quality of mother-infant interaction. *Child Development*, 60, 819-824.
- Barrio, V. R. & Drolet, B. A. (2005). Treatment of hemangiomas of infancy. *Dermatologic Therapy*, 18, 151-159.
- Batniji R. K., Buckingham, E. D., & Williams, E. F. (2005). An aesthetic approach to facial hemangiomas. *Archives of Facial Plastic Surgery*, 7, 301-306.
- Batta, K., Goodyear, H. M., Moss, C., Williams, H. C., Hiller, L., & Waters, R. (2002). Randomized controlled study of early-pulsed dye laser treatment of uncomplicated childhood hemangiomas: Results of a 1-year analysis. *Lancet*, 30, 521-527.
- Bertalanffy, L. (1968). *General systems theory: Foundations, development, applications*. New York: George Braziller, Inc.
- Bradbury, E. T., & Hewison, J. (1994). Early parental adjustment to visible congenital disfigurement. *Child: Care, Health and Development*, 20, 251-266.
- Brogdon, J. (1999). Psychosocial impact of congenital vascular lesions. In M. Waner & J. Suen (Eds.), *Hemangiomas and vascular malformations of the head and neck*, pp. 217-232). New York: Wiley-Liss.
- Bryant, R. A., Harvey, A. G., Dang, S. T., & Sackville, T. (1998). Assessing acute stress disorder: Psychometric properties of a structured clinical interview. *Psychological Assessment*, 10(3), 215-220.
- Campbell, D. T., & Stanley, J. C. (1963). *Experimental and quasi-experimental designs for research*. Boston: Houghton Mifflin.
- Canam, C. (1993). Common adaptive tasks facing parents of children with chronic conditions. *Journal of Advanced Nursing*, 18, 46-53.
- Cardena, E., Koopman, C., Classen, C., Waelde, L. C. & Spiegel, D. (2000).

- Psychometric properties of the Stanford Acute Stress Reaction Questionnaire (SASRQ): A valid and reliable measure of acute stress. *Journal of Traumatic Stress, 13*(4), 719-734.
- Cate-Miller, A., Cate, I. M., Watson, H. S., & Geronemus, R. G. (1999). Stress and satisfaction in parents of children with facial port-wine stains. *Pediatric Dermatology, 16*(3), 190-197.
- Chamlin, S. L. (2005). Infantile hemangiomas: Measuring the burden of disease. Proceedings of a research workshop on infantile hemangiomas. *Pediatric Dermatology, 22*(5), 383-4-6.
- Classen, C. (n.d.). Acute stress disorder as a predictor of PTSD. *Directions in Psychiatry, 19*(25).
- Cox, M. J. & Paley, B. (1997). Families as systems. *Annual Reviews in Psychology 1997, 48*, 243-267.
- Creswell, J. W. (2003). *Research design: Qualitative, quantitative, and mixed methods approaches*. Thousand Oaks, CA: Sage.
- Dellve, L., Samuelsson, L., Tallborn, A., Fasth, A., & Hallberg, L. (2005). Stress and well-being among parents of children with rare diseases: A prospective intervention study. *Issues and Innovations in Nursing Practice, 392-402*.
- Dieterich-Miller, C. A., & Safford, P. L. (1992). Psychosocial development of children with hemangiomas: Home, school, health care collaboration. *Children's Hospital of Cleveland, 21*(2), 84-89.
- Drotar, D., Baskiewicz, A., Irvin, N., Kennell, J. & Klaus, M. (1975). The adaptation of parents to the birth of an infant with a congenital malformation: A hypothetical model. *Pediatrics, 56*(5), 710-717.
- Elhai, J. D., Gray, M. J., Kashdan, T. D., & Franklin, C. L. (2005). Which instruments are most commonly used to assess traumatic event exposure and posttraumatic effects?: A survey of traumatic stress professionals. *Journal of Traumatic Stress, 18*(5), 541-545.
- Endriga, M. C., & Speltz, M. L. (1997). Face-to-face interaction between infants with orofacial clefts and their mothers. *Journal of Pediatric Psychology, 22*(4), 439-453.
- Engel, G. L. (1962). *Psychological development in health and disease*. Philadelphia: WB Saunders.

- Fleming, W. M. (2007). Family systems theory: Challenges and future directions. Retrieved June 11, 2007 from [www.family.jrank.org](http://www.family.jrank.org).
- Fortier, L., M., & Wanlass, R. L. (1984). Family crisis following the diagnosis of a handicapped child. *Family Relations*, 33(1), 13-24.
- Glassman, P. A., Model, K. E., Kahan, J. P., Jacobson, P. D., & Peabody, J. W. (1997). The role of medical necessity and cost-effectiveness in making medical decisions. *Annals of Internal Medicine*, 126(2), 152-156.
- Green, S. B., & Salkind, N. J. (2005). *Using SPSS for windows and macintosh: Analyzing and understanding data (4<sup>th</sup> ed.)*. Upper Saddle River, NJ: Pearson Prentice Hall.
- Hill, R. (1966). Contemporary developments in family theory. *Journal of Marriage and the Family*, 28, 10-25.
- Hochman, M. (2001). Management of cutaneous hemangiomas. *Facial Plastic Surgery Clinics of North America*, 9(4) 621-628.
- Hodapp, R. M., Fidler, D. J., & Smith, A. C. (1998). Stress and coping in families of children with smith-magenis syndrome. *Journal of Intellectual Disability Research*, 42(5), 332-340.
- Hodgkinson, P., Brown, S., Duncan, D., Grant, C., McNaughton, A., Thomas, P., et al. (2005). Management of children with cleft lip and palate: A review describing the application of multidisciplinary team working in this condition based upon the experiences of a regional cleft lip and palate centre in the United Kingdom. *Fetal and Maternal Medicine Review*, 16(1), 1-27.
- Horowitz, M., Wilner, M., & Alvarez, W. (1979). Impact of event scale: A measure of subjective stress. *Psychosomatic Medicine*, 41, 209-219.
- Janssens, A., Buljevac, D., van Doorn, P. A., van der Meche, F., Polman, C., Passchier, J., & Hintzen, R. (2006). Prediction of anxiety and distress following diagnosis of multiple sclerosis: a two-year longitudinal study. *Multiple Sclerosis*, 12, 794-801.
- Jedlicka-Kohler, I., Gotz, M., & Eichler, I. (1996). Parents recollection of the initial communication of the diagnosis of cystic fibrosis. *Pediatrics*, 97(2), 204-209.
- Johnson, B., & Christensen, L. (2004). *Educational research: Quantitative, qualitative, and mixed approaches* (2nd ed.). Boston: Pearson Education.
- Kazak, A. E. (1989). Families of chronically ill children: A systems and social-ecological model of adaptation and challenge. *Journal of Consulting and Clinical Psychology*, 57(2), 25-30.



