Psychological and Functional Outcomes of Treatment for Adolescents with Limb Deficiency Disorders: A Focus on the Family

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ABSTRACT

Psychological and Orthopedic Outcomes of Treatment for Children with Limb Deficiency Disorders: A Focus on the Family

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External fixator treatment (EFT) is an increasingly common yet invasive procedure for adolescents with limb deficiencies, necessitating an intensive rehabilitation regimen that places tremendous demands on the caregivers of adolescents receiving this treatment. This study investigated the role of family functioning and parents' psychological functioning in the psychological adjustment and functional independence of adolescents with limb deficiencies. In addition, differences in psychological adjustment and functional independence were assessed comparing adolescents with limb deficiencies who had received EFT with adolescent with limb deficiencies who had not received EFT. Participants completed standardized questionnaires providing data on medical history, and evaluating family functioning, parents' psychological functioning, child's psychological adjustment, and child's functional independence. The sample was comprised of 46 adolescents with limb deficiencies (36 congenital; 10 acquired), ranging from 11 to 18 years of age. Three multivariate analyses of covariance were used to assess group differences. Six multiple regression analyses were employed to evaluate hypothesized predictors of psychological and orthopedic functioning in this sample. Covariates were determined through preliminary correlation analyses. They included mother's education for CBCL Externalizing and YSR Externalizing analyses; time since last orthopedic surgery in analyses for CBCL Internalizing, as well as child and parent

reports of functional independence; number of orthopedic surgeries in analyses for YSR Externalizing; and, sex in analyses for child reports of functional independence. Adolescents with limb deficiencies in this study did not experience more psychological problems compared to a normative sample. No differences in psychological adjustment and functional independence were found between groups. Results also indicated parental distress and family functioning to be important variables in explaining variability in psychological adjustment; however, this pattern of results varied based on reporter (adolescent versus parent). Results further identified a positive relationship between time since adolescent's last orthopedic surgery and functional independence. These findings indicate a role for psychological assessment and intervention targeting patients, parents, and their families in order to optimize rehabilitation and adjustment for adolescents receiving treatment for limb deficiencies.

CHAPTER 1: INTRODUCTION

The psychological and orthopedic functioning of adolescents with limb deficiencies represents a relatively unexplored area of research. Given that children and adolescents with chronic physical conditions are at an increased risk for behavioral and emotional problems (Lavigne & Faier-Routman, 1992), continued investigation and identification of factors that may influence psychological adjustment is essential. While psychological functioning is typically conceptualized as the ultimate outcome variable in the psychosocial literature on children with chronic physical conditions, it represents only one component of overall adjustment. The degree to which children with chronic physical conditions can function independently may be an equally important component of adaptation. In adolescents with limb deficiencies, the orthopedic impairment they experience may compromise their functional independence, and ultimately compromise their overall functioning. In addition, understanding psychological adjustment and functional independence from a developmental perspective may be particularly important given the likely restrictions in functioning for adolescents with limb deficiencies as they attempt to develop increased autonomy as adolescence progresses. Moreover, orthopedists, orthopedic surgeons, and physiatrists may consider physical functioning as a primary indicator of functional health. As such, in keeping with the multidisciplinary management of adolescents with chronic medical conditions, with the goal of achieving optimal functioning, both psychological and functional physical adaptation was be examined in this study.

Overall adjustment in children and adolescents with chronic physical conditions may be improved by the treatments they receive, designed to sustain life, increase physical capabilities, manage symptomatology and pain, and ultimately improve their quality of life. However, treatment is often invasive, painful, and may require intensive follow-up care, placing tremendous demands on the child's caregivers. For children and adolescents with limb deficiencies, external fixator treatment is being used more commonly to lengthen limbs and correct angular deformities. Reconstruction using this procedure presents the patient and family with a long and stressful process (Morton, 1998). As a result, families' functioning and parents' level of psychological adjustment may be especially important to both psychological and medical outcomes in this population.

The purpose of this study was to evaluate whether adolescents with limb deficiencies are at greater risk than the general population for emotional difficulties. Second, this study investigated group differences in psychological adjustment and functional independence between adolescents with limb deficiencies who had undergone external fixator treatment and those who had not. Third, this research assessed the role of parental adjustment and family functioning in the overall adjustment of adolescents with limb deficiencies, defining overall adjustment as both psychological adaptation and functional independence, as it relates to orthopedic functioning and physical adaptation.

First, a description of limb deficiencies and external fixator treatment will be provided. Next, adolescents' adaptation to chronic physical conditions will be discussed, providing a developmental framework in which to understand psychological adjustment and functional independence. This discussion will include a presentation of two

theoretical models of psychosocial adjustment to chronic pediatric disorders. Following the discussion, the role of family and parental distress as predictors of adjustment in pediatric populations will be addressed. Two theoretical frameworks that address the family context in particular, as well as the empirical literature, emphasizing research on children with limb deficiency disorders will be discussed. In addition, functional independence will be examined as an important indicator of adjustment in pediatric chronic conditions. Finally, the brief literature on psychological and orthopedic outcomes following treatment for limb deficiencies will be reviewed.

CHAPTER 2: LITERATURE REVIEW

Limb Deficiency Disorders

Children and adolescents with chronic physical conditions comprise an estimated 10-20% of the general population (Wallander & Thompson, 1995), however not all of these conditions are debilitating or fatal. A "chronic physical condition" is defined as such when it (1) interferes with daily functioning for more than three months in a year; (2) requires hospitalization lasting more than one month in a year; or, (3) is thought at the time of diagnosis to do either of the above (Pless & Pinkerton, 1975).

Limb deficiency disorders represent one example of a potentially debilitating chronic physical condition affecting children, constituting 5-9.7 of 10,000 live births (Wilson, 1998). The term "limb deficiency" or "limb deformity" incorporates both absence and size reduction of 120 human limb bones, with 205 identified limb deficiency disorders (Wilson, 1998). This group of disorders is etiologically heterogeneous. Limb deformities may be acquired or congenital. Acquired limb deficiencies may be the result of trauma, such as amputations, fractures that do not heal properly, or the result of neuromuscular diseases, such as polio. Examples of congenital conditions include dysplasias, hemimelia, Ollier's disease, Blount's disease (tibia vara), Bowlegs (genu varum), Knock-knees (genu valgum), Rickets, and club foot. (Orthoseek, 2000; Moseley, 1997). In spite of this etiological heterogeneity, the homogeneity of the structural presentation allows the category of limb deficiencies to be defined by symptom condition. Structurally, limb deformities may be categorized as limb length discrepancy or angular deformity (Orthoseek, 2000).

Treatment Approaches for Limb Deficiency Disorders

The aim of treatment for limb deficiencies is to minimize limb length discrepancies and/or angular deformity with the ultimate goal of increasing independent physical function. According to Naudie, Hamdy, Frasier, Morin, and Duhaime (1997), successful management attempts to restore normal weight bearing, limb length, and angulation to achieve a normal gait when walking, for lower limb deficiencies. There are a variety of treatments employed to improve functional health, correct deformities and minimize deficiencies. Shoe raises, step-in prostheses, epiphysiodesis, and femoral shortening may be employed in relatively mild cases, with limb length discrepancies ranging from 2 to 6 centimeters. Epiphyseodesis is a surgical intervention with low morbidity and complication rates that completely and permanently stops physeal growth in the longer leg, ultimately slowing the growth rate in the long leg while allowing the shorter limb to catch up (Moseley, 1990). Femoral shortening may be offered as a treatment alternative to epiphysiodesis when the patient has already reached skeletal maturity, precluding epiphysiodesis, or when a reliable prediction of discrepancy at skeletal maturity cannot be made (Moseley, 1990). Prediction of limb length discrepancy at skeletal maturity is commonly based on the Green-Anderson growth-remaining graph. This graph denotes the growth remaining in the distal femur and proximal tibia of boys and girls as a function of skeletal age (Moseley, 1990). Estimating leg length discrepancy is based on relationships between maturity (skeletal age), stature (limb length), and aging (chronological age). Changes in these factors and the relationships between them vary across children. In particular, children may mature more quickly than they age resulting

in unexpected “growth spurts” that may lead to unreliable discrepancy predictions (Moseley, 1990).

In cases with more severe congenital deficiencies, management is often controversial due to the costs and complications related to the treatment for such conditions (Naudie et al., 1997). Many physicians encourage early amputation to convert the affected limb to a stump suitable for prosthetic fitting (Choi, Kumar, & Bowen, 1990; Mason, 1991). Mason (1991) indicates that while there is some variability in the timing of elective amputation, ideally this surgery is performed around the child’s first birthday to allow the child to reach normal motor development and adjust to the prosthesis. However, lack of a clear diagnosis as to the severity of the deficiency may interfere with decisions regarding early treatment. For fibular hemimelia, the most common deficiency of long bones characterized by the partial or complete absence of the fibula, amputation and prosthetic treatment is generally recommended for discrepancies of 5 to 7.5 centimeters at skeletal maturity and for foot deformity that cannot successfully be made plantigrade by surgical correction (Birch et al., 1999; Naudie et al., 1997). Other guidelines generally indicate amputation for limb length discrepancies greater than 15 centimeters (Moseley, 1990). With appropriate fitting, prosthetic devices can correct malrotation, achieve equal limb length, and increase mechanical stability (Mason, 1991). While amputation typically involves a single surgery with a brief hospital stay and immediate equalization, it is also irreversible and may require additional surgeries to revise the stump. Additional disadvantages to amputation include the resulting lack of normal sensation and proprioception and the periodic replacement and adjustment of prostheses (Naudie et al., 1997).

Severe limb deficiencies may also be managed with external fixator treatment (EFT), typically employed in children with limb deficiencies of 6 to 15 centimeters predicted at skeletal maturity (Moseley, 1990). Limb lengthening by external fixation was first introduced by Codivilla of Bologna approximately 100 years ago. The now commonly used Ilizarov method was developed in the early 1950s by Dr. Gavrill Ilizarov in Siberia, and has been employed in North America since 1981 (Limb Lengthening and Realignment: A Guide for Parents and Families. A.I. duPont Hospital for Children). While EFT can lengthen bones and correct angular deformities, it cannot create bone where none exists or reconstruct deficient structures (Mason, 1991). Consequently this treatment is not available to all children with limb deficiencies.

EFT necessitates surgery during which the bone to be lengthened or rotated is partially cut, a procedure called a corticotomy. External fixator devices are affixed to the bone with pins and a bolt-like apparatus is threaded between the fixators. Following the surgery, the bolts are turned 2 to 4 times per day to lengthen or rotate the bone, a process referred to as distraction. Distraction proceeds very slowly, 1 mm per day for optimal results, so that bone healing occurs as the lengthening or angulation proceeds (Van Der Bauwhede & Sherry, 1996). External fixator treatment employs either a uniplanar fixator, such as the EBI or Garche devices, or ring fixators, such as the Ilizarov device (Van Der Bauwhede & Sherry, 1996).

EFT provides the opportunity for improved independent physical functioning without the loss of limbs and related disadvantages discussed above. However, high complication rates, problems in healing of the bone in particular, were common with the earlier Wagner method of EFT and often interfered with successful achievement of the

initial surgical goal. With the advent of the Ilizarov method, based in the physiology of bone and soft-tissue regeneration under tension-stress conditions, management of complications related to bone healing have improved significantly and treatment goals are typically achieved (Paley, 1990). Nonetheless, concerns regarding other complications remain. Complications that occur during EFT include muscle contractures, neurological or vascular injury, joint luxation, axial deviation, premature or delayed bone consolidation, nonunion, pin site infections, and hardware failure (Paley, 1990). Loss of length, bowing, or re-fracture may also occur late in the EFT, and joint stiffness may remain following the removal of hardware (Paley, 1990). In addition, pain and difficulty sleeping have been reported throughout the EFT process (Morton, 1998; Paley, 1990).

EFT involves a significant and lengthy rehabilitation process. Generally, fixators remain affixed to the bone for 4 to 8 months (Cusack, 1997). During this period, patients are involved in several hours of occupational and physical therapy, 3 to 5 times weekly. In addition, patients and their caregivers must engage in distraction, as well as pin care, three times a day, to ensure that the pin sites, where the fixator is attached to the bone, do not become infected (Jauernig, 1990). Recent innovations in EFT have included automated distraction devices which may relieve some care-giving demands experienced by families. However, this device does not appear to be widely available to date. Following several months of EFT and rehabilitation, a second surgery is often required to remove the fixator device.

Given the costs and benefits of treatments for limb deficiency disorders, particularly those requiring surgical intervention for relatively severe deficiencies, understanding the functional results of treatment, both psychological and physical, may

further guide treatment decision-making for families and doctors. A significant empirical and theoretical literature exists regarding children's adaptation to chronic medical conditions. This literature will be reviewed as a framework in which to understand adjustment of children with limb deficiency disorders. Following this review, research addressing psychological adjustment and functional health specific to the pediatric limb deficiency population will be presented. In addition, factors influencing adaptation will be addressed for both general chronic pediatric conditions and limb deficiencies.

Adaptation to Chronic Pediatric Conditions

Adolescents with chronic conditions, such as limb deficiency disorders, must cope with stresses and responsibilities related to their symptomatology, variations in the course of their illness, and treatment of their conditions over an extended period of time. The demands and stressors inherent in living with a chronic physical condition may compromise the adolescents' overall adjustment. The present study defines adjustment, conceptually related to quality of life, as both psychosocial adaptation and functional independence as it relates to physical adaptation.

Living with a chronic medical condition may present unique challenges for adolescents and their families. Adolescence is a developmental period marked by the process of achieving independence from parents while also maintaining connections with their caregivers (Reed, Richards, Moneta, Holmbeck, & Duckett, 1996). During adolescence, parenting practices must shift in order to be more congruent with developmental changes, such as the adolescent's growing need for autonomy and responsibility (Eccles et al., 1993; Holmbeck, Patkoff, & Brooks-Gunn, 1995). Parents of

adolescents with chronic health conditions may have greater difficulty accommodating their children's growing need for independence. According to Anderson and Coyne (1993), some parents may be hesitant to allow their adolescents control over decision-making, particularly as it relates to medical issues. In addition, parents of adolescents with medical conditions may exert more control and protection in an attempt to assure their child's health and manage their own stress related to parenting an adolescent with chronic illness (Holmbeck et al., 2002). These parents may struggle to encourage the development of autonomy in their adolescents while attempting to protect them from poor health outcomes (Anderson & Coyne, 1993). Moreover, the parent adolescent negotiation around the adolescent's developing independence and control may result in increased conflict, particularly for youth with chronic illness. For example, in sample of 82 mother-adolescent dyads, Miller and Drotar (2003) found more diabetes-related conflict reported by mothers when their adolescents (11-17 years) perceived that they had more autonomy for diabetes-related decision-making than their mothers attributed to them. This research has important implications for individual and family conflict, as well as for adherence to medical regimens, particularly given previous research suggesting decreases in adherence to medical regimens as children move into adolescence (Anderson, Ho, Brackett, Finkelstein, & Laffel, 1997).

From the perspective of the adolescent, increasing cognitive development during this period is likely to be related to the adolescents' growing independence in managing their own conditions, coinciding with their increased needs for autonomy, control, and privacy (Anderson & Coyne, 1993). However, the development of autonomy for adolescents with chronic medical conditions may be inhibited by their reliance on family

members, particularly parents, and medical professionals for support and assistance (Blum, Resnick, Nelson, & St Germaine, 1991).

In the context of this developmental perspective, managing a chronic medical condition clearly presents challenges specific to adolescents and their parents. For adolescents with limb deficiencies, mobility is likely to be limited, contributing to restricted functional independence. This may be especially relevant for adolescents receiving EFT. During treatment and rehabilitation, adolescents may be particularly dependent on their parents, especially during non-weight bearing periods, for self-care, pin site care, and physical therapy. The potential conflict between the developmental tasks of adolescents, particularly the need for autonomy and control, and parents' reluctance to accommodate these needs for adolescents with limb deficiencies provides a framework in which to understand psychological adjustment and functional independence for this developmental stage.

Psychological Adjustment

The psychological adjustment of children and adolescents with chronic physical conditions has long been an important focus of empirical study in the field of pediatric psychology. Epidemiological surveys conducted in the late 1960s consistently revealed a higher proportion of emotional and social problems in children with chronic physical conditions than in healthy children, suggesting that up to 30% of these youths might experience psychosocial maladjustment (Pless & Roughmann, 1971). These results were supported by a more recent, large-scale epidemiological study that found children with

chronic physical disorders to be at two to three times greater risk for psychiatric disorder than were healthy children (Cadman, Boyle, Szatmari, & Offord, 1987).

A meta-analytic study reviewed empirical clinical studies of adjustment in children with chronic physical conditions (Lavigne & Faier-Routman, 1992). Consistent with the epidemiological data, despite general resilience, these children appear to be at risk for the development of psychological distress, including overall adjustment, as well as internalizing, and externalizing symptoms. For example, Wallander, Varni, Babani, Banis, and Wilcox (1989) found that mothers of 50 children (6-11 years) with chronic physical conditions reported their children to have significantly more behavior problems and poorer social competence on average than a large normative sample. Thompson, Hodges, and Hamlett (1990) investigated the psychological adjustment of children and adolescents (6-17 years) with chronic illness. Their results indicated similar levels of worry, poor self-image, and anxiety in the chronically ill group and psychiatrically referred children (Thompson et al., 1990). The results of a meta-analytic review examining psychological adjustment in youths with chronic arthritis revealed that this population appears to be at greater risk for the development of internalizing psychological problems as well as overall adjustment problems (LeBovidge, Lavigne, Donenburg, & Miller, 2003).

Physical Adjustment

The literature on adjustment in children with chronic physical conditions has focused on their psychological and social adaptation while the physical component of adjustment has been largely ignored. This lack of attention is likely due, at least in part,

to the fact that disease and disability parameters have consistently been found to be poor predictors of adaptation (Lavigne & Faier-Routman, 1993). However, the physiological variables previously investigated may not have been sensitive to the important components of physical functioning, such as functional independence.

Functional independence refers to the skills and capability required to function independently in the environment. This capacity is not only related to the structural physical abnormalities associated with a chronic condition. Rather, it reflects the success with which individuals with physical conditions are able to function independently in spite of their physical limitations and related obstacles. This includes the patient's ability to meet the demands of daily living activities, functioning in the community, and communicating with others in an age appropriate manner. Compromised functional independence may represent the fundamental chronic stressor for the child and family (Wallander, Varni, Babani, Banis, DeHaan, & Wilcox, 1989).

In order to comprehensively evaluate the overall adjustment of children with chronic pediatric conditions, it is therefore essential to assess functional independence, in addition to psychosocial adjustment. This may be especially true of youths with orthopedic conditions, such as limb deficiencies, particularly for adolescents for whom restrictions in functional independence may interfere with the pivotal developmental task of developing increased autonomy. Furthermore, the primary goal of medical rehabilitation is to improve patient functioning and independence (Wilkerson, Batavia, & DeJong, 1992). The patient developing optimum functioning despite physical disability is best achieved using an interdisciplinary team approach (DeLisa, 1988). In pediatric settings, an interdisciplinary rehabilitation team is typically comprised a physiatrist,

rehabilitation nurses, a psychologist, a physical therapist, an occupational therapist, an orthopedic surgeon, an orthotist, and a prosthetist (Richards, Elliot, Cotliar, & Stevenson, 1995). In order to ensure children's comprehensive adaptation to their condition, mental health professionals must be able to work in concert with medical and rehabilitation professionals. In this context, functional independence is an additionally important target of investigation for children with chronic conditions.

Relationship between Psychological Functioning and Physical Functioning

The importance of functional physical adaptation is further highlighted in its relationship with psychological functioning. Timko, Stovel, Moos, and Miller (1992) found that a sample of 165 patients with juvenile rheumatoid arthritis (1-18 years) who experienced moderate to severe functional disability had greater psychological problems, and more social and family environment difficulties than those with mild functional disabilities. In addition, orthopedic and psychological functioning were found to be significantly related in a sample of 21 children (10 months-12 years) who had undergone amputation (Herring, Barnill & Gaffney, 1986). Measures of orthopedic functioning in this study, however, did not focus on functional health. Overall, these results suggest that physical and psychological functioning are both significant and related components of overall adjustment for children with chronic pediatric conditions.