- Kazak, A. E. (1992). Stress, change and families: Theoretical and methodological considerations. *Journal of Family Psychology*, 6(2), 120-124.
- Kersting, A., Dorsch, M., Wesselmann, U., Ludorff, K., Witthaut, J., Ohrmann, P., Hornig-Franz, I., Klockenbusch, W., Harms, E., & Arolt, V. (2004). Maternal posttraumatic stress response after the birth of a very low-birth-weight infant. *Journal of Psychosomatic Response*, 57(5), 473-476.
- Lande, R. G., Crawford, P. M., & Ramsey, B. J. (2001). Psychosocial impact of vascular birthmarks. *Facial Plastic Surgery Clinics of North America*, 9, (4,) 561-566.
- Majnemer, A., Limperopoulos, C., Shevell, M., Rohlicek, B., Rosenblatt, B., & Tchervenkova, C. (2006). Health and well-being of children with congenital cardiac malformations, and their families, following open-heart surgery. *Cardiology in the Young*, 16, 157-164.
- Mulliken, J. & Young, A. (1988). *Vascular birthmarks: Hemangiomas and malformations*. Philadelphia, Pa. WB Saunders.
- Ozer, E. J., Best, S. R., Lipsey, T. L., & Weiss, D. S. (2003). Predictors of posttraumatic stress disorder and symptoms in adults: A meta-analysis. *Psychological Bulletin*, 129(1), 52-73.
- Pelchat, D., Ricard, N., Bouchard, J., Perreault, M., Saucier, J., Berthiaume, & Bisson, J. (1999). Adaptation of parents in relation to their 6-month old infant's type of disability. *Child: Care, Health and Development*, 25(4), 377-397.
- Pope A. W. (2000). Psychologic aspects of facial vascular lesions in infancy and early childhood. *NYU Vascular Anomalies 2000 Conference*, p. 70.
- Pope, A. W., Tillman, K., & Snyder, H. T. (2005). Parenting stress in infancy and psychosocial adjustment in toddlerhood: A longitudinal study of children with craniofacial anomalies. *The Cleft Palate-Craniofacial Journal*, 42(5), 556-559.
- Rozell-Shannon, L. (2007). *Modification of the Stanford acute stress reaction questionnaire (SASRQ): Measuring stress from a hemangioma diagnosis*. Manuscript submitted for publication.
- Rozell-Shannon, L., Marshall, C., & Waner, M. (1997). *Birthmarks: A guide to hemangiomas and vascular malformations*. Women's Health Publishing: Minden, NV.
- Rubin, H. J., & Rubin, I. S. (2005). *Qualitative interviewing: The art of hearing data* (2<sup>nd</sup> ed.). Thousand Oaks, CA: Sage.

- Shakin-Kunkel, E., Zager, R. Hausman, C., & Rabinowitz, L. (1994). An interdisciplinary group for parents of children with hemangiomas. *Psychosomatics*, 35(6), 524-532.
- Shaw, R., Deblois, T., Ikuta, L., Ginzburg, K., Fleisher, B., & Koopman, C. (2006). Acute stress disorder among parents of infants in the neonatal intensive care nursery. *Psychosomatics*, 47(3), 206-212.
- Simmons, O., & Chiu, E. S. (2004). Current management of vascular anomalies. *Pediatric Review, Children's Hospital, New Orleans*, 18(12), 1-4.
- Singer, S. J., & Bergthold, L. A. (2001). Cosmetic vs. reconstructive surgery for cleft palate A window into the medical necessity debate. *JAMA*, 286(17), p. 2162.
- St. John, D., Pai, L., Belfer, M., & Mulliken, J. (2003). Effects of a child with a craniofacial anomaly on stability of the parental relationship. *The Journal of Craniofacial Surgery*, 14(5), 704-708.
- Sundin, E. C., & Horowitz, M. J. (2003). Horowitz's impact of event scale evaluation of 20 years of use. *Psychosomatic Medicine*, 65, 870-876.
- Tak, Y. R., & McCubbin, M. (2002). Family stress, perceived social support and coping following the diagnosis of a child's congenital heart disease. *Journal of Advanced Nursing*, 39(2), 190-198.
- Tanner, J. L., Dechert, M. P., & Frieden, I. J. (1998). Growing up with a facial hemangioma: Parent and child coping and adaptation *American Academy of Pediatrics*, 101(2), 446-452.
- Triola, M. F. (2005). *Elementary statistics* (9<sup>th</sup> ed.). Boston: Pearson-Addison Wesley.
- U.S. Department of Health and Human Services. (2003). Coping with a traumatic event: Information for health professionals. Retrieved on June 12, 2007 from [www.dying.about.com](http://www.dying.about.com).
- Vacca, J. J. (2006). Parent perceptions of raising a child with a severe physical disability: Implications for mental health providers. *Best Practices in Mental Health*, 2 (1), 59-73
- Waner, M. & Suen, J. Y. (1999). *Hemangiomas and vascular malformations of the head and neck*. NY: Wiley-Liss.
- Weinstein, J. M., & Chamlin, S. L. (2005). Quality of life in vascular anomalies. *Lymphatic Research and Biology*, 3(4), 256-259.