Psychological Adjustment and Functional Health in Children with Limb Deficiency Disorders

The literature on children with limb deficiencies has focused primarily on the adaptation of amputees whose limb deficiencies resulted from trauma or disease (Varni & Setoguchi, 1991). Early attempts to examine adjustment in children and adolescents with acquired limb loss focused on social, vocational, and educational achievement. Based on these outcomes, youths with acquired amputations were generally found to be well adjusted (Boyle, Tebbi, Mindell, & Mettlin, 1982; Tebbi & Mallon, 1987; Tebbi, Petrilli, & Richards, 1989). More recent research, employing standardized psychological measures, has found these children to manifest depression, anxiety and loss of self-esteem but significant variability in adaptation has been observed (Varni & Setoguchi, 1991). Varni and Setoguchi (1992) found a sample of children and adolescents (N=111) with congenital and acquired limb deficiencies to be at significantly greater risk for psychological maladjustment. The majority of pediatric limb deficiencies are congenital, however, study samples have generally not reflected this statistic. The psychological research on children with limb deficiencies has increasingly included children with congenital disorders and has focused on identifying the potentially modifiable factors influencing psychosocial adjustment in this population (Rubenfeld, Varni, Talbot, & Setoguchi, 1988; Varni, Rubenfeld, Talbot, & Setoguchi, 1989a, 1989b; Varni & Setoguchi, 1993, 1996)

Few studies have investigated psychological adaptation of children who have undergone external fixator treatment. A study by Hrutkay and Eilert (1990) investigated

the psychological functioning of 22 children who underwent the Wagner technique, an external fixator procedure for tibial or femoral lengthening. They found that 64% of these participants experienced psychological problems that included depression and anxiety symtomatology, noncompliance, dependence, anorexia, suicidal ideation, and self-destructive behavior. Hrutkay and Eilert (1990) did not indicate how long these symptoms persisted.

Morton (1998) studied the psychological adjustment of 56 children and adolescents (1-19 years) who received Ilizarov fixator treatments. Additional variables examined included family psychiatric history, family stressors, and potential for cooperation and compliance with treatment. Measures included the Children's Depression Inventory (Kovacs, 1992), Child Behavior Checklist (Achenbach, 1991), Family Environment Scale (Moos & Moos, 1986), Minnesota Multiphasic Personality Inventory for Adolescents (Hathaway & McKinley, 1989), Parenting Stress Index (Abidin, 1990), Piers- Harris Self Concept Inventory (Piers, 1984), and Revised Children's Manifest Anxiety Scale (Reynolds & Richmond, 1985). However, the questionnaires did not appear to comprise a single battery, and participants and parents did not complete all measures. Although there did not appear to be a control group in this study, Morton (1998) suggested substantial psychological costs of Ilizarov external fixator treatment for patients and their families. First, moderate to severe decline of mental status was reported in almost 50% of the sample. Second, approximately half of the sample reported significant family stressors, hypothesized to exacerbate patients' psychological and behavioral problems. Third, sleep disturbance was reported by more than 25% of the sample (Morton, 1998).

Few other studies have investigated both psychological and functional outcomes for children who received external fixator treatment. Ghoneem, Wright, Cole, and Rang (1996) investigated psychological and functional outcomes for 45 children and adolescents (3-18 years) who had undergone Ilizarov external fixator treatment. Their results indicated that all psychological outcome scores fell within normal limits, and that less than 1% of their patients were referred to a psychiatrist for emotional or behavioral difficulties during the lengthening period. In addition, only 7% of their patients experienced functional limitations in daily activities (Ghoneem et al., 1996). However, aside from patients' depression, psychological adjustment was assessed using a measure that specifically evaluated behavior post hospitalization. A more general measure of psychological adjustment with norms that include children who have not been hospitalized may be more useful in evaluating how well these children are functioning in the general population.

Ramaker, Lagro, van Roermund, and Sinnema (2000) investigated the psychosocial functioning for 26 children and adolescents (6-17 years) 2 months following EFT by Ilizarov device. They did not find significant change in anxiety or depression in this sample, and self-report did not indicate changes in school or work functioning related to the limb lengthening procedure. The psychological measures used in this study were not clearly indicated. In addition, patients' subjective report of physical skill after treatment suggested a lack of functional improvement; however, no psychometrically sound measure was used to assess functional outcomes in this study.

Overall, there appears to be variability in results regarding adjustment in children who have undergone external fixator treatment. However, two studies (Hrutkay & Eilert,

1990; Morton, 1998) suggested that psychological adjustment in this population merits further investigation. In particular, it is important to evaluate risk and resistance factors that may contribute to variability in adjustment. Given the extensive self-care demands placed on the parents of children who receive external fixator treatment, identifying family factors that may be related to the adjustment of children who have undergone this procedure is essential. Understanding how the quality and level of family functioning and parental adjustment may influence the child's adaptation is important, ultimately to design family intervention programs to promote healthy adjustment in youth with limb deficiencies.

Theoretical Models of Adjustment to Chronic Pediatric Conditions

While a clear pattern of increased risk for maladjustment in children with chronic physical disorders has emerged in the empirical literature, considerable variation in the adaptation of these children was also found. As a result, the identification of factors that might contribute to the psychological and social functioning of children with chronic physical conditions, and account for this variation, is critical. A variety of models have been hypothesized to explain how risk factors might increase maladjustment in these children and the way in which protective factors might contribute to healthy adjustment (Lavigne & Faier-Routman, 1993).

Three distinct theoretical models of adjustment to pediatric conditions have been discussed in the literature and are reviewed by Garstein, Short, Vannatta, and Noll (1999). The discrete disease conceptualization assumes that each disorder presents specific physical challenges and socio-emotional sequalae. As such, this model examines

adaptation and functioning of children, parents, and families for a single disorder in an attempt to identify disease-specific contributors to adjustment. The noncategorical approach theorizes that regardless of the condition, children living with chronic illness experience common stressors that may result in psychological and social difficulties. A mixed model considers differences between conditions but also identifies nonspecific disease factors. An empirical investigation of these models indicated support for the noncategorical conceptualization (Garstein et al., 1999).

Within the framework of the noncategorical approach, children with chronic physical conditions are hypothesized to display difficulties in adjustment because they are exposed to nonspecific disease-related factors such as negative life events and related stressors. Chronic disorders produce an ongoing strain for both the children and their parents that require continual readjustment and interfere repeatedly with the performance of ordinary activities (Varni, Pruitt, & Seid, 1998). Proposed noncategorical models of adaptation to chronic pediatric conditions attempt to capture the multiple and intricately related factors that may contribute to adjustment in these children. A brief description of the two predominant models is presented below.

Transactional Stress and Coping Model

Thompson (1985) proposed a transactional stress and coping model, identifying the factors hypothesized to influence how the child and family system adapt to the stress of a chronic physical condition. This model is based on ecological-systems theory. System theory posits (1) that systems are composed of interrelated parts, (2) that change in one part is associated with change in all others, (3) that systems maintain a regular

state of balance, and (4) that systems maintain a balance in periods of change and stability (Hoffman, 1981).

Within the context of systems theory, in Thompson's (1985) model, adjustment is proposed to be a function of the transactions between biomedical, developmental, and psychosocial processes. This framework emphasizes the role of child and family adaptation processes as the primary factors influencing the psychological adjustment of children and their mothers (Wallander & Thompson, 1995). These factors include the type and severity of the child's illness; cognitive processes, such as health locus of control, stress appraisals, and self-esteem; methods of coping; and family functioning (Wallander & Thompson, 1995).

Disability- Stress- Coping Model

Wallander and Varni (1992) presented a similar framework for understanding adjustment to chronic physical conditions. Their disability-stress-coping model outlines factors that are organized into a risk-resistance framework. Risk factors in the model include disease/disability parameters, functional independence in activities of daily living, and psychosocial stressors. Resistance factors include interpersonal variables; socio-ecological factors, such as family environment and family member adjustment; and stress processing (Varni, Pruitt, & Seid, 1998). Identification of modifiable risk and resistance factors can ultimately guide the development of new interventions for children with chronic physical conditions (Varni et al., 1998).

The Thompson (1985) and Wallander and Varni (1992) models address many of the same variables purported to influence children's adaptation to chronic conditions.

Similarities between these models include their identification of socio-ecological factors, intrapersonal factors and stress processing as resistance factors, and psychosocial stressors and disability/ disease parameters as risk factors. In addition to these components, the Wallander and Varni (1992) model outlines treatment factors and functional independence, identified as risk factors, as integral components influencing children's psychological adjustment to chronic physical conditions. Furthermore, Wallander and Varni (1992) included physical health in their definition of adaptation, a dimension of adjustment not addressed in Thompson's model. Notably, the Wallander and Varni (1992) model conceptualizes functional independence to be a risk factor for psychological adjustment whereas the conceptualization of the present study theorizes functional independence to be an outcome variable.

Lavigne and Faier-Routman (1993) presented a meta-analytic review of the available studies that have examined many of the correlates of psychological adjustment outlined in these models. The results indicated empirical support for several components of these theoretical frameworks. In particular, when considered as sets, disease/ disability, family/social, and child variables are significantly related to psychological adjustment. Parent/ family and child/ intrapersonal variables appeared to have the greatest predictive ability while disease/ disability parameters were poorer predictors of adaptation (Lavigne & Faier-Routman, 1993). More recently, research has empirically investigated risk and resistance models of adaptation for youths with sickle cell disease (Brown et al., 2000; Casey, Brown, & Bakeman, 2000). Both studies supported the use of the Wallander and Varni's (1992) conceptual framework for predicting adjustment in children with chronic illness. Specifically, Brown and colleagues (2000) found support

for relationships between caregiver coping and caregiver adjustment, children's locus of control and child adjustment, and stress processing and adjustment. In the Casey et al. investigation (2000), adaptive behavior (functional independence) was related to child adjustment, severity was associated with disability-related stress, competence was related to adjustment, and family cohesion and adaptability was associated with disability stress. For both of these studies, disease severity was not correlated with adjustment. In addition, family functioning was not significantly related to caregiver or child adjustment; however, this finding may be due to the unique cultural characteristics of African American families of children with sickle cell disease, as well as the broad age-range of youth who participated in these studies (5-18 years). According to Casey and colleagues (2000), the nonsignificant relationship between family functioning and adjustment may indicate unique interactions between child and family functioning at different developmental stages. A review of the literature investigating the relationship between family factors and adjustment in children for various chronic medical conditions indicated a significant association, in a positive direction, between family functioning and psychosocial adaptation (Drotar, 1997).

While the Wallander and Varni (1992) model has been expanded to include physical health as a component of overall adjustment (Varni, Pruitt, & Seid, 1998), functional independence has received limited empirical attention as outcome variable. Moreover, investigations of functional independence have generally defined this construct as adaptive behavior, frequently measured by the Vineland Adaptive Behavior Scales (e.g Brown et al., 2000; Casey et al., Wiegner, 2000). The present study

conceptualizes functional independence as an outcome variable and defines it in the context of orthopedic functioning, rather than adaptive behavior.

The models discussed above outline important variables and present hypothesized relationships between these factors that may account for varying degrees of adjustment in children with chronic medical conditions. The current study focuses on the role of parental distress and family functioning in the child's adjustment. As such, the following sections will focus on the significant relationship between socio-ecological factors and children's adjustment to chronic physical conditions. The theoretical and empirical literature addressing the role of family functioning and parental adjustment as predictors of adaptation to chronic pediatric disorders will be reviewed.

Chronic Pediatric Conditions and the Family

Chronic pediatric conditions involve and affect the child's family to varying degrees. Children often require assistance to attend medical appointments, understand their disease and treatment, and implement their treatment regimens. In addition, children are unable to give informed consent for treatment (Kazak, Segal-Andrews, & Johnson, 1995). The family is construed as a recipient of such demands intrinsic to chronic illness and may act to improve or exacerbate those stressors (Masters, Cerreto, & Mendlowitz, 1983). The quality of family functioning, may serve to buffer the child from the disruptions and crises related to chronic illness or, intensify the disruptive effects of stressful events (Hamlett, Pellegrini, & Katz, 1992).

Family Factors in Theoretical Models

Both the Thompson (1985) and Wallander and Varni (1992) models identify family factors as important variables mediating children's adjustment to chronic conditions. Several frameworks focus specifically on the role of the family in adaptation to chronic conditions. Family-focused models developed by Rolland (1990) and McCubbin and McCubbin (1993) are discussed below.

Rolland (1990) proposed that patients and their families experience threat and perceived loss as a result of chronic illness. For example, loss may be experienced as loss of function for the child and change in family roles. Rolland (1990) presents the family systems-illness model in which he outlines three dimensions of illness and families to be assessed in evaluating family adaptation. The first dimension includes psychosocial disease-related factors such as onset, course, outcome, and level of incapacitation. The second dimension consists of the phases of illness, such as diagnosis, crisis, and terminal stage. The third dimension is comprised of family system variables that include beliefs about illness and family functioning. The interaction of these dimensions results in varying levels of adjustment to chronic conditions for children and their families.

McCubbin and McCubbin (1993) developed the resiliency model of family stress, adjustment, and adaptation. Illness is defined as a stressor that places demands on the family process that includes reciprocal relationships between the family's quality and style of functioning, problem-solving and coping mechanisms, appraisals of illness and its severity, and family resistance resources. McCubbin and McCubbin (1993) take into account the course of illness over time and the dynamic nature of the family's process of adjustment in response to different phases of the child's chronic condition.

Empirical Support for the Role of Family Factors in Adjustment

Quality and level of family functioning play an important role in the psychological adjustment of medically healthy children. For example, Billings and Moos (1985) found that a negative family environment was related to the children's psychological distress (N=133). In particular, a high level of child dysfunction was associated with more family-related strains, greater family conflict and less family cohesion (Billings & Moos, 1985).

Research in pediatric populations has revealed a relationship between family and psychosocial adjustment of children with chronic medical conditions. A review of the empirical literature revealed that supportive family relationships predicted increased psychological functioning while negative family characteristics were associated with less competent adjustment and more behavioral symptoms (Drotar, 1997). For example, Perrin, Ayoub, and Willett (1993) found families' interpersonal environments to be significantly associated with children's (7-18 years) psychosocial adaptation across a variety of chronic conditions (N=187). A study by Wallander, Varni, Babani, Banis et al. (1989) indicated that internalizing and externalizing problems in children (4-16 years) with chronic conditions were negatively associated with family cohesion and organization, and positively related to conflict (N=153). In addition, higher levels of social competence were significantly associated with family cohesion and expressiveness (Wallander, Varni, Babani, Banis et al., 1989). Hamlett et al. (1992) found family cohesion and family conflict to be significantly related to mother- reported behavior problems in children (6-14 years) with asthma and diabetes (N=30). Furthermore, well adjusted children with chronic conditions tended to live in families that participated

regularly in outside social and recreational activities and were flexible enough to modify family rules. Conversely, the family environments of children who were less well adjusted tended to be less cohesive, more rigid, more disorganized, more socially isolated, and experienced more conflict (Chaney & Peterson, 1989; Wallander, Varni, Babani, Banis, et al., 1989).

With regard to its relationship to parents' psychological functioning, family functioning was found to be associated with psychological problems in caregivers of children (3-12 years) with cerebral palsy and spina bifida (N=94) (Wiegner, 2000). Results from this investigation indicated that difficulties in adaptively assigning family tasks and role restriction were related to greater distress in caregivers. Family functioning was also found to moderate the relationship between externalizing behavior problems in children (5-17 years) with sickle cell syndromes and caregivers' reports of depression, anxiety, and hostility (N=60) (Ievers, Brown, Lambert, Hsu, & Eckman, 1998). Specifically, for children who were perceived as difficult to manage behaviorally, a higher degree of cohesive and adaptive family functioning protected caregivers from experiencing negative affect (Ievers et al., 1998).

Family functioning has been investigated longitudinally in a limited number of studies on adjustment to chronic pediatric conditions. Kazak and Barakat (1997) found that reports of parenting stress and problematic parent-child interaction during a child's cancer treatment were associated with reports of posttraumatic stress symptoms and anxiety in both parents following treatment (N=29). In another study of adaptation to pediatric cancer, Varni, Katz, Colgrove, and Dolgin's (1996) results indicated a relationship between current family cohesion and expressiveness, and child psychological

and social adaptation at diagnosis, at 6 and at 9 months following diagnosis in a sample of 64 children, 5 to 13 years of age. In addition, a relationship between child adaptation at diagnosis and family conflict was found (Varni et al., 1996). Similarly, family conflict was found to be a risk factor for poor adjustment in youths (19 months- 23 years) who received bone marrow transplants (N=64) (Phipps & Mulhern, 1995). Furthermore, Phipps and Mulhern (1995) found that greater family cohesion and expressiveness were associated with fewer behavior problems and greater social competence in the patient.

Chronic Pediatric Conditions and Parents' Psychological Functioning

Psychological Adjustment in Children

In addition to the quality and style of family functioning as a unit, parents' psychological adjustment is similarly associated with adaptation in children and adolescents. Studies of adjustment in medically healthy youths have demonstrated this relationship. Orvaschel, Walsh-Allis, and Ye (1988) found higher rates of psychopathology in 106 children (6-17 years) of depressed parents when compared to children of parents without a psychiatric diagnosis or with estimated rates of disorder for non-referred children from other studies. Children of parents with recurrent major depressive disorder had more severe psychopathology, indicated by their impaired functioning and need for longer-term psychological intervention (Orvaschel, Walsh-Allis, & Ye, 1988). Billings and Moos (1985) found children ($\leq$ 18 years) of depressed parents (N=57) to have more psychological, behavioral, and physical difficulties than children of parents without depression (N=95), a difference in child dysfunction that persisted when the parents' depression remitted. Similarly, in a sample of 59 participants, children (7-12

years) of parents with anxiety disorders exhibited more emotional distress and poorer social adjustment when compared to children of parents without an anxiety disorder (Turner, Beidel, & Costello, 1987).

Parental maladjustment, depression in particular, is thus an important factor that may interfere with a parent's parenting skills. Varni and Setoguchi (1993) summarized a variety of studies that provide examples of impaired parenting by depressed parents. For instance, depressed mothers were found to display a higher proportion of negative critical utterances toward their children (Conrad & Hammen, 1989), to interact with their children with more critical and physically negative behaviors (Webster-Stratton, 1988), and to be less tolerant of child misbehavior (Panaccione & Wahler, 1986). In addition, depressed parents have been described as less involved with their children and showing increased friction, resentment, and helplessness (Orvaschel, Walsh-Allis, & Ye, 1988).

Just as in medically healthy youth, a relationship between psychological functioning of parents' and children with chronic medical conditions has been discussed in the literature. Furthermore, caregivers' of children with chronic medical conditions experience significant psychological distress due to the demands of caring for chronically ill youngster (Jessop, Riessman, & Stein, 1988). The literature addressing psychological problems in caregivers of children and adolescents with various medical conditions suggests that a high percentage of caregivers fall in the clinical range of general psychological distress when compared to the 10% expected in the general population (Derogatis, 1993). For example, Canning, Harris, and Kelleher (1996) reported 19% of caregivers falling in the clinical range (N=116; varying medical conditions). Davis, Brown, Bakeman, and Campbell (1998) reported that 37% of mothers' caring for children

with congenital heart defects experienced significant psychological distress (N=52). For caregivers of children with cerebral palsy (N=60), spina bifida (N=34), and limb deficiencies (N=27), the percentage of those experiencing poor psychological functioning was 48%, 41%, and 37%, respectively (Wiegner, 2000). In this context, parents' psychological functioning is an important factor in the adjustment of children and adolescents with chronic medical conditions.