- Weiss, D. S., & Marmar, C. R. (1997). Impact of event scale – revised (IES-R). Retrieved May 24, 2007, from <http://www.swin.edu.au/victims/resources/assessment/ptsd/ies-r.html>.
- Williams, E. F., Hochman, M., Rodgers, B. J., Brockbank, D., Shannon, L., & Lam, S. M. (2003). A psychological profile of children with hemangiomas and their families. *Archives of Facial Plastic Surgery*, 5, 229-234.
- Winston, F. K., Kassam-Adams, N., Vivarelli-O'Neill, C., Ford, J., Newman, E., Baxt, C., Stafford, P., & Cnaan, A. (2002). Acute stress disorder symptoms in children and their parents after pediatric traffic injury. *Pediatrics*, 109(6), 1-7.

## APPENDIX A: HRQ

### Hemangioma Reaction Questionnaire (HRQ)

#### **DIRECTIONS:**

Recall the stressful events that occurred in your life when your baby was diagnosed with a hemangioma.

**Briefly describe the most profound event that occurred since your baby was diagnosed with a hemangioma. (type your answer in this blank area. Try to keep it to 5 or 6 single spaced lines, if possible):**

#### **How disturbing was this event to you? (Please mark one):**

- Not at all disturbing \_\_\_\_\_  
Somewhat disturbing \_\_\_\_\_  
Moderately disturbing \_\_\_\_\_  
Very disturbing \_\_\_\_\_  
Extremely disturbing \_\_\_\_\_

**DIRECTIONS:** Below is a list of experiences people sometimes have during and after a stressful event. Please read each item carefully and decide how well it describes *your* experience following the diagnosis of your infant's hemangioma. Refer to this event in answering the items below. Use the 0-5 point scale shown below and circle the number that best describes your experience. Place an "X" or a check mark before the number that you are selecting.

- |     | 1-----  | 2-----      | 3-----      | 4-----      | 5-----                  | 6-----      |
|-----|---|-------------|-------------|-------------|-------------------------|-------------|
|     | not   | very rarely | rarely      | sometimes   | often                   | very often  |
|     | experienced   | experienced | experienced | experienced | experienced             | experienced |
| 1.  | I had difficulty falling or staying asleep.   |             |             |             | _ 1 _ 2 _ 3 _ 4 _ 5 _ 6 |             |
| 2.  | I felt restless.  |             |             |             | _ 1 _ 2 _ 3 _ 4 _ 5 _ 6 |             |
| 3.  | I felt a sense of timelessness.   |             |             |             | _ 1 _ 2 _ 3 _ 4 _ 5 _ 6 |             |
| 4.  | I was slow to respond.  |             |             |             | _ 1 _ 2 _ 3 _ 4 _ 5 _ 6 |             |
| 5.  | I tried to avoid feelings about the hemangioma.   |             |             |             | _ 1 _ 2 _ 3 _ 4 _ 5 _ 6 |             |
| 6.  | I had repeated distressing dreams of the hemangioma.  |             |             |             | _ 1 _ 2 _ 3 _ 4 _ 5 _ 6 |             |
| 7.  | I felt extremely upset if exposed to events that reminded me of an aspect of the hemangioma.        |             |             |             | _ 1 _ 2 _ 3 _ 4 _ 5 _ 6 |             |
| 8.  | I would jump in surprise at the least thing.  |             |             |             | _ 1 _ 2 _ 3 _ 4 _ 5 _ 6 |             |
| 9.  | Thoughts of the hemangioma made it difficult for me to perform work or other things I needed to do. |             |             |             | _ 1 _ 2 _ 3 _ 4 _ 5 _ 6 |             |
| 10. | I did not have the usual sense of who I am.   |             |             |             | _ 1 _ 2 _ 3 _ 4 _ 5 _ 6 |             |
| 11. | I tried to avoid activities that reminded me of the hemangiomas.                                    |             |             |             | _ 1 _ 2 _ 3 _ 4 _ 5 _ 6 |             |
| 12. | I felt hypervigilant or "on edge".  |             |             |             | _ 1 _ 2 _ 3 _ 4 _ 5 _ 6 |             |

	1-----2-----3-----4-----5-----6	
	not very rarely rarely sometimes often very often	
	experienced experienced experienced experienced experienced experienced	
13.	I experienced myself as though I were a stranger.	_1 _2 _3 _4 _5 _6
14.	I tried to avoid conversations about the hemangioma.	_1 _2 _3 _4 _5 _6
15.	I had a bodily reaction when exposed to reminders of the hemangioma.	_1 _2 _3 _4 _5 _6
16.	I had problems remembering important details about the hemangioma.	_1 _2 _3 _4 _5 _6
17.	I tried to avoid thoughts about the hemangioma.	_1 _2 _3 _4 _5 _6
18.	Things I saw looked different to me from how I know they really looked.	_1 _2 _3 _4 _5 _6
19.	I had repeated and unwanted memories of the hemangioma.	_1 _2 _3 _4 _5 _6
20.	I felt distant from my own emotions.	_1 _2 _3 _4 _5 _6
21.	I felt irritable or had outbursts of anger.	_1 _2 _3 _4 _5 _6
22.	I avoided contact with people who reminded me of the hemangioma, including my child.	_1 _2 _3 _4 _5 _6
23.	I would suddenly act or feel as if I had learned about the hemangioma all over again.	_1 _2 _3 _4 _5 _6
24.	My mind went blank.	_1 _2 _3 _4 _5 _6
25.	I had amnesia for large periods after the hemangioma diagnosis.	_1 _2 _3 _4 _5 _6
26.	The hemangioma caused problems in my relationships with other people.	_1 _2 _3 _4 _5 _6
27.	I had difficulty concentrating.	_1 _2 _3 _4 _5 _6
28.	I felt estranged or detached from other people.	_1 _2 _3 _4 _5 _6
29.	I had a vivid sense that the hemangiomas was being diagnosed all over again.	_1 _2 _3 _4 _5 _6
30.	I tried to stay away from places that reminded me of the hemangioma.	_1 _2 _3 _4 _5 _6

**On how many days did you experience any of the above symptoms of distress? (Please mark one):**

none	_____	one	_____
two	_____	three	_____
four	_____	five or more	_____

## APPENDIX B: HRQ SCORING BASED ON SASRQ

### Scoring of the Stanford Acute Stress Reaction Questionnaire

#### A. Traumatic Event

The open-ended item or a special series of items tailored to the situation establish the fact that the person has experienced, witnessed or confronted an event involving actual or threatened death or serious injury or threat to the physical integrity of oneself or others. The rating scale from "not at all disturbing" to "extremely disturbing" assess the intensity of the person's response.

#### B. Dissociative Symptoms

##### 1) Subjective sense of numbing, detachment, or absence of emotional responsiveness

Items 20, 28

##### 2) A reduction in awareness of one's surroundings

Items 4, 24

##### 3) Derealization

Items 3, 18

##### 4) Depersonalization

Items 10, 13

##### 5) Dissociative amnesia, i.e., inability to recall an important aspect of the trauma

Items 16, 25

#### C. The traumatic event is persistently reexperienced

Items 6, 7, 15, 19, 23, 29

#### D. Marked avoidance of stimuli that arouse recollections of the trauma

Items 5, 11, 14, 17, 22, 30

#### E. Marked symptoms of anxiety or increased arousal

Items 1, 2, 8, 12, 21, 27

#### F. Impairment in functioning

Items 9, 26

## APPENDIX C: EIS-R

### IMPACT OF EVENT SCALE-REVISED

*Instructions:* The following is a list of difficulties people sometimes have after stressful life events. Please read each item, and then indicate how distressing each difficulty has been for you *during the past 7 days* with respect to the disaster. How much were you distressed or bothered by these difficulties?

		Not at all	A little bit	Moder- ately	Quite a bit	Ex- treme- ly
1	Any reminder brought back feelings about it.	0	1	2	3	4
2	I had trouble staying asleep.	0	1	2	3	4
3	Other things kept making me think about it.	0	1	2	3	4
4	I felt irritable and angry.	0	1	2	3	4
5	I avoided letting myself get upset when I thought about it or was reminded of it.	0	1	2	3	4
6	I thought about it when I didn't mean to.	0	1	2	3	4
7	I felt as if it hadn't happened or wasn't real.	0	1	2	3	4
8	I stayed away from reminders about it.	0	1	2	3	4
9	Pictures about it popped into my mind.	0	1	2	3	4
10	I was jumpy and easily startled.	0	1	2	3	4
11	I tried not to think about it.	0	1	2	3	4
12	I was aware that I still had a lot of feelings about it, but I didn't deal with them.	0	1	2	3	4
13	My feelings about it were kind of numb.	0	1	2	3	4
14	I found myself acting or feeling like I was back at that time.	0	1	2	3	4
15	I had trouble falling asleep.	0	1	2	3	4
16	I had waves of strong feelings about it.	0	1	2	3	4
17	I tried to remove it from my memory.	0	1	2	3	4
18	I had trouble concentrating.	0	1	2	3	4
19	Reminders of it caused me to have physical reactions, such as sweating, trouble breathing, nausea, or a pounding heart.	0	1	2	3	4
20	I had dreams about it.	0	1	2	3	4
21	I felt watchful and on guard.	0	1	2	3	4
22	I tried not to talk about it.	0	1	2	3	4

## APPENDIX D: IES-R SCORING

### IMPACT OF EVENT SCALE- REVISED

**INSTRUCTIONS:** Below is a list of difficulties people sometimes have after stressful life events. Please read each item, and then indicate how distressing each difficulty has been for you **DURING THE PAST SEVEN DAYS** with respect to \_\_\_\_\_, which occurred on \_\_\_\_\_. How much were you distressed or bothered by these difficulties?

Item Response Anchors are 0 = Not at all; 1 = A little bit; 2 = Moderately; 3 = Quite a bit; 4 = Extremely.

The Intrusion subscale is the **MEAN** item response of items 1, 2, 3, 6, 9, 14, 16, 20. Thus, scores can range from 0 through 4.

The **Avoidance** subscale is the **MEAN** item response of items 5, 7, 8, 11, 12, 13, 17, 22. Thus, scores can range from 0 through 4.

The **Hyperarousal** subscale is the **MEAN** item response of items 4, 10, 15, 18, 19, 21. Thus, scores can range from 0 through 4.