Drotar's (1997) review of the literature revealed that poorly adjusted children and adolescents with chronic physical conditions tended to have mothers with more frequent psychological adjustment problems. For example, Daniels, Moos, Billings, and Miller (1987) found parental dysfunction to be a significant risk factor associated with more adjustment problems in youths (1-14 years, age of onset) with rheumatic disease (N=93). Similarly, in a sample of 91 participants (7-17 years) with sickle cell disease, mothers' anxiety was significantly related to behavior problems in their children and adolescents (Thompson, Gil, Burbach, Keith, & Kinney, 1990). In another study, Frank and colleagues (1998) investigated family variables that may contribute to adaptation in a large sample of participants (1-17 years) with JRA (N=107) and insulin-dependent diabetes mellitus (N=114). JRA may be comparable to limb deficiencies by virtue of their orthopedic nature and impairment. Results indicated a relationship between parental distress and adjustment in their children. In particular, mothers' depression was associated with child behavior problems (Frank et al., 1998). The direction of the relationship between children's adjustment and psychological functioning in parents is not yet clear and merits further study. Sawyer and colleagues' (1998) longitudinal study indicated an effect of parental adjustment on the psychological functioning of children (2-

5 years) treated for cancer (N=38). They found that mothers' psychological distress following the child's diagnosis significantly influenced the child's psychological adaptation in the future (Sawyer, Streiner, Antoniou, Toogood, & Rice, 1998).

Physical Adjustment in Children

Although chronic physical conditions typically compromise the physical health and related functional independence of children and adolescents to varying degrees, the psychological research on this population has only recently begun to evaluate psychosocial risk and resistance factors with physical adaptation as the outcome. While the role of physical adaptation or functional independence has not yet been assessed in children with limb deficiencies, the following studies investigated functional and physical outcome variables in other orthopedic and pediatric samples. Daniels et al. (1987) investigated parental functioning, family stressors, and family resources as predictors of physical problems in children (1-14 years, age of onset) with rheumatic disease (N=93). Their results indicated that parental dysfunction was associated with more disease-related physical problems, such as physical disability, morning stiffness, and arthritic pain. In addition, they found that the highest proportion of children with multiple psychological and physical problems were those who experienced high family stressors, low family cohesion, and depressed mothers (Daniels, et al., 1987).

Indicators of physical adaptation vary in the literature from symtomatology (i.e. Daniels et al., 1987) to functional capacity. Functional assessment measures purposeful behavior in interaction with the environment (Halpern & Fuhrer, 1984), and as such, may be more sensitive to changes in adaptation that do not require structural changes. Varni,

Wilcox, Hanson, and Brik (1988) investigated predictors of functional status in participants (4-16 years) with JRA and their mothers (N=23). Their findings suggested that children and adolescents' internalizing and externalizing behavior problems, family functioning, disease activity and chronic musculoskeletal pain were predictive of limitations in activities of daily living, a measure of functional status (Varni et al., 1988).

Boyer, Knolls, and Kafkalas (2000) investigated the relationship between family functioning, functional independence, and posttraumatic stress in a sample of 64 adolescents and young adults (11-24 years) with spinal cord injuries. Their findings indicated that family functioning was significantly related to posttraumatic stress and functional independence. A more recent study investigated posttraumatic stress as a mediator in the relationship between family functioning and functional independence in a sample of pediatric spinal cord injuries. Boyer, Hitelman, Knolls, and Kafkalas (2003) found that lower levels of family functioning contributed to a greater risk of posttraumatic stress that, in turn, related to more impairments in functional independence in a sample of 64 adolescents and young adults (11-24 years). Alternatively, better family functioning appeared to protect participants from posttraumatic stress, and ultimately contributed to greater functional independence. Functional independence in these pediatric spinal cord studies was measured from an orthopedic functional health perspective, using an outcome developed by the Pediatric Orthopedic Society of North America (POSNA). This functional health measure assesses components of physical functioning, including upper extremity function, physical function and sports, and transfer and mobility, rather than adaptive behavior. This measure may be particularly

sensitive as an functional independence measure for children and adolescents with orthopedic conditions.

Family Functioning, Parental Adjustment, and Psychological Adaptation in Children with Limb Deficiency Disorders

The role of family functioning in the adjustment of children with limb deficiencies has received limited empirical attention. Rubenfeld, Varni, Talbot, and Setoguchi (1988) investigated the relationship between family functioning and self-esteem in children (8-13 years) with limb deficiency disorders. Their results indicated a relationship between high family conflict and low self-esteem. Higher organization in the family was related to higher self-esteem. In addition, self-esteem was found to be a significant predictor of depressive symptomatology (Varni, Rubenfeld, Talbot, & Setoguchi, 1989a). Similarly, Varni and colleagues (1989b) found that greater family cohesion, more family organization, and greater moral-religious emphasis were associated with fewer internalizing behavior problems, fewer externalizing behavior problems, and increased social competence. Greater internalizing and externalizing behavior problems were associated with more family conflict (Varni et al., 1989b). In another study, Herring and colleagues (1986) found family stress to influence psychological functioning in a sample of children (10 months- 12 years) with Syme amputations, through its relationship with behavior, self-concept, and IQ.

Varni and Setoguchi (1993) investigated parental psychological adjustment as a predictor of psychological adaptation in children (8-13 years) with limb deficiencies (N=54). They found parental distress and marital discord to be significant risk factors for

poor psychosocial adjustment. Specifically, marital discord, paternal depression and paternal anxiety were related to more child depressive symptomatology, higher levels of trait anxiety, and lower self-esteem.

These studies of family functioning, parental adjustment, and adjustment in children with limb deficiencies establish an important foundation. However, several limitations and gaps in the research must be addressed. The data in these studies generally represent a small single sample, limiting the generalizability of the findings. With the exclusion of the Herring (1986) study (N=22), sample sizes ranged from 41-54 and appear to be collected from the same site, solicited from the UCLA Child Amputee Prosthetics Project. In addition, these studies did not assess participants' functional capacity. Furthermore, the UCLA Amputee Prosthetic Project studies did not address the kinds of treatments that the participants had received. The lack of information regarding the course of treatment prevents evaluation of whether psychological adjustment related more strongly to the orthopedic condition, the treatment procedures, or both. Furthermore, the kind of treatment employed may differentially influence the relationship between factors that have been found to predict adjustment in children with limb deficiencies, such as family functioning and parental adjustment. Moreover, samples in these studies generally did not include adolescent participants. As previously discussed, adolescents living with chronic medical conditions likely experience a unique set of challenges that may contribute to adjustment. This context may be particularly relevant for adolescents with limb deficiencies who may be more likely to experience limitations in mobility and functional independence at a time when increasing autonomy is a crucial developmental task. The present study investigated the role of treatment in the context of

these family factors and the adolescent's overall adjustment, defined as both psychological adjustment and functional independence.

The Role of the Family in the Treatment of Pediatric Conditions

Family members assume a major role in implementing the medical regimen of children with chronic physical conditions. Parents have been found to be primarily responsible for the daily care of their children with chronic illness (Canam, 1993; Gallo & Knafl, 1998). The demands of the child's treatment regimen may lead to changes in the family's level of functioning, restricting or interfering with daily routines and established roles, and may impede successful treatment adherence (LaGreca & Schuman, 1995). A brief review of the literature will illustrate the role of family in the treatment of children with chronic conditions, as well as the effects of treatment regimens on the family.

Treatment adherence following external fixator procedures has received limited empirical attention. Morton's (1998) investigation of psychological factors related to the Ilizarov external fixator procedure indicated that in approximately one third of the sample, noncompliance with exercise occurred. Difficulty adhering to the prescribed treatment regimen following surgery was exacerbated by loss of appetite and sleep, family stressors, and possibly, the presence of premorbid family psychiatric history (Morton, 1998). The effects of demands on the family to implement the extensive treatment regimen following the child's external fixator procedure have not yet been investigated.

Given the scarcity of research on the psychological considerations in the treatment children with limb deficiencies, examples from other orthopedic populations can further illustrate the demands on caregivers to help their child follow prescribed treatment regimens. The degree to which children with chronic conditions can function independently may be related to the demands placed on caregivers. For example, the functional impairment of children with spinal cord injuries places extensive and potentially life-altering demands on their parents. Mulcahey, Smith, and Betz (1994) studied the impact of pediatric spinal cord injury (N=156) on parents' daily routines of work, leisure, and rest. They found that every parent reported an alteration in their routines. Forty percent reported decreased number of hours they worked for income by at least 10 hours a week, with 20% resigning completely from their job to care full-time for their injured child. An increase of at least 20 hours each week in homemaking activities, related to the additional parenting activities required for the injured child was reported by 55% of the sample. In addition, 56% reported a decrease in leisure and self-improvement activities and 57% reported spending less time with friends and relatives.

The effects of caring for a chronically ill child have been investigated in patients with juvenile rheumatoid arthritis. JRA is a chronic disease that requires a daily treatment regimen of anti-inflammatory medications, splints, and specialized exercises (Frank et al., 1998). The symptomatic joint inflammation, pain and tenderness, morning stiffness, and limited mobility result in varying levels of functional impairment that necessitates aide from caregivers to follow the prescribed treatment (Frank et al., 1998). McCormick, Stemmler, and Athreya (1985) investigated mothers' perceptions of the impact of JRA on family stress. They found that the most important predictor of greater family stress was

the number of activities in daily living limited by the child's condition and related functional impairment. Presumably, children's limited independence in daily living places higher caregiving demands on their family members.

The demands facing parents of children and adolescents with chronic medical conditions likely include implementing and ensuring adherence to their offsprings' prescribed treatment regimen. Parents of youths with JRA have indicated difficulties with treatment adherence, reporting problems getting their children to comply with medication and exercise regimens (Hayford & Ross, 1988; Litt, Chusky, & Rosenberg, 1982; Rapoff, Lindsley, & Christopherson, 1985). Given the caregiving roles and demands faced by parents of children with chronic medical conditions, family functioning is an important target of investigation in the context of treatment compliance. Chaney and Peterson (1989) investigated the role of family functioning and medication compliance in 25 youths (7-17 years) with JRA. They found better medication compliance for children in families with high levels of cohesion and adaptability (Chaney & Peterson, 1989). In another study, Bender and colleagues (1998) evaluated the effect of psychological factors on adherence to treatment for 24 children (6-12 years) with asthma. They found that nonadherence increased with the level of family dysfunction. Specifically, this was the case in families where affection was not demonstrated and where expectations and consequences for children's behavior were not clearly communicated (Bender, Milgrom, Rand, & Ackerson, 1998). These results suggest that family functioning may be an important focus of intervention in order to improve treatment compliance. In turn, improved compliance to a prescribed treatment regimen presumably results in greater physical and psychological adaptation to chronic pediatric conditions.

Comparison of Outcomes in Children after External Fixator Treatment and Amputation

McCarthy and colleagues (2000) reported that when physicians and families must decide between amputation and limb lengthening, the condition of the foot, the presence of associated anomalies, bilaterality, the desires of the family, and cultural differences must be considered. While the psychological literature on the adjustment of children with limb deficiency disorders has yet to address differences between children who have undergone external fixator treatment and those who have not, a review of the orthopedic literature revealed three studies that compared limb lengthening by external fixation procedures and amputation. McCarthy, Glancy, Chang, and Eilert (2000) compared orthopedic functioning for 25 children and adolescents who had undergone limb lengthening (5-18 years) or amputation (7 months-2 years) to treat fibular hemimelia. Their findings indicated that while external fixator treatment can achieve good results, children who had undergone early amputation had less pain, had fewer complications, were more active, underwent fewer procedures, and were more satisfied with the results of their treatment (McCarthy et al., 2000). Naudie, Hamdy, Fassier, Morin, and Duhaime (1997) found that the 12 patients (6-24 years) in their study who had amputation to treat fibular hemimelia had significantly fewer hospital admissions and less than half the hospital stay than those 10 patients who underwent external fixator treatment (1-16 years). In addition, the patients who had undergone amputation had fewer clinic visits and missed fewer school days due to treatment. Similarly, Choi, Kumar, and Bowen (1990) found a greater percentage of "satisfactory" results for 32 adolescents and young adults (9 –23 years) who underwent amputation compared to 11 patients who had

received external fixator treatment. However, determination of outcomes was not clearly defined and functioning did not appear to be assessed by standardized measures.

While these studies consistently indicate that amputation is often a better treatment option for fibular hemimelia, psychological adjustment has not been adequately addressed. In a review of the literature on investigating psychosocial adjustment in children with amputations, Tyc (1992) noted the lack of empirically-based interviews of psychopathology in these studies, as well as the use clinical impressions of "satifactory" social contacts, educational and occupation achievement as indicators of psychological adjustment. As a result, comparative psychological outcomes based on sound, standardized measures of children receiving these treatments are not evident. In addition, it is not surprising that youth who have undergone amputation had fewer clinic visits, fewer hospital admissions, and a related fewer number of days missed from school given the relatively circumscribed nature of amputation compared to the ongoing process of external fixator treatment. Furthermore, functional independence has not been investigated as a measure of orthopedic functioning in these studies.

Important research evaluating orthopedic and psychological outcomes for children with limb deficiencies has been conducted. The present study built on the existing literature in several ways. First, this investigation compared psychological adjustment for adolescents who have undergone external fixator treatment with those who have not. To date, studies comparing these groups have yet to evaluate psychological factors. Second, the proposed study evaluated functional independence across these groups. Functional independence refers to the skills and capability required to function independently in the environment. This capacity is not only related to the structural physical abnormalities

associated with the chronic condition. Rather, it reflects the success with which individuals with physical conditions are able to function independently in spite of their physical limitations and related obstacles (Wilkerson et al., 1992). This includes the patient's ability to meet the demands of daily living activities, functioning in the community, and communicating with others in an age appropriate manner. Compromised functional independence may represent the fundamental chronic stressor for the child and family (Wallander, Varni, Babani, Banis, DeHaan, et al., 1989). Furthermore, one of the fundamental goals of medical rehabilitation is to improve patient functioning and independence (Wilkerson et al., 1992). Third, studies investigating adjustment in youth with limb deficiencies have generally not focused on adolescent samples, a population that may yield issues specific to this developmental stage.

Fourth, this investigation built on existing research by evaluating the role of family functioning and parents' psychological functioning on the psychological adjustment and functional independence of adolescents who have undergone external fixator treatment and those who have not. An investigation of these family factors has not yet been conducted in the context of treatment for limb deficiencies. In addition, functional independence has yet to be evaluated in relation to family variables in this population.

Understanding the role of family factors in the psychological and functional adjustment of adolescents receiving treatment for limb deficiencies is important in order to guide the development of interventions to support and promote optimal functioning in this population.

Purpose of the Present Study

The purpose of the present study was to investigate whether adolescents with limb deficiencies experiencing more psychological distress than a normative sample. In addition, this study evaluated whether differences in psychological adjustment and functional independence exist between children with limb deficiencies who have undergone external fixator treatment and those who have not. To date, external fixator treatment has received minimal attention in the empirical literature and the difference between these two groups has not yet addressed psychological adjustment and functional independence. Furthermore, this investigation assessed risk and resistance factors that may account for variability in the adjustment of children with limb deficiencies. These variables included treatment variables, family functioning, and parents' psychological adjustment.

Hypothesis I. In this study, adolescents with limb deficiencies were expected to experience greater psychological difficulties compared to adolescents in a normative sample. The limb deficiency sample consisted of youth who have undergone EFT and those who have not.

Hypothesis II. This study hypothesized that patients who have undergone external fixator treatment (EFT) for their orthopedic condition would differ from patients with the similar conditions who have not undergone EFT. Specifically, the EFT group was expected to show greater functional independence, and lower general psychological difficulties than the TC group (treatment control: patients with comparable conditions but no EFT). The external fixator procedure was expected to correct the limb deficiency disorder to a degree that allowed the adolescent to function more independently, while

salvaging their limbs, than adolescents who had not undergone EFT. While research on adjustment in youth who have undergone amputation or external fixator treatment revealed equivocal results, this study hypothesized that a child whose limb has been salvaged and who ultimately does not require prosthetic support to function would evidence greater overall functioning over the long term.

Hypothesis III. While adolescents with limb deficiencies were expected to experience more psychological problems than a normative sample, variability in their level of psychological adjustment was expected. This investigation proposed that family functioning, parents' psychological adjustment, treatment, and severity of the limb deficiency would be significant predictors of psychological adjustment for youth with limb deficiencies. In addition, these predictor variables were expected to account for variability in the adolescent's functional independence, an indicator of orthopedic functioning.

CHAPTER 3: METHODOLOGY

Participants

Participant Recruitment

Through the patient registry and surgical logs at duPont Hospital for Children (DHC) in Wilmington, Delaware, adolescents, age 11- 18, with limb deficiency disorders were identified for two groups of study. Children with limb deficiency disorders and their parents were eligible to participate as part of the External Fixator Treatment (EFT) group if they had received at least one limb-lengthening/straightening procedure with external fixation within the last 6 years and were no longer in active EFT treatment (even if still followed in the outpatient clinics). Children and their parents were eligible to participate in the Treatment Control (TC) group if they had a diagnosed limb deficiency such as Blounts disease, Genu Varum (bowlegs), Genu Valgus (knock knees), clubfoot, leg length discrepancy, hypoplasia, S/P Polio, Rickets, S/P limb fracture, S/P amputation, they had never received limb-lengthening/straightening procedure with external fixation, and they were not currently receiving EFT. Children and their families were not recruited if they did not speak English and if the child had cognitive deficits.

Based on eligibility criteria described above, 51 families were invited to participate in the study, with one family having 2 eligible child participants for a total of 52 potential participants over an 8-month period. Caregivers were contacted one to two weeks prior to a scheduled orthopedic clinic appointment. Using a pre-constructed telephone script as a guide, the research coordinator approached eligible families by telephone, providing a detailed description of the study and its intent to learn more about

emotional and orthopedic adjustment in children with limb deficiency disorders. If caregivers agreed to participate with their children, the research coordinator scheduled to administer the questionnaires at participants' already scheduled orthopedic follow-up appointments. Potential participants were not approached unless they were already intending to be at the hospital for a scheduled orthopedic appointment.

Fifty participants were successfully recruited of the 52 potential participants. The two non-participants reportedly declined participation because their children were not interested in completing the study. Four of the 50 participants were consented but did not complete or return questionnaires for reasons related to the additional time at their clinic visit required to complete the study (30 to 60 minutes). As such, a total of 46 participants completed the study, including 26 in the EFT group and 20 in the TC group. Data were collected from January, 2003 through September, 2003.

Several issues in recruitment were apparent. Families were only invited to participate in the study if they were visiting the hospital for an existing orthopedic appointment. This approach to recruitment was employed in the interest of convenience for potential participants and to recruit participants who were still being followed for their orthopedic condition, with the goal of increasing participation and completion of the study. As a result, the study yielded an 88% completion rate. However, this procedure also limited the number of potential participants, excluding children who had undergone EFT but were no longer attending orthopedic clinic. Recruitment was further limited for the control group sample, comprised of adolescents with amputations and adolescents whose orthopedic conditions did not typically necessitate a surgery as intensive as EFT, with regard to length of treatment and rehabilitation. Consequently, these children were

not followed in clinic as frequently and were more difficult to capture for recruitment. In addition, the age range of children for this study was limited to 11-18 years. Review of clinic lists for the amputation clinic identified a preponderance of children with congenital limb deficiencies. Children born with congenital amputations generally undergo surgeries, when necessary, as infants and toddlers in order to facilitate prosthetic fitting and training to coincide with normative motor development (Tooms, 1990). As such, many children attending these clinics on a more consistent basis were younger than the study age range and, as such, not eligible for recruitment.

Measures

Demographic & Diagnostic Data

The following information was collected from families by questionnaire (Appendix A): sex, age at time of EFT, date of birth, race, clinical diagnosis necessitating treatment, degree of loss, degree of length and/or angulation discrepancy, number of EFTs of patient, and anatomical site of fixation (if EFT patient), number and type of surgeries (for all EFT and TC patients), number and type of complications from surgery or treatment (e.g., pin infections, etc.; for all EFT and TC patients), number and length of admissions (for all EFT and TC patients). In addition, all participants were asked whether external fixator treatment had been offered as a treatment option, as well as the reasons EFT was or was not employed. Any medical and treatment information that parents were unable to provide was obtained from medical chart reviews.

Similarly, a questionnaire (Appendix B) was used to collect highest grade of schooling completed by patient, mother (or mother-figure) and father (or father-figure); and family income.

Severity

Given the heterogeneous nature of the total sample, the structural severity of limb deficiencies fell into one of four categories: percentage of length achieved, limb length discrepancy, degree of angulation, or degree of limb loss. For participants who had limb length discrepancies and who underwent EFT to lengthen the shorter limb, severity was measured as the percentage of length achieved through EFT. This measurement was advised by a senior attending orthopedic surgeon at DHC in order to account for the proportion of discrepancy and length achieved in different limbs. For example, a 3 centimeter lengthening in an ulna bone differs in severity from a 3 centimeter lengthening in a femur. Data regarding bone length before and after EFT for lengthening, and centimeters of length achieved were obtained from review of medical records, scanograms and X-rays interpreted by a senior attending orthopedic surgeon at DHC. For participants who had limb length discrepancies but who did not undergo lengthening treatment, it was not possible to calculate a percentage of bone lengthened. As such, severity for these participants was measured by centimeters discrepancy between limbs. For participants with angular deformities, degree of angulation was used as an indicator of severity. The Degree of Limb Loss scale, developed by Varni and colleagues (1989b), was employed to capture severity for participants with amputations. Amputations,

depending on site, are assigned numerical values (Table 1). Upper and lower limb loss ratings are summed to obtain a measure of total limb loss.

Table 1. Degree of Limb Loss Scale

Upper Body	Rating	Lower Body
Partial Hand	1	Partial Foot
Transcarpal/ Metacarpal	2	Transtarsal
Wrist Disarticulation	3	Ankle Disarticulation
Below Elbow	4	Below Knee
Elbow Disarticulation	5	Knee Disarticulation
Above Elbow	6	Above Knee
Shoulder Disarticulation	7	Hip Disarticulation
Forequarters	8	Hemipelvectomy
_____	9	Hemicorporectomy

Given that the variation of the structural presentation of participants' limb deficiencies necessitated four measurements of severity, these four measures were converted to z-scores. Z-scores allow for comparisons between scores from different sets of data with different units by converting scores to a common scale. Each participant then received one z-score for severity that was used in data analysis. This z-score reflected either percentage of length achieved, discrepancy in centimeters, degree of angulation, or degree of limb loss, depending on the category in which the participant fell.

Adolescent Psychological Adjustment

The Child Behavior Checklist (CBCL) and Youth Self-Report (*YSR*) (Achenbach & Rescorla, 2001) consist of 113 items that yield the following subscales: Withdrawal, Somatic Complaints, Anxious/Depressed, Social Problems, Thought Problems, Attention Problems, Delinquent Behavior, and Aggressive Behavior. These are summed to create three Total scores: Internalizing behavior problems (INT), Externalizing behavior problems (EXT), and Total behavior problems. The CBCL, which parents complete about their child, is appropriate for children aged 11-18. The YSR is also appropriate for children aged 11 to 18 years of age. Higher scores on both indicate more emotional and behavioral difficulties. The CBCL and YSR internalizing and externalizing scales are used as indicators of parent- and self-reported psychological functioning for participants in this study.

The CBCL and YSR have demonstrated acceptable psychometric properties. Test-retest reliability coefficients range from .80 to greater than .90. Interrater reliablity coefficients range from .59 for parents to .64 for teachers, indicating adequate levels of agreement. In addition, these measures have demonstrated good convergent and discriminant validity (Achenbach & Edelbrock, 1983a; Achenbach & Edelbrock, 1983b).

Although the use of the CBCL and YSR were not specifically designed or recommended for children with chronic physical conditions, Perrin, Stein, and Drotar (1991) reported that this measure has become the most appropriate measurement for the assessment of psychological functioning in children, including those with pediatric conditions. However, Perrin and colleagues highlight important considerations

concerning the use of these measures with medical populations. Of particular importance for the present study is the manner in which social competence is assessed using this measure. The "social competence" component evaluates the child's participation in peer relationships, school, sports, other activities, and household responsibilities (Perrin et al., 1991). Children with limb deficiencies may be less able to participate in such activities, due to the degree of their physical handicap, special transportation requirements, and medical and rehabilitation appointments. Consequently, the Social Competence section would not be an accurate indicator of the child's ability to interact with peers and engage in social activities. Moreover, Social Competence scores may reflect functional independence to some degree, rather than psychological adjustment. As such, this section was excluded for the purpose of this study.

Parent Psychological Adjustment

The *Brief Symptom Inventory* (BSI; Derogatis, 1993) is a 53 item, Likert-type self-report questionnaire designed to evaluate the current psychological symptom status of psychiatric patients, medical patients, and individuals in the community who are not currently patients. Subscales include Somatization, Obsessive Compulsive, Interpersonal Sensitivity, Depression, Anxiety, Hostility, Phobic Anxiety, Paranoid Ideation, and Psychoticism. In addition, the BSI yields the Global Symptom Index (GSI) as a measure of overall psychological functioning. The GSI was used assess to the psychological functioning of parents. Higher scores indicate greater psychological distress.

The BSI has demonstrated adequate internal consistency ranging from .71 for the Psychoticism subscale to .85 for the Depression subscale. Test-retest reliability was

generally strong, with a coefficient of .90 for the Global Severity Index. The BSI has also demonstrated good convergent and predictive validity (Derogatis, 1993). In addition, this measure has been frequently used in studies investigating psychological functioning in caregivers of children with chronic medical conditions (e.g Canning et al., 1996; Davis et al., 1998; Ievers et al., 1998; Wiegner, 2000).

Family Functioning

The *Family Assessment Device* (FAD; Epstein, Baldwin, & Bishop, 1983) is a 60 item self-report measure of family functioning based on the McMaster Model of Family Functioning. This model describes structural and organizational properties and patterns of interaction among family members that have been found to discriminate between healthy and unhealthy families. The FAD identifies seven subscales: Problem Solving, Communication, Roles, Affective Responsiveness, Affective Involvement, Behavior Control & General Functioning. A form is available for each family member (Epstein, et al., 1983). Parent and child reports for the FAD General Functioning scale (FAD Parent; FAD Child) were used in analyses as indicators of overall family functioning.

The FAD is a psychometrically sound instrument. Internal consistency of the seven subscales range from .72 for Roles and Behavioral Control, to .92 for General Functioning (Epstein et al., 1983). Test-retest reliability coefficients ranged from .66 for Problem Solving to .76 for Affective Involvement (Miller, Epstein, Bishop, & Keitner, 1985). In addition, the FAD has demonstrated strong discriminant validity, as well as adequate predictive and concurrent validity (Epstein et al., 1983; Miller et al., 1985). Furthermore, the FAD has been found to be a valid and reliable measure for use in

medical populations (Kabacoff, Miller, Bishop, Epstein, & Keitner, 1990) and has been frequently employed in studies of pediatric populations (e.g Ammerman, Kane, Slomka, Reigel, Franzen, & Gadow, 1998; Bender, et al., 1998; Luescher, Dede, Gitten, Fennell, & Bernard, 1999; Sawyer, et al., 1998; Wade, Taylor, Drotar, Stancin, & Yeates, 1996).

Functional Independence

The *Pediatric Orthopedic Surgeons of North America Pediatric Musculoskeletal Functional Health Questionnaire* (POSNA FHQ; Daltroy, Leang, Fossel, & Goldberg, 1998) is a self-report measure of physical functioning and functional independence (FI) with the following subscales: Upper Extremity Function, Physical Function/Sports, Transfer and Mobility, Comfort, Expectations, and Happy/Satisfied. The instrument has been reported to show excellent internal reliability (>.80), and good to excellent test-retest reliability. Significant correlations between the instrument of interest, physician assessments, and a widely used general function scale indicate the validity of the POSNA FHQ. The POSNA FHQ has forms available for parents to complete regarding children aged 11-18, and for patients' self-report (Daltroy et al., 1998). Parent and child reports on the General Function scale (FI Parent and FI Child) were used in analyses for this study.

Procedure

Parents of adolescents who met inclusion criteria were contacted by telephone to invite them to participate in the study. Families who chose to participate in the study were met at the A.I. duPont Hospital for Children (Wilmington, Delaware) when their children attended their outpatient orthopedic follow-up appointment. At this time, informed

consent was obtained from the adolescent's parent for the adolescent to complete questionnaires, and assent was obtained from the adolescent. In addition, informed consent was obtained from parents so that they might complete questionnaires for themselves. Parents and their children were provided with their questionnaires that were generally completed in approximately 30 to 60 minutes. They were asked to complete the questionnaires independently while they waited for their appointment and return them in envelopes provided before leaving the hospital. Families were also informed that their child's medical chart would be reviewed for information regarding diagnosis and medical complications. Participants did not receive renumeration for their participation in this study.

The present study was approved by the Institutional Review Boards of A.I. duPont Hospital for Children (Wilmington, DE) and Drexel University.

Data Analyses

Data analyses were completed using a computerized statistical analysis program (Statistical Product and Services Solutions: SPSS 11.0 for PC). In order to control for Type I error, as well as address the exploratory nature of this study and concerns regarding low power for the sample size, analyses were based on the standard alpha level of .05. The concern regarding limited power was further addressed through use of preliminary analyses to identify the variables to be included in later analyses, ultimately limiting the number of variables to be included. In addition, MANOVAs were conducted to account for intercorrelation between dependent variables, reducing the number of

analyses. Combining multiple dependent variables into a single multivariate analysis affords greater control over Type I error (Hair, Anderson, Tathan, & Black, 1995)

Preliminary analyses assessed equality of groups for demographic variables that included sex, age, child's current grade, ethnicity, family income, as well as mothers' and fathers' levels of education. Medical factors including severity of limb deficiency, number of orthopedic surgeries, and time since last orthopedic surgery were also examined for equality of groups. Depending on the nature of the variable (continuous or categorical), independent samples t-tests or chi square analyses were conducted to investigate equality between groups for the variables outlined above. In addition, the relationships between demographic and medical variables with psychological and orthopedic measures of adjustment were examined through zero-order correlations in order to identify possible covariates to be included in subsequent analyses.

Dependent measures comprised of scores on the measures of child's psychological adjustment and child's functional independence/ orthopedic adjustment. These criterion variables included CBCL Externalizing Problems (EXT), CBCL Internalizing Problems (INT), YSR Externalizing Problems (EXT), YSR Internalizing Problems (INT), POSNA FHQ parent report (FI Parent), and POSNA FHQ child report (FI Child).

Hypothesis I: Given that no comparison group was available, the psychological adjustment of the limb deficiency sample was compared to the normative sample for the Child Behavior Checklist and Youth Self Report (Achenbach & Rescorla, 2001). A one-sample t- test was performed to assess the mean difference between the limb deficiency sample as a whole and the CBCL/ YSR norms for 2 subscales of these measures:

Internalizing Problems and Externalizing Problems. Two additional one-sample t-tests on the same variables were conducted for the EFT group and the TC group independently.

Hypothesis II: Differences between children with limb deficiencies who had undergone external fixator treatment (EFT group) and children with limb deficiencies who had not received EFT (TC group) were analyzed using 3 multivariate analyses of covariance. For each MANCOVA, Treatment served as the three- level (EFT vs. Amputation vs. Other) independent variable. Initially, group was to be the two-level (EFT vs. TC) independent variable, however, the split between amputation and other treatments in the TC group required analyses to determine whether the control group could be maintained as a single sample. The first MANCOVA examined differences between groups on CBCL INT and CBCL EXT. The second MANCOVA assessed differences between groups on YSR INT and YSR EXT. The third MANCOVA investigated differences between groups for FI Parent and FI Child.

Hypothesis III: Six separate hierarchical multiple regression analyses were conducted to statistically predict the six criterion measures: CBCL INT, CBCL EXT, YSR INT, YSR INT, FI Parent, and FI Child. Hierarchical regression analysis provides data on the unique proportion of variance shared by each newly added variable with the criterion measure (Grimm & Yarnold, 1997). Demographic and medical variables that were found to be significantly associated with the criterion variables for the zero-order correlations were included in the regression analyses on the first step. On the second step, parents' scores on the BSI global symptom index were entered. The Global Functioning scores for child and parent on the FAD were entered on the third step. The predictor variables were entered in the same order for each hierarchical regression analysis.

Entering significant demographic and clinical variables on the first step allowed for the measurement of the additional proportion of variance in each dependent measure accounted for by parents' psychological functioning (GSI) and family functioning (FAD Parent and FAD Child). The BSI Global Symptom Index was entered prior to the FAD General Functioning measure to reflect the theoretical relationship between these two variables.

CHAPTER 4: RESULTS

Participants

Since the TC group was comprised of 13 participants who had undergone amputation and 7 participants who received other treatments for their limb deficiencies, group differences on demographic and medical variables were assessed between these two possible subgroups within the control group. T-tests and chi square analyses did not yield differences within the TC group, indicating that the TC group could be treated as a single No-EFT control group. Table 2 details demographic and medical characteristics for the EFT and TC (No- EFT) groups. In addition, t or χ^2 values are included to denote results from analyses testing differences between the EFT and TC groups for these demographic and medical factors.

Table 2. Demographic and Medical Characteristics of the EFT and TC Groups[a]

Variable	EFT (N=26)	TC (N=20)	t or χ^2 value[d]	p value
Sex			.03	.875
Male	15(58%)	12(60)		
Female	11(42%)	8(40)		

Table 2 (Continued)

Variable	EFT	TC	t or χ^2 value[d]	p value
Ethnicity[b]			4.37	.224
Caucasian	17(65%)	16(80)		
African American	5(19%)	3(15)		
Hispanic American	1(4%)	1(5)		
Native American	1(4%)	0(0)		
Family Income[c]			8.24	.410
< $ 10 000	1(4%)	0(0)		
$10 000- 19 999	0(0%)	1(5)		
$20 000- 34 999	1(4%)	4(20)		
$35 000- 49 999	3(12%)	1(5)		
$50 000- 74 999	8(31%)	2(10)		
$75 000- 99 999	2(8%)	3(15)		
$100 000-124 999	4(15%)	3(15)		
> $ 125 000	4(15%)	3(15)		
Age			.07	.941
M(SD)	14.35(2.08)	14.30(2.10)		

Table 2 (Continued)

Variable	EFT	TC	t or χ^2 value[d]	p value
Grade Level			-.23	.966
M(SD)	8.2(2.20)	8.05(1.90)		
Mother's Years			.38	.703
Education				
M(SD)	13.85(2.54)	14.15(2.82)		
Father's Years			-1.25	.965
Education	14.57(2.91)	14.72(2.66)		
M(SD)				
Severity of Condition			.06	.954
M(SD)	-.01(.94)	-.01(.89)		
Time Since Last			-1.14	.220
Surgery (in months)				
M(SD)	8.63(14.84)	13.74(12.31)		
Number of Surgeries			1.25	.259
M(SD)	3.19(1.70)	2.59(1.85)		

Note. [a] Values indicate frequencies (%) unless otherwise noted

[b] Missing data for 3 participants in EFT group

[c] Missing data for 3 participants in EFT group

[d] No significant differences between groups were found

External Fixator Treatment (EFT) Group

Review of demographic variables indicated that the EFT group was comprised of 15 male and 11 female participants. Mean age was 14.35 years (SD=2.08, range= 11 to 18). The mean grade level was 8.2 (SD= 2.20, range= 5 to 12). There were 17 Caucasian participants, 5 African American participants, 1 Hispanic American participant, and 1 Native American participant in the EFT group. One family reported less than $10, 000 annual household family income, 1 family fell between $20, 000 and $34, 999, 3 families reported income between $35, 000 and $49, 999, 8 families fell between $50, 000 and $74, 999, 2 families reported income between $75, 000 and $99, 999, 4 families fell in the $100, 000 to $12, 999 range, and 4 families reported income equivalent or greater than $125, 000. The mean numbers of years of education in the EFT group was 14.57 (SD= 2.91, range 10- 20) for fathers and 13.85 (SD=2.54, range= 9 to 20) for mothers.

Participants in the EFT group had a mean structural severity score of -.01 (SD= .94, range= –1.68 to 2.02). Mean number of orthopedic surgeries was 3.19 (SD= 1.70, range= 1 to 8). Mean number of months since external fixators were removed was 11.10 (SD= 15.20, range= .50 to 72.00). Time since participants' last orthopedic surgery was a mean of 8.63 months (SD= 14.84, range= .50 to 74.00). External fixator surgery was employed a mean number of 1.31 times (SD= .55, range= 1-2) with a mean number of 1.42 (SD=.58, range= 1-2) external fixators used for treatment. Over the course of treatment, 16 participants (61.5%) were treated with one external fixator, 9 participants (34.6%) were treated with two external fixators, and 1 participant (3.8%) was treated with three external fixators. Nineteen participants in the EFT group (73.1%) underwent EFT

on one occasion, 6 participants (23.1%) underwent EFT twice, and 1 participant (3.8%) underwent EFT three times. Six children (23.1%) in this group were treated with two external fixators simultaneously, with placement on the tibia and femur. For eight children (30.8%) the external fixator was placed on the tibia, 10 children (38.5%) had femural placement, 1 child (3.8%) had placement on the foot, and 1 child (3.8%) had placement on the ulna. All but one participant underwent EFT on lower body limbs. The Ilizarov fixator was used for eighteen participants (69.2%) while the EBI fixator was used with the remaining eight participants (30.8%). The majority of the sample (76.9%) experienced at least one complication associated with EFT. Complications included pin site infections, muscle contractures, and fractures during EFT. Twenty- one adolescents (81%) in the EFT group were diagnosed with congenital limb deficiencies including achondroplasia, dysplasia, fibular hemimelia, Blount's disease, genu valgus, and club foot. Five adolescents in the EFT group were diagnosed with acquired limb deficiencies such as status post fracture limb length discrepancy and genu valgus.

Treatment Control (TC) Group

Analyses of demographic variables indicated 12 male and 8 female participants in the TC group. Mean age was 14.30 years (SD=2.10, range= 11 to 18) with a mean grade level of 8.05 (SD= 1.90, range= 5 to 12). This group comprised 16 Caucasian participants, 3 Hispanic American participants, and 1 African American participant. One participant's family income was reported to be between $10, 000 and $19, 999 annually, 4 families fell in the range of $20, 000 to $34, 000, 1 family reported an income between $35, 000 and $49, 999, 2 families fell between $50, 000 and $74, 999. The $75, 000-

$99, 999, $100, 000-124, 999, and greater than $125, 000 ranges were each comprised of 3 families. The mean numbers of years of education in the TC group was 14.72 (SD= 2.66, range= 12- 19) for fathers and 14.15 (SD=2.82, range= 9 to 18) for mothers.

Review of medical measures indicated a mean severity score of -.01 (SD= .89, range= –1.14 to 3.00) in the TC group. Mean number of orthopedic surgeries was 2.59 (SD= 1.85, range= 1 to 9). Mean time since participants' last orthopedic surgery was 13.74 months (SD= 12.31, range= .75 to 38.00).

In the TC group, 13 participants had amputations with 9 congenital amputations and 4 acquired amputations. The remaining 7 participants in the TC group underwent a variety of alternative treatments for their limb deficiency disorders. For these 7 participants, treatments included epiphyseodesis, shortening of the longer limb, and osteotomy with internal fixation. For participants with amputations, Table 3 outlines frequencies and percentages for amputation sites. The TC group was comprised of 15 adolescents (75%) with congenital limb deficiencies such as fibular hemimelia, absence of the tibia, proximal focal femoral deficiency, osteochondromatosis, and displasias. Five participants (15%) were diagnosed with acquired limb deficiencies including acquired amputation due to pseudoarthrosis, Streeter Band syndrome, meningococcemia, and lawn mower accident.