1. Any reminder brought back feelings about it.
2. I had trouble staying asleep.
3. Other things kept making me think about it.
4. I felt irritable and angry.
5. I avoided letting myself get upset when I thought about it or was reminded of it.
6. I thought about it when I didn't mean to.
7. I felt as if it hadn't happened or wasn't real..
8. I stayed away from reminders of it.
9. Pictures about it popped into my mind.
10. I was jumpy and easily startled.
11. I tried not to think about it.
12. I was aware that I still had a lot of feelings about it, but I didn't deal with them.
13. My feelings about it were kind of numb.
14. I found myself acting or feeling like I was back at that time.
15. I had trouble falling asleep.
16. I had waves of strong feelings about it.
17. I tried to remove it from my memory.
18. I had trouble concentrating.
19. Reminders of it caused me to have physical reactions, such as sweating, trouble breathing, nausea, or a pounding heart.
20. I had dreams about it.



21. I felt watchful and on-guard.

22. I tried not to talk about it.

Citations:

Weiss, D.S. & Marmar, C.R. (1997). The Impact of Event Scale-Revised. In J.P. Wilson, & T. M. Keane (Eds.), *Assessing Psychological Trauma and PTSD: A Practitioner's Handbook*. (pp. 399-411). New York: Guilford.

Weiss, D. S. (2004). The Impact of Event Scale-Revised. In J. P. Wilson, & T. M. Keane (Eds.), *Assessing psychological trauma and PTSD: A practitioner's handbook* (2<sup>nd</sup> ed., pp. 168-189). New York: Guilford Press.

## APPENDIX E: Scoring for SASRQ used for HRQ

### Scoring of the Stanford Acute Stress Reaction Questionnaire

#### A. Traumatic Event

The open-ended item or a special series of items tailored to the situation establish the fact that the person has experienced, witnessed or confronted an event involving actual or threatened death or serious injury or threat to the physical integrity of oneself or others. The rating scale from "not at all disturbing" to "extremely disturbing" assess the intensity of the person's response.

#### B. Dissociative Symptoms

##### 1) Subjective sense of numbing, detachment, or absence of emotional responsiveness

Items 20, 28

##### 2) A reduction in awareness of one's surroundings

Items 4, 24

##### 3) Derealization

Items 3, 18

##### 4) Depersonalization

Items 10, 13

##### 5) Dissociative amnesia, i.e., inability to recall an important aspect of the trauma

Items 16, 25

#### C. The traumatic event is persistently reexperienced

Items 6, 7, 15, 19, 23, 29

#### D. Marked avoidance of stimuli that arouse recollections of the trauma

Items 5, 11, 14, 17, 22, 30

#### E. Marked symptoms of anxiety or increased arousal

Items 1, 2, 8, 12, 21, 27

#### F. Impairment in functioning

Items 9, 26

## APPENDIX F: CONFIDENTIALITY AGREEMENT

During the course of my activity in collecting data for this research, “Assessing Maternal Response to the Impact of Hemangiomas on the Family System,” I will have access to information, which is confidential and should not be disclosed. I acknowledge that the information must remain confidential, and that improper disclosure of confidential information can be damaging to the participant.

By signing this Confidentiality Agreement I acknowledge and agree that:

1. I will not disclose or discuss any confidential information with others, including friends or family.
2. I will not in any way divulge copy, release, sell, loan, alter or destroy any confidential information except as properly authorized.
3. I will not discuss confidential information where others can overhear the conversation. I understand that it is not acceptable to discuss confidential information even if the participant’s name is not used.
4. I will not make any unauthorized transmissions, inquiries, modification or purging of confidential information.
5. I agree that my obligations under this agreement will continue after termination of the job that I will perform.
6. I understand that violation of this agreement will have legal implications.
7. I will only access or use systems or devices I’m officially authorized to access and I will not demonstrate the operation or function of systems or devices to unauthorized individuals.

Signing this document, I acknowledge that I have read the agreement and I agree to comply with all the terms and conditions stated above.

*Linda Rozell-Shannon*

**Signature:**

July 20, 2007

**Date:**

## APPENDIX G: WEB SITE ANNOUNCEMENT

### **Assessing Maternal Response to the Impact of Hemangiomas on the Family System**

#### Website Announcement for Vascular Birthmarks Foundation Websites and the MSN Vascular Birthmarks Support Group Website

Linda Rozell-Shannon, President and Founder of the Vascular Birthmarks Foundation, is currently earning her PhD in Education at Walden University. Linda has researched and written about vascular birthmarks for the past twelve years and is currently focusing on them in her doctoral studies. Linda is conducting her PhD dissertation study to assess the mother's response to having a baby diagnosed with a hemangioma.

In order to substantiate her research, Linda needs some volunteers. These volunteers must be first-time mothers of a baby diagnosed with a hemangioma within one year of participating in this study. The mother must be between 25 and 40 years of age and have not been treated for stress at any time prior to participating in this study. There will be two short questionnaires entitled the "Hemangioma Reaction Questionnaire" and the "Impact of Event Scale – Revised." It should take less than 30 minutes to complete these questionnaires. They are located on the SurveyMonkey website for ease in completing and for ease in analyzing the data from the completed surveys.

*If you would like to volunteer to participate in this study, please go to this link and complete the two surveys:*

<a href="http://www.surveymonkey.com/s.aspx?sm=2LJ5sAx0\_2fMEFRA1eDO6OSA\_3d\_3d">Click Here to take survey</a>

*If you are unable to go to the surveys from this link, please copy and past this link into your browser:*

[http://www.surveymonkey.com/s.aspx?sm=2LJ5sAx0\\_2fMEFRA1eDO6OSA\\_3d\\_3d](http://www.surveymonkey.com/s.aspx?sm=2LJ5sAx0_2fMEFRA1eDO6OSA_3d_3d)

Before you complete the surveys, please download and complete the Participant Form, and the Hemangioma Consent Form at the following link (they are 2 separate forms but they are linked together as one word document for your ease in completing and for ease in emailing back to [hvbf@aol.com](mailto:hvbf@aol.com) when you have finished.

Save a copy of the Participant Form and the Hemangioma Consent Form to your desktop. You can fill them in by backspacing over the areas you need to complete or by placing an

“x” where indicated. All personal information will remain anonymous and confidential and will not be disclosed or shared with anyone.

Your participation is greatly needed and greatly appreciated. If you have any questions, please send them to [hvbf@aol.com](mailto:hvbf@aol.com).

## APPENDIX H: CONSENT FORM ON SURVEYMONEKY WEB SITE

### CONSENT FORM

You are invited to participate in a research study to explore the possible relationship between stress and having a baby diagnosed with a hemangioma. You were selected as a possible participant due to fact that you are the mother of a baby with a hemangioma that has been diagnosed within the past year. Please read this form and ask any questions you may have before acting on this invitation to be in the study.

This dissertation study is being conducted by Linda Rozell-Shannon, a doctoral candidate at Walden University, and President and Founder of the Vascular Birthmarks Foundation. This study has been tested and it will take approximately 10 minutes to complete the entire study.

#### Background Information:

The purpose of this study is to assess and explore if there is a relationship between stress and the diagnosis of an infant with a hemangioma.

#### Procedures:

If you agree to be in this study, you will be asked to complete two questionnaires. There are two consent forms. This is the first consent form and it applies to the first instrument, the HRQ. The second consent form follows this form and it is for the IES-R. A separate consent form is required for each instrument.

#### Voluntary Nature of the Study:

Your participation in this study is strictly voluntary. Your decision whether or not to participate will not affect your current or future relations with the Vascular Birthmarks Foundation. If you initially decide to participate, you are still free to withdraw at any time without affecting those relationships. If at any time during your completion of the survey or following the completion of the survey you feel traumatized or need to talk to someone regarding this subject matter, Please feel free to contact the following Mental Health Specialist.

Elissa Rifkin, M.Ed. CMHC  
605 S. Central Ave.  
Clayton, MO 63105  
Clinical Mental Health Specialist  
Vascular Birthmark Foundation  
314-495-7531  
psyelectro@aol.com

**Risks and Benefits of Being in the Study:**

There are no risks associated with participating in this study. However, this study may provide a process for physicians to refer affected families to an appropriate mental health counselor, if needed. It may also assist families with providing justification for treatment of the hemangioma. In the event you experience stress or anxiety during your participation in the study you may discontinue your participation. You may refuse to answer any questions you consider invasive or stressful.

**Compensation:**

There will be no compensation provided for your participation in this study.

**Confidentiality:**

The records of this study will be kept private. In any report of this study that might be published, the researcher will not include any information that will make it possible to identify you. Research records will be kept in a locked file, and only the researcher will have access to the records.

**Contacts and Questions:**

The researcher conducting this study is Linda Rozell-Shannon. The researcher's Dissertation Chairman is Dr. Mel Finkenberg at [mfinkenb@waldenu.edu](mailto:mfinkenb@waldenu.edu)

## APPENDIX I: PARTICIPANT FORM

### Assessing Maternal Response to the Impact of Hemangiomas on the Family System

#### Participant Qualification Form

##### Instructions:

Complete this form if your infant has been diagnosed with a hemangioma within one year of completing this form. All personal information will remain anonymous and confidential and will not be disclosed or shared with anyone.

Mother's Name (Last)

(First)

---

Phone (home)

Email:

Today's Date

---

Phone (cell or work)

Mother's current age:

---

Your age when the baby was born?

Have you ever been treated for stress?

---

Where was/is hemangioma located?

Date when hemangioma was diagnosed

---

How old was the baby when diagnosed?

Is this your first baby? (yes or no)

---

Your Name (Print/type)

Your Signature (electronic or write)

---

Your participation is greatly appreciated. If you have any questions, feel free to email Linda directly at [hvbf@aol.com](mailto:hvbf@aol.com).



## APPENDIX J: WALDEN IRB APPROVAL

Dear Ms. Rozell-Shannon:

This email is to notify you that the Institutional Review Board (IRB) has approved your application for the study entitled, "Assessing Maternal Response to the Impact of Hemangiomas on the Family System."

Your approval # is 11-30-07-0320781. You will need to reference this number in the appendix of your dissertation and in any future funding or publication submissions.

Your IRB approval expires on November 30, 2008. One month before this expiration date, you will be sent a Continuing Review Form, which must be submitted if you wish to collect data beyond the approval expiration date.

Your IRB approval is contingent upon your adherence to the exact procedures described in the final version of the IRB application materials that have been submitted as of this date. If you need to make any changes to your research staff or procedures, you must obtain IRB approval by submitting the IRB Request for Change in Procedures Form.

You will receive an IRB approval status update within 1 week of submitting the change request form and are not permitted to implement changes prior to receiving approval.

Please note that Walden University does not accept responsibility or liability for research activities conducted without the IRB's approval, and the University will not accept or grant credit for student work that fails to comply with the policies and procedures related to ethical standards in research.

When you submitted your IRB application, you made a commitment to communicate both discrete adverse events and general problems to the IRB within 1 week of their occurrence/realization. Failure to do so may result in invalidation of data, loss of academic credit, and/or loss of legal protections otherwise available to the researcher.