Table 3. Frequency and Percentage for Sites of Amputations in TC Group (N=13)

Site	N	(%)
Above knee (AK)	1	(7.6)
Below knee (BK)	5	(38.5)
Knee disarticulation (KD)	2	(15.4)
Partial foot (PF)	1	(7.6)
AK, BK, & bilateral elbow disarticulation	1	(7.6)
BK, PF, & partial hand(PH)	1	(7.6)
AK, BK, & PH	1	(7.6)
Hip disarticulation & PH	1	(7.6)

Preliminary Analyses

The sampling distribution for demographic and medical variables of interest, the three predictors (GSI, FAD Parent and FAD Child), and the six criterion variables (CBCL INT, CBCL EXT, YSR INT, YSR EXT, FI Parent, and FI Child) were examined. Statistical tests of normality and examination of histograms and normal probability plots revealed that the GSI was not normally distributed. A LOG transformation of the BSI GSI created a normal distribution of this variable. All other variables met the assumption of normality.

Between group equality (EFT vs. TC) for the following demographic and medical variables were assessed through independent samples t-tests and chi

square analyses: Participant's age, participant's current grade, mother's education, father's education, family income, ethnicity, sex, severity of condition, time since last orthopedic surgery, and number of orthopedic surgeries. No statistical differences were found between groups on any of these factors, indicating group equality with regard to demographic and medical characteristics. Mean scores and standard deviations are reported in Table 4 for independent variables, GSI, FAD Parent, and FAD Child, as well as for outcome measures CBCL INT, CBCL EXT, YSR INT, YSR EXT, FI Parent, and FI Child. In addition, the frequencies and percentage of scores falling in the clinical range (T score $\geq$ 63; 90^{th} percentile) for measures of psychological functioning for children and their parents are presented in Table 5. Parent- and self-reports of psychological adjustment and functional independence were remarkably consistent. Both parent and adolescent reports of functional independence were approximately one standard deviation below the mean for the general population indicating lower levels of functional independence. Notably, the percentages of participants and caregivers falling in the clinical range for psychological distress were generally low and mean scores on measures of psychological functioning for adolescents and their caregivers did not differ significantly from normative mean scores. Family functioning was also found to fall non-pathological range.

Table 4. Means and Standard Deviation Scores by Group

Measure	M(SD) by Group		
	EFT	TC	Total
	N=26	N=20	N=46
Independent Variables			
GSI[a]	48.81(12.49)	49.90(10.03)	49.28(11.38)
FAD Parent[b]	1.85(.51)	1.70(.31)	1.78(.44)
FAD Child[b]	1.85(.38)	1.73(.45)	1.80(.41)
Dependent Variables			
CBCL EXT[a]	48.19(11.06)	51.60(9.55)	49.67(10.46)
CBCL INT[a]	50.46(11.55)	50.40(10.47)	50.44(10.97)
YSR EXT[a]	48.19(11.46)	50.15(8.77)	49.04(10.32)
YSR INT[a]	47.58(11.63)	49.21(10.88)	48.29(11.21)
FI Parent[c]	82.21(11.87)	82.81(15.35)	82.47(13.34)
FI Child[c]	85.55(11.01)	84.93(12.85)	85.28(11.71)

Note. [a] T scores. M= 50, SD= 10

[b] Scores range from 1 to 4. Higher scores equal greater dysfunction. M= 1.84, SD= .43 for nonclinical normative sample. Clinical range $\geq$ 2.

[c] Standardized scores range from 0 to 100. Higher scores equal better functioning. M= 95.15, SD= 7.24 for general population normative sample.

Table 5. Frequencies and Percentage of Clinical T Scores for Parent and Child Measures of Psychological Functioning By Group

Measures	N (%) in Clinical Range by Group[a]		
	EFT	TC	Total
	N=26	N=20	N=46
Child Psychological Functioning			
CBCL EXT	4(15.4%)	2(10%)	6(13%)
CBCL INT	5(19.2%)	2(10%)	7(15.2%)
YSR EXT	3(11.5%)	0(0%)	3(6.5%)
YSR INT	1(2.2%)	1(5%)	2(4.3%)
Parent Psychological Functioning			
GSI	2(7.8%)	1(5%)	3(6.5%)

Note. [a] $T \geq 63$; 90^{th} percentile. Higher T scores equal more psychological problems

Pearson correlation coefficients were completed to identify relationships between medical and demographic variables with outcome measures. The following demographic and medical factors were found to be significantly related to dependent variables. Sex (FI Child r= -.35, p= .019), father's education (CBCL EXT r= -.36, p= .014), mother's education (CBCL EXT r= -.31, p= .038), family income (CBCL EXT r= -.52 , p= .0001; YSR EXT r= -.29, p= .049), number of orthopedic surgeries (YSR EXT r= -.36, p= .023), and time since last orthopedic surgery (FI Parent r= .38, p=.011; FI Child r= .35, p= .023; CBCL INT r= -.35, p= .023). Demographic and medical variables that did not yield

significant correlations with outcome variables included: Participant's age, ethnicity, severity of condition, and treatment type.

Demographic and medical variables were included in MANCOVA and regression analyses to control for their effects on dependent variables. However, in the interest of limiting the number of independent variables in the analyses to maximize power, only those demographic and medical factors found to be significantly correlated with outcomes measures were included in the MANCOVA and regression analyses. Mother's education was included as the most parsimonious representation of other demographic variables for analyses assessing CBCL EXT and YSR EXT. Although father's education and family income were also found to be significantly correlated with these dependent variables, mother's education was significantly correlated with both of these factors (Father's education r=.63, p=.0001; Family income r= .50, p=.0001) as well as with ethnicity (r= -.38, p=.01). In addition, the time since last orthopedic surgery variable was included in analyses for outcome measures CBCL INT, FI Child, and FI Parent, based on significant correlations between this medical factor and each of these dependent variables. Similarly, number of orthopedic surgeries was included a covariate in analyses for the dependent measure YSR EXT, and sex was included for FI Child.

Hypothesis I

In order to determine whether children with limb deficiency disorders experienced greater psychological problems than a normative sample, four one-sample t-tests compared means on the following outcome measures with standardized norms for these scales: CBCL INT, CBCL EXT, YSR INT, and YSR EXT. Analyses were conducted for

each group and the total sample. For both groups (EFT and TC) and the total sample, no differences were found between children with limb deficiency disorders and normative samples (M=50, SD=10) for CBCL EXT, CBCL INT, YSR EXT, and YSR INT. Therefore, contrary to the hypothesis, children with limb deficiency disorder did not experience greater psychological difficulties than the normative sample, regardless of whether they were treated with external fixation or not. Results are presented in Table 6.

Table 6. Means, Standard Deviations, t- test, and p values for each Outcome Measure by Group

Outcomes	Group	M(SD)	t(df)	p
CBCL EXT				
	EFT[a]	48.19(11.06)	-.83(25)	.412
	TC[b]	51.60(9.55)	.75(19)	.463
	Total[c]	49.67(10.46)	-.21(45)	.833
CBCL INT				
	EFT[a]	50.46(11.55)	.20(25)	.840
	TC[b]	50.40(10.47)	.17(19)	.866
	Total[c]	50.44(10.97)	.27(45)	.789

Table 6 (Continued)

Outcomes	Group	M(SD)	t(df)	p
YSR EXT				
	EFT[a]	48.19(11.46)	-.80(25)	.429
	TC[b]	50.15(8.77)	.08(19)	.939
	Total[c]	49.04(10.32)	-.63(45)	.533
YSR INT				
	EFT[a]	47.58(11.63)	-1.06(25)	.298
	TC[b]	49.21(10.88)	-.32(19)	.750
	Total[c]	48.29(11.21)	-1.04(45)	.306

Note. [a] N=26; [b] N=20; [c] N=46

Hypothesis II

To assess for differences between the EFT and TC groups, 3 MANCOVAs were planned to investigate parent report of child's psychological functioning (CBCL INT and CBCL EXT), children's reports of their own psychological functioning (YSR INT and YSR EXT), and parent and child reports of functional independence (FI Parent and Child). However, due to the composition of the TC control group (13 amputations and 7 other varying surgeries), 3 MANOVAs were conducted to assess differences between treatments (EFT, Amputation, and Other), instead of groups, to determine whether the TC group could be interpreted as a single No- EFT group with regard to the variables of interest. If differences on the CBCL, YSR, or FI scores were found between Amputation

and Other, these treatments would comprise two separate treatment control groups. In addition, preliminary analyses previously discussed identified covariates to control for relationships between demographic and medical variables with outcome measures. Mother's education and time since surgery were included as covariates in the analysis of CBCL INT and CBCL EXT. For the analysis investigating differences across treatment for YSR INT and YSR EXT, mother's education and participant's number of orthopedic surgeries were entered as a covariates. Sex and time since last orthopedic surgery was included as a covariate for the analysis assessing differences across treatments for FI Parent and Child.

For all three MANCOVAs, no significant effect for treatment was found (Table 7). Results indicated that the TC group could be assessed as a single No- EFT group. Based on these findings, three additional MANCOVAs were conducted to assess differences between the EFT group and TC (No-EFT) group. No differences in psychological adjustment and functional independence were found between the EFT and TC groups (Table 8). These results did not support the hypothesis that children in the EFT group would demonstrate fewer psychological problems and greater functional independence than children who had not undergone EFT.

Table 7. Multivariate Analyses of Covariates: Differences Between Treatments

Dependent Variables	F	(df)	p
CBCL INT and CBCL EXT	.99	(2,42)	.416
YSR INT and YSR EXT	.13	(2,41))	.971
FI Parent and FI Child	.67	(2,41)	.618

Note. Pillai's trace *F* is reported to address unequal sample size across treatments (EFT N=26; Amputation N=13; Other N=7)

Table 8. Multivariate Analyses of Covariates: Differences Between Groups

Dependent Variables	F	(df)	p
CBCL INT and CBCL EXT	.057	(2,41)	.302
YSR INT and YSR EXT	.004	(2,41))	.927
FI Parent and FI Child	.010	(2,41)	.806

Note. Pillai's trace *F* is reported to address unequal sample size across groups (EFT N=26; TC N=20)

Hypothesis III

Six hierarchical regression analyses were conducted to determine the unique contribution of parents' psychological distress (GSI) and both parent and child reports of family functioning (FAD) on the psychological adjustment and functional independence of children with limb deficiency disorders. The proposed hypothesis included severity of limb deficiency and treatment as additional predictor variables, however, these factors were found to be unrelated to the outcome measures in preliminary zero-order correlation analyses. As detailed in the Preliminary Analyses section, zero-order correlations identified alternative demographic and medical factors to be included in the regression equations. In addition, given that between- group analyses indicated that treatment groups did not differ with regard to outcome measures, variability in psychological adjustment and functional independence were investigated for the total sample.

Hypothesis IIIa

Table 9 details the results of the hierarchical regression analysis for the CBCL EXT. Based on results of preliminary analyses already discussed, to control for the effects of mother's education this variable was entered into the equation at the first step. Theoretically, and based on the existing literature, parents' distress was presumed to pre-date family functioning. As such, in each initial regression model for all six outcome variables, GSI was entered independently immediately before the simultaneous entry of FAD parent and FAD child at the last step.

Following the first step of the prediction equation, the R value was significant and maintained significance through each additional step of the model. Mother's education

accounted for a significant amount of variance in the criterion ($\Delta R^2 = .09$, ΔF (1,44)= 4.56, $p = .038$). Similarly, the GSI entered at Step 2 accounted for a unique proportion of variance in CBCL EXT ($\Delta R^2 = .09$, ΔF (1,43)= 4.71, $p = .036$). The addition of FAD Parent and FAD Child entered on Step 3 contributed to the prediction equation over and above mother's education and the GSI ($\Delta R^2 = .12$, F (2,41)= 3.44, $p = .042$). The entry of all 4 variables at Step 3 accounted for 30.1% of the variance in CBCL EXT scores ($R^2 = .30$, $F(4,41) = 4.41$, $p = .005$).

None of the beta (β) values and partial correlations in the final step were significant. However, on Step 2, prior to the addition of the FAD variables, the β values and partial correlations indicated that mother's education ($\beta = -.33$, $p = .021$) and GSI ($\beta = .30$, $p = .036$) were the most significant predictors of the variance in CBCL EXT. Notably, the contribution of these variables was not apparent when the FAD variables were included in the model. These results indicated shared variance between the predictors included on the final step of this model that may be interfering with the identification of the significant predictors. Further, a variable that has a significant unique contribution to a dependent measure but does not yield a significant β weight (i.e. GSI and Mother's education) is indicative of a possible indirect effect. In this case, GSI and mother's education may be associated with CBCL EXT indirectly through their respective relationships with the FAD variables.

Table 9. Hierarchical Multiple Regression Predicting CBCL EXT

Step	Variable(s)	Partial r with CBCL EXT	β	Δ R^2
1	Mother's Education	-.20	-.19	_____
2	BSI GSI	.16	.15	.09*
3	FAD parent	.24	.27	.12*
	FAD child	.19	.19	

All 4 variables: Multiple R^2 = .30*, Adjusted R^2 = .23*, Multiple R= .55*, df: 4,41

* $p < .05$

Does FAD mediate GSI → CBCL EXT?

Based on the loss of statistical significance in the GSI β coefficient when the FAD variables were added to the model, the FAD variables were identified as potential mediators in the relationship between the GSI and CBCL EXT. A mediator is a variable that accounts for the relationship between the independent and dependent measures. In this case, parents' psychological distress was hypothesized to be associated with child's externalizing behavior problems through its relationship with family functioning. According to Baron and Kenny (1986), in order to function as a mediator, there are three preliminary statistical conditions that must be met: 1) The independent variable and proposed mediator must be significantly related; 2) there must be a significant relationship between the independent and dependent measures; and, 3) the proposed mediator must be significantly associated with the dependent measure. These conditions

are tested through a series of regression analyses (Holmbeck, 1997). For this hierarchical regression model, these conditions were met for FAD Parent but not for FAD Child. As a result, FAD Child was eliminated as a possible mediator.

For the mediator variable to be significant, a fourth and final condition must be met. If the relationship between the independent variable (GSI) and the dependent variable (CBCL EXT) is significantly reduced after controlling for the effects of the mediator variable (FAD Parent) the mediation model will be supported (Holmbeck, 1997). Based on guidelines presented by Holmbeck (1997) and Baron and Kenny (1986), in order to test this final condition, an additional hierarchical regression analysis was conducted forcing GSI and FAD Parent into the model simultaneously at the last step of the model. In this analysis, the GSI β weight fell to a nonsignificant level ($\underline{\beta}$= .14, $\underline{p}$= .360) while FAD Parent yielded a significant β weight ($\underline{\beta}$= .35, $\underline{p}$= .027). The $\underline{F}$ statistics, β coefficients, and $\underline{p}$ values for the regressions testing the necessary conditions for mediation through FAD Parent are presented in Table 10.

Table 10. Statistical Tests of Conditions for Mediation

Condition	F	P	β	P
1) GSI → FAD	8.15	.001	.46	.001
2) GSI→ CBCL EXT	4.83	.013	.30	.036
3) FAD → CBCL EXT	7.53	.002	.42	.003
4) FAD → CBCL EXT (with GSI)	5.92	.003	.354 .139	.027 .360

These results suggest that FAD Parent acts as a mediator between GSI and CBCL EXT; however, Sobel's post hoc equation was conducted to further assess the significance of the indirect association between these variables. This follow-up analysis tests whether the reduction in the relationship between GSI and CBCL EXT is enough to be considered statistically significant (Holmbeck, 2002). Sobel's post hoc analysis involves the calculation of the standard error of the indirect effect. A *z*- score is then calculated by dividing the unstandardized path coefficient (b) for the indirect effect by the standard error (se) of the indirect effect. If the absolute value of *z* is greater than 1.96, the mediation relationship is considered to be statistically significant ($p < .05$) (Holmbeck, 2002). In this case, $z = 1.79$ ($p = .073$), indicating the indirect relationship between GSI and CBCL EXT is not mediated by FAD Parent. These results suggest a

complex interrelationship between these variables that does not appear to be one of mediation.

Does FAD mediate Mother's education → CBCL EXT ?

Mother's education, entered on the first step of the original hierarchical regression, accounted for significantly unique variance in CBCL EXT, however the β coefficient for this variables failed to remain significant with the addition of FAD Parent and Child to the model. In this case, the preliminary statistical conditions for mediation were met for FAD Child but not for FAD Parent. Based on these findings, FAD Parent was not further investigated as a possible mediator. As the mediation model states, if the relationship between Mother's education and CBCL EXT is reduced to non-significance when FAD Child is included in the model, the mediation is supported (Holmbeck, 1997; Baron and Kenny, 1986). To test this, a final hierarchical regression analysis was conducted entering Mother's education and FAD Child into the model simultaneously in the last block. The β coefficient for Mother's education dropped to a non-significant level (β= -.20, p= .185) and a significant β weight was observed for FAD Child (β= .33, p= .027). Sobel's post hoc test was conducted to further assess the significance of FAD Child as a mediator for indirect relationship between Mother's education and CBCL EXT. A z- score of 1.64 (p = .101) was found, suggesting that the nature of the relationship between these variables is not one of mediation. The F statistics, β coefficients, and p values for the regressions testing the necessary conditions for mediation through FAD Child are presented in Table 11.

Table 11. Statistical Tests of Conditions for Mediation

Condition	F	p	β	P
1) Mother's education → FAD	5.47	.024	-.33	.024
2) Mother's Education→ CBCL EXT	4.56	.038	-.31	.038
3) FAD → CBCL EXT	8.31	.006	.40	.006
4) FAD → CBCL EXT with education	5.14	.010	.33 -.20	.027 .185

Hypothesis IIIb

Table 12 outlines the results of the hierarchical regression analysis for CBCL INT. In this model, previously discussed preliminary analyses indicated the inclusion of time since the child's last orthopedic surgery at Step 1. As in the first regression analysis, GSI was entered at Step 2 while FAD Parent and FAD Child were entered simultaneously at Step 3. This model yielded a significant R value when time since surgery was added to the model at Step 1, with this variable uniquely contributing to the prediction model ($\Delta R^2 = .11$, ΔF (1,44)= 5.44, p= .024). At Step 2, the GSI accounted for an additional

proportion of variance in the criterion measure ($\Delta R^2 = .14$, ΔF (1,43)= 7.73, $p = .002$) as did the inclusion of the FAD Parent and FAD Child at Step 3 ($\Delta R^2 = .13$, $F(2,41) = 4.37$, $p = .001$). The entry of all 4 variables at Step 3 accounted for 42.4% of the variance in CBCL INT scores ($R^2 = .37$, $F(4,41)=6.24$, $p = .001$).

The β values and partial correlations for the full model indicated that FAD Parent was the most important contributor to the prediction model ($\beta=.35$, $p=.033$). Of note, as in the CBCL EXT regression model, prior to the addition of the FAD variables the β values and partial correlations indicated that GSI was a significant predictor of the variance in CBCL INT ($\beta=.37$, $p = .008$), as was time since surgery ($\beta = -.321$, $p = .020$). With the addition of the FAD variables in the final step, the effects of GSI and time since surgery were no longer observed. As previously discussed, shared variance between these independent variables, as well as the possible mediating role of FAD in the indirect relationship between GSI and CBCL INT, and time since surgery and CBCL INT, may explain why GSI and time since surgery did not maintain their significance in the last block of the model.

Table 12. Hierarchical Multiple Regression Predicting CBCL INT

Step	Variable(s)	Partial r with CBCL INT	β	Δ R^2
1	Time Since Surgery	-.30	-.26	.11*
2	BSI GSI	.23	.21	.14**
3	FAD parent	.33*	.35*	.13*
	FAD child	.11	.10	

All 4 variables: Multiple R^2 = .37**, Adjusted R^2 = .32**, Multiple R= .62**, df: 4,41

* p < .05; **p< .01

Does FAD mediate GSI → CBCL INT?

As in the previous regression model, GSI's failure to maintain a significant β coefficient when the FAD variables were added to the model suggested that these variables may be potential mediators in the relationship between the GSI and CBCL INT. Parents' psychological distress was hypothesized to be associated with parents' report of child's internalizing problems through its relationship with family functioning. The preliminary statistical conditions for mediation described above were met for FAD Parent but not for FAD Child. As such, only FAD Parent was assessed for its mediational effect. The mediation model states that in order for FAD Parent to be a significant mediator for the indirect relationship between GSI and CBCL INT, the association between GSI (IV)

and the CBCL INT (DV) must be significantly diminished after controlling for the significant contribution of the FAD Parent (mediator variable). Without the inclusion of FAD Parent in the hierarchical model GSI contributed significantly to the prediction model for CBCL INT (β=.37, p= .008). When the hierarchical regression analysis was conducted entering FAD Parent and GSI simultaneously at the last step, the GSI β coefficient did not maintain significance (β=.21, p= .13) and a significant β weight was found for FAD Parent (β=.35, p= .03). Table 13 outlines the F statistics, β coefficients and p values for the regression tests of the conditions for mediation.

Table 13. Statistical Tests of Conditions for Mediation

Condition	F	p	β	p
1) GSI → FAD	5.29	.009	.43	.003
2) GSI→ CBCL INT	7.00	.002	.37	.008
3) FAD → CBCL INT	11.02	.0001	.48	.0001
4) FAD → CBCL INT with GSI	8.27	.0001	.40 .20	.006 .149

Sobel's post hoc equation was conducted to further assess the significance of the mediating role of FAD Parent in the relationship between GSI and CBCL INT. The

mediation model was supported by this analysis (z= 2.28, p= .023). By dividing the indirect path coefficient by the total path coefficient between GSI and CBCL INT, the proportion of variance in this indirect relationship explained by the mediator can be determined. In this case, FAD Parent accounts for 46% of the path between GSI and CBCL INT. These findings indicate that GSI has an indirect effect on CBCL INT through its relationship with the mediating FAD Parent variable. The positive value of these β coefficients suggests that greater psychological distress in parents is associated with more family dysfunction, and greater family dysfunction is related to more internalizing problems in children, reported by parents. Alternatively, lower levels of psychological distress experienced by parents is related to less family dysfunction which in turn is associated with fewer parent reported internalizing problems in their children.

Does FAD Mediate Time since Surgery → CBCL INT?

The loss of significance for the time since surgery β coefficient with the addition of the FAD variables to the model suggested that family functioning may mediate the relationship between the time since surgery and CBCL INT. However, neither FAD Parent nor FAD Child met the preliminary statistical conditions for mediation. Consequently, no further analyses investigating mediation for these variables were conducted. Based on these results, family functioning does not appear to mediate the relationship between time since surgery and parent reports of their children's internalizing problems.

Hypothesis IIIc

Table 14 presents the findings of the hierarchical regression analysis for the YSR EXT. Preliminary analyses suggested that mother's education be entered at Step 1, followed by the number of orthopedic surgeries experienced by the children at Step 2. GSI was added at Step 3 followed by the simultaneous addition of FAD Parent and FAD Child at Step 4. Number of surgeries accounted for a unique proportion of variance in YSR EXT ($\Delta R^2 = .10$, ΔF (1,43)= 5.21, $p = .028$), yielding a significant R value at the second step of this model. Neither the GSI ($\Delta R^2 = .06$, ΔF (1,42)= 3.33, $p = .075$) entered at Step 3, nor the FAD Parent and FAD Child ($\Delta R^2 = .08$, ΔF (2,40)= 2.14, $p = 130$) entered at Step 4 yielded unique contributions to the prediction equation. The β coefficients and partial correlations for the full model indicated that number of surgeries was the most important contributor to the prediction of child's report of their externalizing problems (YSR EXT) ($\beta = -.37$, $p = .009$). In particular, a greater number of surgeries was associated with fewer reported behavior problems. This model did not yield a significant β coefficient for the GSI at any block of the model. The entry of all 5 variables at Step 4 accounted for 28.4% of the variance in YSR EXT scores ($R^2 = .28$, F (5,40)=3.17, $p = .017$).

Table 14. Hierarchical Multiple Regression Predicting YSR EXT

Step	Variable(s)	Partial r with YSR EXT	β	Δ R^2
1	Mother's Education	-.07	-.07	____
2	# Surgeries	-.40	-.39	.10*
3	BSI GSI	.24	.24	.06
4	FAD parent	-.06	.06	.08
	FAD child	.300	.318	

All 5 variables… Multiple R^2 = .28*, Adjusted R^2= .19*, Multiple R= .53*, df: 5,40

* p < .05

Hypothesis IIId

Results from the hierarchical regression model for YSR INT are detailed in Table 15. Preliminary analyses did not indicate significant correlations between demographic or medical variables with this outcome. As such, GSI was added at Step 1 followed by the simultaneous addition of FAD Parent and FAD Child at Step 2. This model yielded a significant R value at Step 1, with GSI uniquely contributing to the prediction model (Δ R^2= .19, ΔF (1,44)= 10.37, p= .002). The FAD variables did not contribute uniquely to the model at Step 2 (Δ R^2= .09, ΔF (2,42)= 2.73, p= .077). In this model, the β coefficients and partial correlations for the full model identified GSI as a significant contributor to the prediction of child's report of their internalizing problems (YSR INT) (β= .35, p=.022). Specifically, with increased parents' psychological distress, children

reported greater internalizing problems. When parents reported lower levels of psychological distress, their children similarly endorsed less internalizing problems. The total model explained 28.4 % of variance in CBCL INT (R^2= .28, F= (3,42)= 5.55, p=.003).

Table 15. Hierarchical Multiple Regression Predicting YSR INT

Step	Variable(s)	Partial r with YSR INT	B	Δ R^2
1	GSI	.35*	.35*	___
2	FAD parent	.14	.16	.09
	FAD child	.21	.21	

All 3 variables… Multiple R^2 = .28**, Adjusted R^2= .23**, Multiple R= .53**, df: 3,42

* p < .05; **p= .01

Hypothesis IIIe

Table 16 outlines the results of the hierarchical regression analysis for child's functional independence reported by parents (FI Parent). To control for significant medical factors, time since surgery was entered into the model at Step 1. GSI was included at Step 2 followed by the FAD variables entered simultaneously at Step 3. A significant R value was observed at the first block of this model, with time since surgery accounting for a unique proportion of variance in FI parent (Δ R^2= .14, ΔF (1,44)= 7.37, p= .009). The inclusion of the GSI at Step 2 additionally contributed to the prediction model (Δ R^2= .13, ΔF (1,43)= 7.73, p= .008) beyond what was accounted for by time

since surgery. The entry of FAD Parent and FAD Child at Step 3 did not significantly predict unique variance in the criterion measure ($\Delta R^2 = .03$, ΔF (2,41)= .89, p=420). The entry of all 4 variables at Step 3 accounted for 30.4% of the variance in FI Parent scores ($R^2 = .30$, F= (4,41)= 4.48, p=.004).

The full model yielded a significant β coefficient and partial correlation for time since surgery, suggesting that this variable was the most important contributor in the prediction equation ($\beta = .37$, p=.009). That is, more time since a child's last orthopedic surgery was related to parents' reports of better functional independence for their children. Alternatively, the less time elapsed since a child's last surgery, the lower their level of functional independence, as reported by their parents. However, parents' psychological distress (GSI) was found to have a significant β coefficient ($\beta = -.36$, p=.008) at the second step of the model, prior to the addition of FAD Parent and FAD Child at Step 3. As previously noted, this suggests both significant shared variance between the factors entered on the last two steps and the possible indirect effect of the GSI on FI Parent, through its relationship with the FAD variables.

Table 16. Hierarchical Multiple Regression Predicting FI Parent

Step	Variable(s)	Partial r with YSR EXT	β	Δ R^2
1	Time since Surgery	.40**	.37**	____
2	GSI	-.24	-.23	.13**
3	FAD parent	-.25	-.27	.03
	FAD child	.06	.06	

All 4 variables… Multiple R^2 = .30**, Adjusted R^2 = .24**, Multiple R= .55**, df: 4,41

* p < .05; **p< .01

Does FAD Mediate GSI → FI Parent?

Similar to regression models for CBCL EXT and CBCL INT, the GSI did not maintain a significant β coefficient when the FAD variables were added to the equation, indicating that family functioning may mediate the relationship between the GSI and FI Parent. Parents' psychological distress was hypothesized to be associated with family functioning that, in turn, was expected to be related to parents' report of child's functional independence. As in the models for CBCL EXT and CBCL INT, the three preliminary statistical conditions for mediation were only met for FAD Parent. Therefore, FAD Child was not further assessed as a possible mediator. F statistics, β coefficients, and p values for the regressions testing the conditions for mediation are presented in Table 17.

Table 17. Statistical Tests of Conditions for Mediation

Condition	F	p	β	P
1) GSI → FAD	5.29	.009	.43	.003
2) GSI→ FI	8.11	.001	-.36	.008
Parent				
3) FAD → FI	6.29	.004	-.29	.037
Parent				
4) FAD → FI	5.88	.002	-.16	.263
Parent with GSI			-.29	.049

As the mediation model states, if the association between the GSI (IV) and the FI (DV) is significantly diminished after controlling for the contribution of the FAD Parent (mediator variable), a mediational effect is supported. When FAD Parent was not entered into the regression equation, GSI uniquely contributed to the prediction model for FI Parent (β= -.36, p= .008). When FAD Parent and GSI were forced into the equation simultaneously, the relationship between GSI and FI parent was not reduced, with GSI maintaining a significant β coefficient (β= -.291, p= .049). The findings from both the initial hierarchical model for FI Parent and subsequent analyses indicate a complex relationship of shared variance between these factors than cannot be explained by a mediation model.

Hypothesis IIIf

The results of the hierarchical regression analysis for the FI Child are presented in Table 18. To control for the variance in the criterion explained by sex, this demographic factor was entered at Step 1, followed by time since surgery at Step 2. GSI was forced into the equation at Step 3 and the FAD variables were entered simultaneously at Step 4. The entry of sex at the first step yielded a significant R value that was maintained at each subsequent step of the model. Sex uniquely contributed to the prediction equation (ΔR^2= .12, ΔF (1,44)= 5.96, p= .019). Time since surgery did not contribute significantly at the second step of the model. Additional unique variance in FI Child was accounted for by GSI at Step 3 (ΔR^2= .09, ΔF (1,42)= 5.27 p= .027) but not by parent and child reports of FAD at Step 3 (ΔR^2= .08, ΔF (2,40)= 2.31, p= .112). When all 5 variables were included at Step 4, the model explained 35.5% of the variance in FI Child scores (R^2= .36, F(5,40)= 4.40, p= .003).

Examination of β coefficients and partial correlations for this model indicated that although time since surgery did not contribute uniquely when entered at Step 2 of the model, this medical factor significantly contributed to the prediction equation when all variables were included in the final block (β= .31, p=.027). In particular, more time since a child's last orthopedic surgery was related to better functional independence as reported by the child whereas less time since a child's last surgery was associated with the child's report of decreased functional independence. As in the regression model for FI Parent, parents' psychological distress (GSI) had a significant β coefficient (β= -.30, p=.027) at the third step of the equation but did not maintain its significance when the FAD

variables were added in the final block. These results suggest a high degree of shared variance between the variables entered on the last two steps, as well as the possible mediating role of FAD on the relationship between GSI and FI Child.

Table 18. Hierarchical Multiple Regression Predicting FI Child

Step	Variable(s)	Partial r with YSR EXT	β	Δ R^2
1	Sex	-.28	-.24	_____
2	Time since Surgery	.34*	.31	.07
2	GSI	-.22	-.20	.09*
3	FAD parent	-.29	-.31	.08
	FAD child	.27	.27	

All 4 variables… Multiple R^2 = .36**, Adjusted R^2 = .27**, Multiple R= .60**, df: 5,40

* $p < .05$; ** $p < .01$

Does FAD mediate GSI → FI Child?

Based on the necessary statistical conditions for mediation, only FAD parent was investigated as a mediator between GSI and FI Child. F tests, β coefficients, and p values for regression analyses testing the conditions of mediation are presented in Table 19.

Table 19. Statistical Tests of Conditions for Mediation

Condition	F	P	β	p
1) GSI → FAD	3.46	.025	.42	.004
2) GSI→ FI	5.46	.003	-.31	.027
Child				
3) FAD → FI	4.97	.005	-.27	.050
Child				
4) FAD → FI	4.49	.004	-.17	.241
Child with GSI			-.23	.121

In the regression analysis testing the final condition necessary to support a mediation model (GSI and FAD Parent entered simultaneously), a diminished relationship between GSI and FI Child was observed. Results from the series of regression analyses suggest that FAD Parent may function as a mediator between these variables (β= -.25, p= .095). However, the β coefficient for FAD Parent was also not significant (β= -.18, p= .232), suggesting mediation may not be the mechanism through which parents' psychological distress and self-reported functional independence are related. Sobel's post hoc test for mediation confirmed that the indirect relationship described above was not mediated by parent reports of family functioning (z = 1.09, p= .276). These results again suggest a complex relationship between FAD Parent, GSI, and FI Child that cannot be explained by a simple mediation model.

Role of Sex in FI Child

The initial regression equation for FI Child indicated that Sex accounted for a significant proportion of variance in children's reports of their own functional independence when no other predictors were included in the model. Follow-up analyses were conducted to further assess the effect of sex on this outcome. Independent one-sample t-tests were run across sex for the following variables: GSI, FAD Parent, FAD Child, and FI Child. Based on these analyses, differences between male and female participants were found only for FI Child ($t(44) = 2.44$, $p = .019$). As a group, boys reported significantly higher functional independence scores than did girls in this sample. Additional independent one sample t-tests indicated that this finding was not due to sex differences in severity ($t(44) = .67$, $p = .505$) or number of orthopedic surgeries ($t(44) = .71$, $p = .338$). Male participants were approximately 6 additional months post surgery compared to female participants; however this difference in time since the last orthopedic surgery across sex was not statistically significant but may be cautiously considered a trend toward significance ($t(44) = 1.47$, $p = .150$).

CHAPTER 5: DISCUSSION

The purpose of the present study was to investigate the psychological functioning of adolescents with limb deficiency disorders for each group (EFT versus TC) compared to a normative sample. In addition, psychological adjustment and functional independence were compared for adolescents with limb deficiencies who had received EFT with those who had not. Based on relationships outlined in the predominant risk and resistance models of adjustment for youth with chronic medical conditions (Thompson, 1985; Wallander & Varni, 1992), this investigation also studied parents' psychological distress and family functioning as important socio-ecological predictors of psychological adjustment and functional independence for adolescents with limb deficiencies.

This study proposed that adolescents with limb deficiency disorders would be at greater risk for psychological problems. However, participants in this study were not found to experience greater psychological difficulties than a normative sample, regardless of treatment. Although the literature on adaptation to chronic pediatric conditions suggests greater vulnerability to psychological maladjustment, this is not the typical outcome (Wallander, Thompson, & Arlickson- Schmidt, 2003). Recent studies have indicated no increased risk for psychological difficulties (e.g Bachanas et al., 2001; Nichols et al., 2000; Reiter- Purtill, Gerhardt, Vannatta, Passo, & Noll, 2003) and families of youth with chronic medical conditions show more similarities than differences when compared to families of healthy children (Kazak et al., 1995). In addition, families of children with pediatric conditions generally function within normal limits, as was observed in this study. Within this framework, the results of this study reflect resilience

for youth with limb deficiencies in the face of the stressors related to living with this condition, rather than psychopathology. Resilience refers to "good outcomes in spite of serious threats to adaptation or development" (Matsen, 2001, p. 228). Accoring to Matsen (2001) resilience is not extraordinary, as initially believed, but rather it is an ordinary phenomenon that generally results from the functioning of basic human mechanisms of adaptation.

In addition, contrary to the hypothesis, no differences in psychological or physical adjustment were found depending on the type of treatment participants received. Orthopedic studies comparing outcomes in youth following EFT and amputation had previously indicated that amputation may result in better functioning when compared to EFT (Choi et al., 1990; Naudie et al., 1997; McCarthy et al., 2000). However, outcomes for these studies focused on hospital admissions, clinic visits, reports of pain and satisfaction with treatment results, and missed school days but did not include standardized measures of psychological adjustment.

Risk and resistance models identify factors that contribute to resilience in youth with chronic medical conditions. In the present study, parents' psychological distress and family functioning were expected to account for variability in adjustment for adolescents with limb deficiencies. This hypothesis was generally supported regardless of treatment received; however, the pattern of results varied for the different criterion measures, depending on reporter of outcomes (child versus parent) and with regard to the influence of medical variables. When parents reported their child's psychological functioning, family functioning was found to mediate the relationship between parents' psychological distress and their children's internalizing problems. Parents' psychological distress and

family functioning were also found to be related to parent-reported externalizing problems; however, the mechanism through which these variables are associated remains unclear. These results are generally consistent with Varni and colleagues' (1989b) findings that indicated higher levels of family functioning to be associated with fewer internalizing behavior problems, fewer externalizing behavior problems, and increased social competence in children with limb deficiencies. Greater internalizing and externalizing behavior problems were associated with more family conflict (Varni et al., 1989b). In addition, Varni and Setoguchi (1993) found parental distress to be significant risk factors for poor psychosocial adjustment. Specifically, marital discord, paternal depression and paternal anxiety were related to more child depressive symptomatology, higher levels of trait anxiety, and lower self-esteem.

In the present study, results differed when children reported on their own psychological functioning. With regard to internalizing problems, parents' psychological distress was found to have the greatest contribution. The number of orthopedic surgeries experienced by participants was most strongly associated with their own report of externalizing behavior problems in a negative direction. With regard to physical adaptation, more time elapsed since the child's most recent surgery was related to greater functional independence, regardless of reporter. In addition, the level of self-reported functional independence was found to differ depending on the sex of the reporter, with male participants reporting a higher level of functional independence as a group. Overall, while parental distress and family functioning appear to play roles in the psychological adjustment and functional independence of adolescents with limb deficiencies, the relationship between these variables and certain outcomes were not clear. Specifically,

parental distress uniquely predicted variability and was significant until the family functioning factors were included in the prediction equations for parent-reported externalizing problems in their children, and parent- and self-reported functional independence. Although these results suggested possible indirect mediational relationships between these predictors and outcomes, mediation models were not supported in these cases. As a result, the mechanism through which parental distress and family functioning were associated with parent- reported externalizing problems, and parent- and self-reported functional independence remains ambiguous.

These findings suggest that disease-related risk factors, such as number of orthopedic surgeries and time since the participants' last orthopedic surgery, generally played a larger role in adolescents' self-reported functioning than for parent-reported functioning in their children. In addition, risk factors explained more variability in parent- and self-reported functional independence in participants, which is notable given the physical foundation of this measure of adjustment. In contrast, socio-ecological resistance factors, such as parental distress and family functioning, generally accounted for more variability in adolescent psychological functioning, except for self-reported externalizing problems. It is important to note, however, that the sample mean scores for parent and adolescent reports of internalizing and externalizing problems fell within normal limits, suggesting significant resilience in response to the stressors of living with a limb deficiency. Within this context of resilience, there is variability in functioning that may be explained, at least in part, by levels of parental distress and family functioning.

Psychological Problems in Adolescents with Limb Deficiencies

The findings of this study suggest a need to identify concerns regarding parent and family functioning in families of adolescents with limb deficiency disorders in order to optimize adaptation. In addition, obtaining information from caregivers and their children may highlight different variables of interest in predicting outcomes of adjustment. Notably, in this study, parents' psychological distress and family functioning appeared to play a significant role in their children's psychological adjustment but the nature of this relationship and its association with other predictors of interest varied depending on whether psychological functioning was parent- or child- reported.