Both the Adverse Event Reporting form and Request for Change in Procedures form can be obtained at the IRB section of the Walden web site or by emailing [irb@waldenu.edu](mailto:irb@waldenu.edu): [http://inside.waldenu.edu/c/Student\\_Faculty/StudentFaculty\\_4274.htm](http://inside.waldenu.edu/c/Student_Faculty/StudentFaculty_4274.htm)

Researchers are expected to keep detailed records of their research activities (i.e., participant log sheets, completed consent forms, etc.) for the same period of time they retain the original data. If, in the future, you require copies of the originally submitted IRB materials, you may request them from Institutional Review Board.

Please note that this letter indicates that the IRB has approved your research. You may not begin the research phase of your dissertation, however, until you have received the **Notification of Approval to Conduct Research** (which indicates that your committee

and Program Chair have also approved your research proposal). Once you have received this notification by email, you may begin your data collection.

Jeff Ford  
Walden University Institutional Review Board  
Email: [IRB@waldenu.edu](mailto:IRB@waldenu.edu)  
Tollfree : 800-925-3368 ext. 1210  
Fax: 626-605-0472  
Office address for Walden University:  
155 5th Avenue South, Suite 200  
Minneapolis, MN 55401

## APPENDIX K: APPROVAL TO USE IES-R

Good luck with your research. Consider this email as approval to use this instrument for your dissertation studies.

---

Daniel S. Weiss, Ph.D.  
Professor of Medical Psychology  
Department of Psychiatry  
University of California San Francisco  
San Francisco, CA 94143-0984  
Phone: 415 476 7557  
FAX: 415 476 7552  
Mail Code: UCSF Box 0984-F

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### CONFIDENTIALITY NOTICE

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At Saturday 08:13 PM 7/21/2007, you wrote:

Dr. Weiss:

I am a PhD student and also President of the leading not for profit in the world for children and adults affected by such vascular birthmarks as hemangiomas, port wine stains, arteriovenous malformations, lymphatic malformations, etc. For 12 years I have been fighting insurance companies to approve treatment of hemangiomas. Insurance companies consider treatment of these often disfiguring benign tumors to be cosmetic. My dissertation is "Assessing the Impact of Hemangiomas on the Family System" will focus on establishing the fact that the diagnosis of a infant with a hemangioma is a traumatic event (IES-Revised) which can cause acute stress or post traumatic stress disorder symptoms (HRQ - the HRQ is the Hemangioma Reaction Questionnaire which is a modification of the SASRQ).

I need permission to use your IES-R for my dissertation. I have referenced you and the co-author throughout my dissertation. Please, may I have permission. I have found a copy of the instrument and how it is measured and the validity and reliability data on the instrument but if you have original documents that you could email me, that would be even better than referencing the website.

Thank you so much for your assistance. My hypothesis will help families because if I can establish that the diagnosis is traumatic and leads to PTSD symptoms, then insurance companies will have to cover treatment and the HRQ can be used by primary care physicians to refer the families for mental health counseling, if needed. No psychometric instrument currently exists to measure the burden of hemangiomas on the family.

**Linda Rozell-Shannon**  
President/Founder  
Vascular Birthmarks Foundation  
[www.birthmark.org](http://www.birthmark.org)  
877-823-4646

Join VBF to Raise Awareness. Tell your own personal birthmark story.  
Sign up today to participate in a Day of Awareness event. Go to  
[www.birthmark.org](http://www.birthmark.org)

## APPENDIX L: APPROVAL TO USE SASRQ

Best of luck with your work. ASD predicts the development of PTSD. I think it would be best to repeat the SASRQ to provide the best evidence of a change in symptoms by using the same measure. I have attached the info you requested. This email serves as approval to use this instrument for your studies.

David Spiegel, M.D.

At 04:43 PM 2/2/2007, you wrote:

David Spiegel, M.D.  
Willson Professor in the School of Medicine  
Associate Chair  
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Dr. Spiegel:

I am the Presiden and Founder of the leading not for profit in the world for children and adults affected by such vascular birthmarks as hemangiomas, malformations, and port wine stains. John Merrick, the Elephant Man had vascular anomalies, such as the lesions we are concerned with.

After 10 years of fighting for families to get the insurance companies to cover treatment of removal, I decided to study for a PhD and use my dissertation to establish a medical necessity for treatment. After much, much, much research and analysis I have determined that if I could modify the Stanford Acute Stress Reaction Questionnaire to a disease specific questionnaire for hemangiomas, this would help to establish a medical necessity for treatment. My plan is to survey 50 mothers before treatment and after treatment and to show a decrease in stress (possibly also use the PSI) and then to establish that PTSD does not go away and that unless they cover treatment they will be paying for a lifetime of therapy. As one of these mothers, I can tell you that 12 years after surgery on my daughter, the pain is still there, as are the nightmares and memories.

I've searched using cleft palates as a comparison but I really need to establish this medical necessity and I think just using the PSI is not enough. I think that since PTSD is a diagnosed disorder that requires treatment that I must get something closer to a clinical diagnosis in order to give weight to the need to remove these massive tumors from the children. Over 85% of the requests for surgery are denied because they say it is cosmetic. These tumors are anything BUT cosmetic.

One of my committee chairs is at UCIrvine and I will be having an international conference there in November (2-3) and I hope to present the results of my pilot.

I need your help. I need the SASRQ or please tell me how to obtain the questionnaire and any supporting documentation.

Do you think I have a good case? Do you think I should use the SASRQ to establish the potential for PTSD and use the PSI as a pre and post treatment test to show how stress goes down after treatment but redo the SASRQ and show that the PTSD is still there? What are your thoughts?

Please look at my website if you get a chance at [www.birthmark.org](http://www.birthmark.org). I am the one in the picture with Laura Bush and also with Hillary Clinton (on the front of the site, scroll down).