Although regression analysis results varied with parent- and adolescent self-reports of psychological functioning, parent and self-reports were remarkably consistent, yielding statistically significant moderate correlations for internalizing and externalizing problems. These correlations are generally consistent with those reported for Internalizing and Externalizing CBCL and YSR normative data (Achenbach & Rescorla, 2001). Research has suggested higher agreement for parent- and self-reports of observable behaviors with less concurrence for behaviors that represent their children's internal emotional states (Lachar & Gruber, 1993). However, agreement between parent and adolescent reports for internalizing problems was comparable to agreement for externalizing problems for both CBCL and YSR norms (Achenbach & Rescorla, 2001), and in the present study. This finding may be explained by the additional assistance and support from caregivers required by adolescents with limb deficiencies, particularly as they work toward increased functional independence. The resulting time spent together (for example, during physical therapy and frequent doctor visits) may afford parents

greater insight into their children's socio-emotional adjustment. In addition, adolescents may be better able to express internalizing symptoms, compared to children, resulting in greater parental awareness of these problems and higher levels of cross-informant agreement with regard to depression, anxiety, and withdrawal symptomatology, comparable to correlations for observable behaviors.

Parent Reports

Findings indicated that greater emotional distress in parents was related to a higher level of family dysfunction that, in turn, was associated with more parent- reported internalizing problems in their children. Alternatively, parents who reported a lower level of psychological distress also reported better family functioning and less internalizing psychological distress in their children. However, the cross-sectional, correlational nature of this research precludes information on the direction of the above relationships. In addition, the likely reciprocal dynamic of interactions within the family cannot be ignored. Furthermore, the source of the data and possible biases in reporting must be discussed.

There are several possible interpretations of this finding. Parents' experience of psychological difficulties may modify how the family functions, which ultimately may contribute to more anxious, depressive, and withdrawal symptomalogy in their children. For example, a mother who feels depressed and anxious may be less able to appropriately set limits regarding her child's rehabilitation following EFT, and she may be less likely to adequately manage her roles in the family, such as accompanying her child to frequent medical visits while maintaining a job and attending to her other children. Her

relationships with all members of her family are likely to suffer. From a developmental perspective, difficulty in care-giving for a parent experiencing psychological distress may be further exacerbated by the struggle between parent and adolescent, as the adolescent asserts increasing autonomy and the parent attempts to appropriately accommodate developmental changes. Potentially impaired parenting resulting from parental distress may contribute to increased depression and anxiety in the adolescent. It equally plausible that having an adolescent with internalizing problems may lead to greater psychological distress in parents, which may be compounded by the stress of caring for a child with chronic illness. For example, parents may become distressed when they observe their adolescents to be withdrawn and isolated, particularly when parents know that their children's limb deficiencies require them to seek assistance in order to physically negotiate their environments. In addition, parent responsibilities such as frequent doctor visits, overseeing several hours of home-based physical therapy weekly, and pin site care may also contribute to parental distress. Furthermore, an adolescent with more psychological problems may result in greater family dysfunction, or, vice versa. An adolescent who has recently received EFT, living in a family in which there are few structured guidelines regarding adherence to rehabilitation, and where family members are unable to clearly communicate their needs regarding medical support, attention to other children, and time for parents, for instance, may be more likely to develop anxious and depressive symptomatology as a result of the lack of predictability, consistency, and structure in the home. A family functioning in this capacity may also lead to emotional distress in parents. For example, stressors associated with limb deficiency disorders and related treatments, as well as distress in the adolescent, may interfere with optimal family

functioning and contribute to parental distress. In exploring the different possible pathways between parents' psychological distress, family functioning, and adolescent distress it is important to consider that, as a group, parent, family and adolescent psychological functioning fell within normal limits in this study, indicating general resilience to the demands of living with limb deficiencies.

These illustrations indicate how parents' psychological distress, family functioning and adolescents' psychological adjustment are interrelated but they do not clarify the direction of these relationships. In fact, the complexity of these interactions is exacerbated by the reciprocal dynamic and transactional nature of families. Regardless of direction, an emotionally distressed parent of an adolescent with a limb deficiency is likely to experience more distress in response to poor family functioning and in managing an adolescent with psychological problems, which, in turn, may contribute to even more family dysfunction and emotional distress in the child. Strong family functioning may temper the demands of caring for an adolescent with a medical condition, and alleviate distress in the caregiver and child. This may be particularly relevant for adolescents with impaired mobility and functional independence at a time when they are attempting to increase autonomy. A high level of family functioning may also facilitate parents' negotiation of their adolescents' developmental shifts, and vice versa. Alternatively, for a family experiencing significant problems as a unit, psychological distress in parents and children may further increase family dysfunction, especially as they attempt to manage the challenges of limb deficiency treatment and rehabilitation in the context of adolescent development. Furthermore, a child with psychological problems may experience more distress in reaction to diminished parent and family functioning. High levels of parent and

family functioning likely contribute to better management and support of the adolescent with a limb deficiency while affording sensitivity to the adolescent's social and emotional development. Each of these variables likely influences the other and these effects may be understood as reciprocal or circular in nature.

It is notable that family functioning mediates the relationship between parental emotional distress and internalizing problems in their children only when parents reported their own emotional distress, family functioning, and their children's adjustment. Previous research has documented a depressive bias that may influence caregivers' perceptions of their children's psychological functioning. Specifically, depressed and anxious caregivers have a tendency to report elevated levels of psychological problems in their children compared to an independent observer of their children's behavior in similar situations (Boyle & Pickles, 1997; Chilcoat & Breslau, 1997; Najman et al., 2000; Youngstrom, Izard, & Ackerman, 1999). The mediating role of family functioning (parent-reported) in the relationship between parents' self-reported emotional distress and parent reports of their children's internalizing problems should be considered in this context. That is, the results may indeed indicate that adolescents experience more distress when their parents have more psychological problems and the family experiences dysfunction. From a developmental perspective, lower levels of family functioning may be associated with the inherent negotiation and potential conflict between parent and adolescent, as the adolescent asserts greater independence and control, and the parent struggles to accommodate these shifts. This issue may be particularly relevant for adolescents with medical conditions, given some parents' reluctance to afford their adolescents control over decision-making, especially with

regard to medical concerns (Anderson & Coyne, 1993). Alternatively, adolescents' psychological functioning in this context may be more a reflection of their parents' perception than reality. The adolescent tasks of increasing autonomy, control, and privacy should also be considered in interpreting this explanation of results. In this context, as adolescents retreat from parents, parents, particularly those with depressive symptomatology, may perceive normal adolescent behavior as indicative of withdrawal, depression, or oppositional and defiant behavior. While the nature of the relationship between predictors of parent-reported externalizing behavior in their children could not be clearly identified, results did indicate a role for both parents' emotional distress and family functioning in explaining variability in oppositional, defiant, or aggressive behavior for adolescents in this sample.

Adolescent Self-Reports

Family functioning and parents' emotional distress both accounted for variability in adolescents' reports of their own internalizing problems; however, parents' distress was found to have the strongest relationship. Results indicated a direct relationship between parental distress and child-reported internalizing problems, rather than an association mediated by family functioning. A higher level of parental distress related to more self-reported internalizing problems, such as anxiety, depression, and withdrawal. As described above, the cross-sectional, correlational nature of this research does not allow for the determination of direction in this relationship. That is, adolescents who report feeling anxious and depressed may contribute to their parents' reports of emotional difficulties just as parents' reported distress may lead to their children's reported

internalizing problems. In addition, the reciprocal or circular dynamic of this relationship must again be considered; Adolescents' distress may be exacerbated in response to parents' emotional difficulties.

Children's reports of their own symptomatolgy provided data free of the potential parental reporting bias discussed above. In this context, explanations for the relationship between parents' psychological distress and children's internalizing problems are presented. There are two primary interpretations of the directional relationship between parental distress and adolescent internalizing problems. First, parental distress may lead to depression, anxiety, and withdrawal symtomatology in their children. This direction may be explained by impairments in parenting. The empirical literature has indicated that parental distress, particularly depression, may impede appropriate parenting. The interactions of depressed parents with their children have been characterized by less involvement, more critical utterances, and more physically negative behaviors (Conrad & Hammond, 1989; Orvachel et al., 1988; Webster-Stratton, 1989). These patterns of interaction may contribute to an adolescents' own reported experience of depression, anxiety, and withdrawal. The association between parent and adolescent psychological problems may also be explained, at least in part, by the biological transmission of mental illness (Najman et al., 2000). Second, caring for an adolescent with internalizing problems may lead to distress in the parent. This distress may be exacerbated by the demands and stressors of caring for an adolescent with a limb deficiency. Regardless of direction, in the context of this significant relationship, parents and adolescents were generally well-adjusted in this study.

Previously discussed risk and resistance models of adaptation to pediatric conditions (Thompson, 1985; Wallander & Varni, 1992) identify additional factors that may further explain the relationship between parents' psychological distress, family functioning, and psychological adjustment in the adolescent. In particular, parent and child cognitive processes (e.g self-esteem, health locus of control, stress appraisals), coping strategies (e.g palliative, adaptive, pain coping), intrapersonal factors (e.g temperament, competence), and additional social ecological factors (e.g social support) have been found to play a role in the adaptation of youth with chronic medical conditions. For example, Rubenfeld et al.'s (1988) investigation of the relationship between family functioning and self-esteem in children with limb deficiency disorders found high family conflict to be related to low self-esteem whereas higher organization in the family was related to higher self-esteem. In a related study, self-esteem was significantly related to children's depressive symptomatology (Varni et al., 1989a). Additionally, in a sample of children with limb deficiencies, perceived classmate social support was associated with less trait anxiety, less depressive symptomatology, and increased self-esteem (Varni et al., 1991). Casey and colleagues (2000) found children's competence to be related to adjustment in a sample of children with sickle cell disease. Furthermore, for children with sickle cell disease, children's internal health locus of control was related to their own adjustment, while coping strategies of primary caregivers was associated with caregiver adjustment. Research investigating parental distress and adjustment in children and adolescents with JRA has similarly found parents' passive and avoidant coping strategies to be associated with more psychological problems in parents. Parental distress was in turn related to children's psychological functioning (Frank et al., 1998; Timko et al.,

1992). These studies highlight additional pathways and variables through which the relationship between parental distress, family functioning, and child adjustment may be explained.

The combined variables of number of orthopedic surgeries, parental distress, and family functioning were found to account for variability in children's reports of their own externalizing problems; however, the parent and family factors did not uniquely explain variability in this outcome measure. In this case, the number of orthopedic surgeries experienced by the child was significantly related to the child's report of externalizing problems. Specifically, fewer surgeries were associated with more behavioral difficulties, such as oppositional and aggressive behavior. In the context of risk-resistance models of adjustment in children with chronic illness (Thompson, 1985; Wallander & Varni, 1992), the number of orthopedic surgeries represents an important risk factor in explaining variability in self- reported externalizing problems in adolescents with limb deficiencies whereas resistance factors (parental distress and family functioning) did not appear to be as significant.

There are several possible explanations for this result. Children who undergo numerous surgeries may be less able to display externalizing behaviors given the physical limitations associated with surgical interventions. In addition, children who experience more surgeries may habituate to related demands and stressors and, consequently, they may not display psychological and behavioral difficulties. Alternatively, children who undergo more surgeries may not perceive their behavior as inappropriately oppositional, defiant, or aggressive, in the context of the demands and stressors of surgery and hospitalization. As such, they would not endorse symptoms of externalizing problems.

Number of orthopedic surgeries was not related to parents' report of their children's externalizing behaviors. That is, the number of surgeries did not appear to influence parent reports of their children's oppositional, defiant, or aggressive behavior. Children may perceive their externalizing behavior as inappropriate based on discipline and consequences from their parents. It is possible that parents may be less likely to discipline or set limits for children who undergo frequent surgeries. The empirical literature suggests that child-rearing practices for children with chronic medical conditions may differ from those for healthy children. For example, Barlow and colleagues (1998) found that parents of children with JRA reported being overprotective, overinvolved, fostering dependence, and spoiling their children. In another study of children with JRA, fathers reported difficulty punishing their children (Gerhardt et al., 2003). Similarly, mothers of children with insulin-dependent diabetes mellitus and cystic fibrosis were found to be significantly less likely to set limits for their children's behavior when compared to mothers of healthy children (Ievers, Drotar, Dahms, Doershuk, & Stern, 1994). In a study investigating attributions and responses to descriptions of the misbehavior of children with physical and emotional conditions, the misbehavior of children with medically explained pain was rated as less intentional, more excusable, and due to causes outside of the children themselves (Walker, Garber, & Van Slyke, 1995). Given this context, adolescents with limb deficiencies may be treated more leniently by their parents with regard to limit setting and discipline, and as a result, may not perceive their behavior to be inappropriate. However, further research is needed to investigate this hypothesis. This research may be particularly useful given the demands of rehabilitation during and following EFT. Additionally, the question of whether the degree to which parents'

consistently set limits may be associated with improved compliance to medical and rehabilitation regimens requires investigation. Moreover, given that disease-related risk factors (i.e. number of surgeries) may interfere with behavioral functioning, consistent limit setting, consequences and structure post treatment may be particularly important for adolescents with limb deficiencies.

Functional Independence in Adolescents with Limb Deficiency Disorders

For both parent and child reports of functional independence, the combined variables of time since surgery, parents' psychological distress and family functioning explained a significant amount of variance in the outcome; however, for both reporters time since last surgery was the most important predictor of functional independence. As would be expected, time elapsed since the child's last surgery was associated with greater functional independence, according to both parents and children. Less time since surgery was related to lower levels of functional independence. Time since surgery constitutes a disease- related risk factor in understanding adaptation to pediatric conditions, as outlined by the predominant risk- resistance models (Thompson, 1985; Wallander & Varni, 1992). These findings therefore suggest time since surgery to be a significant risk factor for functional health outcomes. In contrast, the roles of socio-ecological resistance factors were not as significant in explaining variability in physical adaptation.

When functional independence was self-reported, outcomes varied based on participants' sex. Male participants' self-reports indicated better functional independence than female participants. There is a lack of previous research to support this finding, particularly given that functional independence was not assessed in other studies of

children with limb deficiencies. In this study, the average additional six-months post surgery for the group of male participants may explain, at least in part, their higher reported level of functional independence. While this finding was not statistically significant and must be interpreted with caution, a larger sample size would have yielded greater power to detect differences across sex.

The Relationship between Parents' Psychological Distress and Family Functioning

For four regression analyses, parents' psychological distress was found to be a significant contributor to the prediction models for parent-reported child internalizing and externalizing behavior problems, as well as child and parent reports of functional independence, until family functioning variables were added to the equation. This finding suggested significant shared variance between these predictors and each respective outcome. As previously discussed, only in the case of parent reports of their children's internalizing problems could this relationship be explained by mediation. For parent reports of their children's externalizing problems, and parent- and child- reported functional independence, the relationships could not be explained by mediation. These variables are significantly related to each other, however, not only the direction but the mechanism through which they are related are unclear.

The relationship between parents' psychological distress and family functioning may be further complicated by important demographic variables. In particular, mother's education was found to explain a significant amount of variability in parent-reported psychological adjustment of their children, specifically externalizing behavior problems. However, the contribution of mother's education was diminished when parent and family

functioning variables were included. These findings suggest relationships between all of these factors. In addition, mother's education was associated with father's education, family income, and ethnicity, and as such, was used as an indicator of socioeconomic and cultural variables. As reviewed by Gorman-Smith and colleagues (2000), research investigating these demographic variables as well as parent and family functioning indicates that, regardless of ethnicity, economic hardship is associated with greater psychological distress; however, the effect appears to be greater in African Americans compared to Anglo Americans. Link and Phelan (1995) reported lower education and lower socioeconomic status to be associated with less supportive family environments, a lower degree of social integration, and increased depressed mood. In addition, low socioeconomic status compounded by having a health problem has been found to be associated with increased distress (Willis, 2002). Individual and family distress have been found to be exacerbated by diminished health status, decreased access to health care, and inequality in medical treatment common in low socioeconomic status and low income populations (Dressler, 1993; Navarro, 1990). Furthermore, particular components of family functioning have been found to vary across ethnicity in low-income populations. Gorman-Smith and colleagues (2000) reviewed research that found less consistent, less involved, less supportive, and less sensitive child-rearing practices in parents of lower socioeconomic status. Such parenting practices as well as stricter, more harsh, and more controlling approaches to discipline may be more likely to be associated with oppositional, defiant, or aggressive behavior in children, depending on ethnicity (Deater-Deckard, Dodge, Bates, & Pettit, 1996). Moreover, ethnicity, independent of socioeconomic status, was found to moderate parent and family outcomes in children

with traumatic brain injury, with greater distress for African American parents 6 to 12 months post- injury, compared to Anglo Americans (Yeates et al., 2002). Taken together, these findings suggest complex interrelationships between socioeconomic and cultural variables, emotional distress, and family functioning, particularly in the context of medical conditions.

For this sample, the relationships between distress, family functioning, and socioeconomic and cultural factors are illustrated in the following case example: Rodney W. is a 12 year-old African American boy who lives at home with his parents and three siblings. Rodney is diagnosed with Blount's disease and was treated with EFT. He has recently had his external fixator removed but he continues to require physical therapy, twice weekly. Mrs. W. has reported feeling overwhelmed in managing Rodney's rehabilitation in addition to meeting the needs of her other three children and working 40 hours a week at a minimum wage job. Mr. W. works frequent double-shifts as a security guard. Mr. and Mrs. W., both high school graduates, work long hours to earn enough money to pay their bills. However, the extra fuel costs required for frequent trips to the hospital for follow-up appointments and extra daycare expenses for Rodney's siblings while Rodney and his mother attend these appointments has added significant financial burden. In addition, Rodney's government-funded medical insurance requires referrals for x-rays, requested for each orthopedic appointment. Mrs. W. indicated that she often forgot to obtain referrals because she was so busy with work, and caring for Rodney and his siblings. On a few occasions, she decided to cancel Rodney's medical visit rather than attend without the necessary paperwork. Mrs. W. reported feeling depressed and unable to elicit support from her husband in managing the children, particularly around

Rodney's medical appointments and rehabilitation. She also indicated that Rodney is often defiant in response to requests to complete his schoolwork and physical therapy exercises, and that he has been fighting frequently with his siblings.

This case illustration highlights the complicated, likely transactional nature of the relationships between psychological and family functioning, and ethnic and socioeconomic factors. In this case, financial strain, occupational demands, and related demands from publicly- funded medical assistance may contribute to increased levels of distress in Mrs. W. In addition, it appears that Mr. W.'s demanding work schedule leaves his wife to manage all four children, doctors' visits, Rodney's rehabilitation, discipline, and household duties on her own, despite working at her own full-time job. Furthermore, these factors are likely associated with Mrs. W.'s reported family dysfunction and Rodney's externalizing behaviors. Although the direction of these relationships is not clear, the literature discussed above suggests that socioeconomic factors and ethnicity should be considered important variables in understanding modifiable predictors, such as parental distress and family functioning, and outcomes for adjustment in children and adolescents with pediatric conditions. In the present study, 72% of the sample was Caucasian, with an average middle to upper class socioeconomic status. Previous research (e.g Rubenfeld et al., 1988; Varni et al., 1989a; Varni et al., 1989b; Varni & Setoguchi, 1993) investigating psychological adjustment in children and adolescents with limb deficiencies have generally noted middle class socioeconomic status. Information on the ethnicity of these samples was not available. While the sociodemographic composition of these samples do not indicate generally greater risk for these participants

as a group, adjustment in children and families may be maximized by identifying specific cases that may be at greater risk given these sociodemographic factors.

Psychological Adjustment in Adolescents with Limb Deficiencies

In this study, adolescents with limb deficiency disorders were not found to be at greater risk for psychological problems, regardless of treatment. Parent and adolescent reports of psychological functioning were comparable to a normative sample. These results are not consistent with earlier findings that indicated significantly more psychological problems in children and adolescents with limb deficiencies when compared to the CBCL normative sample (Varni & Setoguchi, 1992). In interpreting the results of the present study, it is important to consider that effect sizes for comparisons between groups are increased with more careful matching of chronic illness and comparison samples (Lavigne & Faier-Routman, 1992). Therefore, use of the non-matched CBCL and YSR normative samples in the present study may have interfered with the detection of differences in psychological problems between adolescents with limb deficiencies and the normative group. This issue was noted in a meta-analytic review of youth with chronic arthritis in which children and adolescents had significantly more psychological problems than study-recruited controls but not when compared to normative data (LeBovidge et al., 2003). In addition, the CBCL and YSR were designed to identify psychopathology, however, these measures are not sensitive to the more subtle psychological problems that are characteristic of youth with pediatric conditions (Drotar, Stein, & Perrin, 1995). This may have been the case in the present study in which the

percentage of participants falling in the clinical range for psychological problems was generally low.