My Committee member in UCIrvine is Dr. Stuart Nelson of the Beckman Laser Institute.

*Linda Rozell-Shannon*  
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## CURRICULUM VITAE

***Linda Rozell-Shannon***

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### **Education:**

1972 Rensselaer Jr.-Sr. High School – Honor Society – Regents Diploma  
1976 Art Instruction School, Minn., MN – A.S.  
1979 College of St. Rose, Albany, NY – B.S. – Bus. Admin. – Magna Cum Laude  
1984 Russell Sage College, Troy, NY – M.S. – Health Ed., Summa Cum Laude  
2008 Walden University, Doctoral Candidate, Minn., MN

### **Employment History:**

#### **1995-Present – Vascular Birthmarks Foundation**

Founder and President of the leading not for profit in the world for children and adults affected by a vascular birthmark. Manage a board of 15 members including 3 part-time staff. Author of the only book for parents on this subject, entitled “Birthmarks: A Guide To Hemangiomas and Vascular Malformations.” First non-physician to address the American Academy of Dermatology at their annual millennium conference in San Francisco in March 2000. Member of several task forces on birthmarks, including the American Academy of Pediatric Otolaryngologists—the only non-physician on the task force. Developed website materials and maintain website. Developed numerous programs such as Orphans With Birthmarks. These children are labeled “hard to place, special needs, etc.” and so we network them to an adoptive family in the US and then to a physician who donates services. Also developed “Babies With Birthmarks” which are guidelines recommending that all infants at the well baby check up at 4 weeks be referred to a birthmark specialist and pediatric endocrinologist if the birthmark has changed in any way or has not resolved. These guidelines are being reviewed by 4 major medical academies. Established chapters of VBF in Israel, Europe and soon to be Latin America and India and Canada. Assisted in establishing numerous multi-disciplinary treatment centers in the U.S. Speak around the world on this subject. Co-authored numerous medical journals and papers on this subject. Author of numerous newsletters for the VBF. Design all brochures, pamphlets, newsletters and publications relating to the foundation. Have appeared on national television on the Discovery Channel several times concerning this topic. Developed and maintain website at [www.birthmark.org](http://www.birthmark.org). Answer hundreds of emails each week. Network children/adults to proper physician. Provide physicians with information to establish clinics. Subject of numerous newspaper stories on the subject.

Developed and implemented National Day of Awareness campaign for VBF. Appeared on local television numerous times to promote awareness about vascular birthmarks and tumors.

**Jefferson Award Recipient** – April 2005 local and regional Jefferson Award Recipient for Public Service.

**2007 VOICES Campaign Award** – February 2008 Award Presented by Montel Williams

**1998-Present – Massachusetts General Hospital Vascular Birthmarks Clinic**–

Assisted in establishing clinic for birthmarks in 1998 and am currently the Clinic Coordinator. Do intakes on patients, take photos, prepare letters for physicians, generate patients through website.

**1995-2003 – Albany Medical Center Vascular Birthmarks Clinic**. Assisted in establishing clinic for birthmarks in 1996 and worked as clinic coordinator until 2003 when clinic moved to a private practice.

**1972-Present – NYS Department of Motor Vehicles.**

2001-Present – Highway Safety Program Representative. Division of Vehicle Safety. Analyze and prepare reports dealing with the businesses we regulate. Forms design and maintenance. Prepared scanning and imaging analysis and project plans. Worked on data base design and systems development processes. Used various planning tools and quality development workflows. Prepared numerous reports, manuals, annual reports, guidelines, business rules, work flows, general correspondence and fiscal reports analysis.

**1979-2001 – Program Analysis – Sr. Administrative Analyst**. Responsible for program and legislative analysis, numerous correspondence and reports. Worked on many major project teams. Conducted focus groups, public speaking, conference materials and presentations and designed license and registration documents.

**1992-1994 – Custom Plates Project – Program Assistant**. Assisted in the development of a statewide custom plates program including design, marketing and communications.

**1994-1995 – License Scofflaw Program Manager** – Established the first concerted effort to address the habitual offender. Presented to enforcement agencies, magistrates and national motor vehicle associations.

**1972-1979 – DMV – various offices**. Coordinated first Affirmative Action Program. Developed a Career Opportunities for Women Program. Budget analysis and preparation and was the Loaned Executive for SEFA in 1982. Piloted a statewide wellness program in DMV as part of my graduate degree work.



**Publications** – Numerous journals in the field of vascular birthmarks. Book on Birthmarks. Wrote a chapter in a medical textbook on vascular birthmarks. Author of numerous publications and pamphlets on this subject. Provide information to other organizations for publication, concerning birthmarks.

**Speaking:** - Public speaking for DMV work and extensive public speaking for VBF.

**Teaching Experience:** - Adjunct teaching at Russell Sage College 1984-1985 various courses. Nutrition class for Hudson Valley Community College – guest lecturer series. Guest lecturer Albany Medical College – Dermatology and Surgery rotations concerning vascular birthmarks. Taught numerous bible study courses through Grace Fellowship Church.

**Hobbies/Other Skills:** Small group bible study leader for many years. Developed program in church for children with special needs. Coordinate annual Veteran's Day Visitation Program. Member of a Calypso band. Very active in Ballroom Dancing.

**References:** Ken Trela, DMV, 473-5110 (supervised me for 20 years). Dr. John Pelizza (Russell Sage College Professor) 270-2357; John Burgher (former Vice President of VBF) Inspector General for Worker's Compensation – 473-3839; Dr. Martin Mihm (current mentor) 617-724-1350 – Mass. General Hospital.