In spite of these considerations, the data from the present study suggest that youth with limb deficiencies and their families are generally well- adjusted and resilient in the face of the demands and stressors of living with a medical condition. This finding is consistent with results in a variety of studies investigating adjustment in other pediatric populations. For example, in a study of school-aged children (6-16 years) infected with HIV (N=68), Bachanas and colleagues (2001) found that participants generally did not experience significant psychological distress. In addition, significant differences between the HIV-infected group and healthy control group were not observed on measures of caregiver psychological functioning, caregiver reported internalizing and externalizing problems in their children, or on child self-reported psychological adjustment (Bachanas et al., 2001). Pilot data from the development of a brief screening instrument designed to identify high risk families in pediatric oncology indicated that most families did not present with multiple indications of elevated risk for psychological difficulties (Kazak et al., 2001). Rather, the majority of families in this study were functioning competently, consistent with the investigators' expectations of general resilience in the face of illness and related stressors (Kazak et al., 2001). In another study, no group differences were found between preadolescents (8-9 years) with spina bifida (N=68) and physically healthy participants (N=68) on measures of internalizing and externalizing problems, global self-worth, or expressed affect in family discussions (Holmbeck et al., 2003). These findings indicated resilience for children with spina bifida in these domains. This study also addressed the need to assess multiple dimensions of psychosocial functioning

by multiple informants in order to obtain more accurate and comprehensive information regarding adjustment to chronic illness compared to the generally broadband, single dimension, single informant focus typical of adjustment studies in pediatric psychology (Holmbeck et al., 2003).

Resilience has also been observed with regard to social functioning in youth with chronic medical conditions. Reiter-Purtill et al. (2003) found that youth with JRA (8-15 years; N=57) did not differ significantly from healthy control participants (8-15 years; N=63) on peer-, teacher-, and self-reports of social functioning. In addition, no significant differences in social adjustment, social performance, or social skills were found between children with insulin dependent diabetes mellitus (N=25), asthma (N=19), and physically healthy children (N=24) further suggesting resilience in social functioning for youth with pediatric conditions (Nassau & Drotar, 1995).

Treatment and Adjustment

Hypothesized differences in psychological adjustment and functional independence between groups and treatment were not found in the present study. Adolescents with limb deficiencies likely assert increased autonomy, as expected during this developmental stage, however, their attempt to do so may be inhibited by decreased mobility and restrictions in functional independence. The findings from the present study suggest that EFT, amputation, and other types of treatment for adolescents with limb deficiencies do not differentially affect psychological and functional outcomes. That is, treatment type does not appear to exacerbate or minimize restrictions in functional independence or psychological adjustment, which is notable, given the struggle to

develop increased independence throughout adolescence. Based on these results, treatment type does not appear to be a risk factor for poor adjustment and lower levels of functional independence in adolescents with limb deficiencies.

There are several possible explanations for these results. First, there was significant difficulty in creating an appropriate control group. As discussed in the literature review, treatment decisions for youths with limb deficiency disorders are based on a variety of factors, including the extent of discrepancy or rotation and degree of foot deformity. Therefore, not all adolescents with limb deficiencies are given the option of EFT. The limb deformity may be too severe, not severe enough, or the structural presentation may not lend itself to EFT. As such, it is very challenging to find a comparison group of adolescents with the same limb deformities who could have received EFT but chose another appropriate treatment. Therefore, in the present study, adolescents who had undergone any type of treatment for limb deficiencies other than EFT were included in the control group. In addition, there was a need to include a broad range of participants with limb deficiencies in order to accrue an adequate number of participants to conduct appropriate statistical analyses. Other studies investigating adjustment in children with limb deficiencies have reported sample sizes ranging from 12 to 54 participants, with data collected over 3 to 25 years (Birch et al., 1999; Choi et al., 1990; Ghoneem et al., 1996; Herring et al., 1986; Hrutkay & Eilert, 1990; McCarthy et al., 2000; Naudie et al., 1997; Varni et al., 1991). As a result, both the EFT and TC groups were heterogeneous with respect to type, structure, and extent of limb deficiency. Attempts were made to control for varying severity of conditions within and across groups, and results suggested that severity of condition did not play a significant role in

adjustment. This is consistent with results from Lavigne and Faier-Routman's (1993) meta- analytic review of psychological adaptation to pediatric conditions as well as more recent studies (e.g Brown et al., 2000; Casey et al., 2000) in which severity was not found to predict outcomes. In a series of studies of children with limb length deficiencies, degree of limb loss was also found to be unrelated to psychological functioning (Varni et al., 1989a; Varni et al., 1989b; Varni et al., 1991; Varni & Setoguchi, 1996). However, the measure of severity in the present study was based only on structural presentation (length discrepancy, angulation, or degree of limb loss) and should be interpreted with caution. Taken together, the difficulty in finding a well-matched control group, with respect to condition and treatment options, and the related heterogeneity of the sample may have interfered with the identification of differences in adjustment between adolescents who had undergone EFT and those who did not.

Second, the small sample size and limited power in the present study may have made it difficult to detect true differences between groups. While the sample size in the present study is comparable or greater than previous studies investigating children with limb deficiency disorder, observed power was low, ranging from .08 to .30 for the multivariate analyses of variance ($p = .05$). Larger sample size per group (N=44) would be necessary to achieve power of .80 to detect a medium effect between three groups, for two dependent variables (Hair et al., 1995). Small sample size and related power issues may also have contributed to the non-significant findings in all but one of the mediation analyses.

Third, the non-significant results for between-group analyses may indeed indicate no differences in psychological functioning and functional independence between

children who have undergone EFT and those who have not. Although EFT and amputation each involve different stressors during surgery, treatment, rehabilitation, and subsequent functioning, each surgical intervention carries its own costs and benefits. For example, EFT may salvage and improve an existing limb; however, the treatment and rehabilitation is lengthy. Amputation results in limb loss and prosthetic management but the primary surgical component of treatment is completed immediately. Both EFT and amputation are characterized by unique cosmetic issues that may be particularly relevant for adolescents, given the increased focus on physical appearance during this period, and with regard to the physical changes characteristic of early to middle adolescence (Lerner et al., 1991). Findings from the present study cautiously suggest that, although EFT and amputation each have different cosmetic issues (visible frames versus limb loss and prosthesis), these differences may not differentially influence adaptation across treatment types. However, this study did not specifically investigate perceived physical appearance and how this may relate to adjustment and functional independence. In previous research, Varni and Setoguchi (1996) found higher perceived physical appearance to be associated with less depressive symptoms, lower trait anxiety, and greater overall self-esteem in adolescents with limb deficiencies. Moreover, the relationship between perceived physical appearance and psychological adjustment in adolescents with limb deficiencies was mediated by self-esteem. The Varni and Setoguchi (1996) study did not assess functional independence or compare adaptation across treatment types. Their study suggests a need for further investigation of self-esteem and perceived physical appearance as predictors of adjustment, both psychological and functional, for adolescents with limb deficiencies, in the context of treatment.

The Lavigne and Faier-Routman (1993) meta- analytic review and other studies (e.g, Brown et al., 2000; Casey et al., 2000) indicated that disease and disability parameters did not play important roles in the psychological adjustment of children with limb deficiencies. Modifiable variables, such as parents' psychological distress and family functioning, may be more important contributors to adjustment than the unique sequalae associated with specific surgical interventions.

Limitations and Future Research

Many of the limitations of the present study have already been addressed throughout the Discussion section but will be reviewed here. First, group differences are more difficult to detect given the small sample size, low power, and heterogeneity of the sample. Sample size can be increased through collaborative studies across sites. Greater access to children with limb deficiencies may also allow for more homogeneous groups in terms of etiology and clinical presentation and minimize within group variability, resulting in greater clarity for between- group differences.

Second, the direction of relationships between predictors and outcomes cannot be determined due to the cross-sectional nature of the study. In addition, the correlational nature of the research precludes collection of data on participant functioning over time. Future research should include prospective studies to assess pre-treatment functioning, functioning over the course of treatment, and post-surgical adjustment to help identify the direction of relationship between parent, family, and patient variables, as well as more clearly highlight the role of medical factors. Prospective studies can also help ascertain changes in parental distress, family functioning, and psychological problems in children

at different stages of treatment and rehabilitation. With this information, psychosocial interventions could be targeted to families at periods of greater risk.

To follow up the present study, a closer investigation focusing on psychosocial adjustment in children and adolescents who receive EFT throughout their treatment may highlight important factors related to successful rehabilitation and subsequent functioning. Given the unique challenges and invasive nature of EFT, psychological distress is expected to be greater during EFT compared to ratings completed after the removal of the fixator as in the present study. However, future studies should assess multiple, more specific domains of psychosocial functioning, beyond CBCL and YSR Internalizing and Externalizing behaviors since these measures may not be as sensitive to the sub-clinical distress more commonly observed in pediatric sample. Investigation of psychosocial adjustment during EFT should include assessment of social functioning, pain, and quality of life, in addition to internalizing and externalizing problems. Important adjustment information could also be obtained by including a measure of emotional and behavioral problems specific to youth in medical settings, such as the Pediatric Behavior Scale (Lindgren & Koeppl, 1987). Inclusion of such a measure would provide information regarding behavior as it relates to living with a chronic medical condition, issues that may not be elucidated with a general measure of psychological functioning (i.e. CBCL/ YSR).

Social functioning is an important target of investigation given disease and treatment parameters that may influence peers relations. For example, school absences during EFT that may due to frequent clinic visits, restricted mobility, and pain could lead to social isolation and interfere with developing social skills. According LaGreca (1990),

conditions that affect physical appearance may lead to negative self-perceptions or negative peer interactions. For adolescents undergoing EFT, the visibility of the external fixator may cause the adolescent to feel embarrassed, or may result in negative reactions from peers. LaGreca (1990) also noted that restrictions in physical activities may interfere with the ability to engage with peers in many social activities. This may be particularly relevant for youth undergoing EFT given related restrictions in mobility. Measurement of social functioning in youth with chronic illness must be sensitive to the above issues. For example, the CBCL Social Competencies scale evaluates the child's participation in peer relationships, school, sports, other activities, and household responsibilities (Perrin et al., 1991). Children with limb deficiencies may be less able to participate in such activities, due to the degree of their physical handicap, restrictions in mobility, and medical and rehabilitation appointments. As a result, the CBCL Social Competencies scale may not be the most appropriate measure of social functioning in this population. Research in youth with pediatric conditions has more recently focused on other components of social functioning that may not be as affected by the child's physical limitations, such as behavioral reputation, social acceptance, social skills, social self-concept, social anxiety, and loneliness (Reiter- Purtill & Noll, 2003). In addition, a measure of social functioning specifically for youth with chronic illness, such as Living with Chronic Illness (LCI; Adams, Streisand, Zawacki, & Joseph, 2002) highlights illness-related and non-illness related social difficulties.

Due the nature of the treatment and the demands of rehabilitation, an investigation of psychosocial functioning in youth undergoing EFT should also assess pain. Young, Bell, and Anthony (1994) evaluated pain in adolescent patients during EFT using the

Adolescent and Pediatric Pain Tool (APPT; Savedra, Tesler, Holzemer, & Ward, 1989). The APPT is an assessment of current pain and includes a visual analogue scale, a listing of 42 words that describe pain, as well as two body diagrams. Young et al. (1994) found that adolescents undergoing EFT experience significant pain as a result of the initial corticotomy incision, acute post-operative edema, soft tissue tension due to distraction and related muscle and skin stretching, infected pin sites, instability in the fixator, and gait training/ rehabilitation. In children with cerebral palsy, pain has been noted during assisted stretching (Hadden & Von Baeyer, 2002), a finding that may be comparable to pain experienced during EFT rehabilitation. Studies have shown pain to be related to internalizing and externalizing problems (Vaalamo, Pulkkinen, Kinnunen, Kaprio, & Rose, 2002) and quality of life (Hunfeld et al., 2001). The evaluation of pain may include multiple components. For example, the Pediatric Pain Questionnaire (PPQ; Varni & Thompson, 1985) assesses pain intensity and location (using visual analogue scale ratings and body diagrams), sensory and affective components of pain (using a list of pain descriptors), as well as the child and family's pain histories. The Waldron/ Varni Pediatric Pain Coping Inventory evaluates pain coping strategies, found to be associated with pain and adjustment outcomes (Varni et al., 1996).

An investigation of psychosocial adjustment in youth with limb deficiencies undergoing EFT should also include an assessment of health-related quality of life. Quality of life refers to objective and subjective well-being in multiple domains of life (Wallander, 2001). Health related quality of life (HRQOL) focuses on disease- and treatment related symptoms, functional implications, and how the medical condition influences psychosocial well- being (Wallander et al., 2003). For example, in children

with cystic fibrosis, HRQOL has been found to be related to general health status and pulmonary function (Czyzewski, Mariotto, Bartholomew, LeCompte, & Sockrider, 1994). In addition, research has indicated chronic pain in adolescents to be negatively associated with quality of life (Hunfeld et al., 2001). This finding may be particularly relevant given concerns regarding pain during EFT rehabilitation. Generic (i.e. not disease-specific) measures of HRQOL generally evaluate functioning across multiple domains (Quittner, Davis, & Modi, 2003). For example, the Child Health Questionnaire (CHQ; Landgraf, Abetz, & Ware, 1996) includes both adolescent self-report and parent-proxy report questionnaires that assess physical functioning, social-emotional/ behavioral functioning, social-physical functioning, bodily pain, behavior, mental health, self-esteem, general health, parental impact- emotional, family activities, family cohesion, and changes in health (Waters, Salmon, & Wake, 2000). Varni and colleagues (2001) developed the Pediatric Quality of Life Inventory 4.0 (PedsQL 4.0) that includes self- and parent reports for generic core scales (Physical Functioning, Emotional Functioning, Social Functioning, and School Functioning) that can be used across pediatric populations, as well as disease-specific modules for pediatric asthma, rheumatology, diabetes, cancer, and cardiac conditions (Quittner et al., 2003). Given that an HRQOL measure does not exist specifically for youth with limb deficiencies, either the CHQ or PedQL generic measure would be appropriate for the assessment of children and adolescents undergoing EFT.

As research in youth with limb deficiencies progresses, comparisons of school-aged children and adolescents undergoing EFT may highlight different patterns of psychosocial adjustment, given the developmental tasks unique to each stage. While

differences in general measures of adjustment (i.e. CBCL/ YSR) may not be observed across developmental stages, assessment of specific domains of psychosocial functioning may yield patterns of adjustment that vary with age. For example, based on a review of social functioning in youth with chronic illness, Reiter-Purtill and Noll (2003) reported that older children with chronic medical conditions appear to be more likely to experience social problems than younger children. This finding may be understood in the context of adolescent developmental tasks which are integrally linked to identification with and acceptance by peers, such as increasing autonomy and identity formation, (Reiter- Purtill & Noll, 2003). Greater importance of peer and social relations during adolescence and possible disruptions in typical social development and peer relations due to living with a chronic medical condition (i.e. missed school days, treatment regimens, changes in appearance) may contribute to increased social difficulties in adolescents with chronic illness. In addition, adolescents with cancer have been found to be at greater risk for lower levels of health- related quality of life when compared to younger children (Barrera et al., 2003). While social functioning and quality of life may be more important domains of psychosocial functioning for adolescents with chronic medical conditions, behavior problems are more common in younger children. For example, preschoolers with cancer have been found to experience more externalizing difficulties than adolescents. These findings suggest that specific components of psychosocial adjustment in youth with chronic illness may highlight areas of risk that appear to coincide with typical developmental issues.

Additional research investigating more specific targets of psychological functioning, such as posttraumatic stress (PTS), may further elucidate the relationship

between parent and family factors and adjustment in children with limb deficiency disorders. Previous research has identified PTS in parents and children following physical injury, and in pediatric cancer and spinal cord injury populations (Aaron, Zaglul, & Emery, 1999; Barakat, Kazak, Meadows, Casey, Meeske, & Stuber, 1997; Boyer et al., 1999a; Boyer et al., 1999b). In children with cancer, PTS has also been found to be predictive of overall adjustment (Barakat, Kazak, Gallagher, Meeske, & Stuber, 2000). For children with spinal cord injuries, the relationship between family functioning and functional independence was found to be mediated by PTS (Boyer et al., 2003). Investigation of PTS in the context of treatment for children with limb deficiencies may identify additional targets of intervention in facilitating adjustment in children, parents, and families.

Future research addressing possible explanations for the relationship between orthopedic surgeries and externalizing behavior problems may identify additional targets of psychological intervention. As previously discussed, children in this study who experienced a greater number of orthopedic surgeries reported less externalizing problems. Although there may be many possible explanations for this finding, one possible interpretation is that adolescents were not disciplined for their inappropriate behaviors and, as such, did not perceive their behaviors to be oppositional, defiant, or aggressive. Research in other pediatric populations suggests that parents of children with chronic health conditions tend to report greater difficulty with discipline and limit setting for their children (Gerhardt et al., 2003; Ievers et al., 1994). Future research for children with limb deficiency disorder could highlight child-rearing patterns that may interfere with optimal treatment and rehabilitation.

Clinical Implications

Results from the present study suggest a need to offer psychological services to adolescents with limb deficiency disorders and their families. In the present study, family functioning was generally within the normal range and only a small percentage of caregivers reported clinically significant distress; however, even within a non-clinical range, variability in family functioning and caregiver distress appeared to be associated with varying levels of psychological adjustment in their children. In this context, even caregivers and families experiencing seemingly mild variations in functioning should be targeted for support and intervention. In the context of general resilience for adolescents with limb deficiencies and their families, identification of families at risk for psychosocial difficulties may facilitate efficient and cost-effective provision of psychological services to those patients and families who truly require support and intervention. Kazak et al. (2001) developed the Psychosocial Assessment Tool (PAT) to identify high- risk families in pediatric oncology. Pilot data suggested the utility of this screening instrument in identifying families at risk for poor adjustment and in guiding levels of psychosocial services (Kazak et al., 2001). Clinical services should also focus on identification of families at risk for poor adjustment prior to major treatment decisions. In addition, screening of family and individual functioning should be made available before and during the course of orthopedic treatment and rehabilitation to identify adolescents and families who may require services. This may be particularly valuable for adolescents and families undergoing lengthy and demanding orthopedic interventions, such as EFT. By monitoring patients and their parents throughout treatment, psychological problems, or shifts in functioning, in children, parents, and the

family as a unit may be identified and managed, and further exacerbation of psychological symptomatology may be prevented. Depending on the level of apparent need, the intensity and frequency of psychological support should vary from an initial consultation/ single contact (with opportunity to follow-up) to weekly therapy sessions, for example. In addition, obtaining information on functioning prior to EFT would provide baseline data against which to compare changes in adjustment throughout treatment and rehabilitation. Awareness of change in functioning over time may further highlight periods of increased risk and distress for patients and their families.

Consider the case of the W. family previously discussed. If Rodney and his family had been identified prior to his EFT, parent, family, and child functioning could have been evaluated and monitored throughout the course of treatment and rehabilitation. Mrs. W. could have been referred for clinical services to address her own depression and anxiety. Family therapy may have facilitated improved functioning for the family as a whole. For example, shifts in roles and responsibilities with guidance from a mental health provider may have relieved some of Mrs. W.'s burden, which may have contributed to improved mood, and ultimately may have helped Rodney to better manage his rehabilitation. In addition, parent- training and instruction of behavior management strategies may have provided Rodney's parents with the tools to more effectively handle his oppositional and defiant behavior, particularly as it related to his resistance to physical therapy. With individual therapy, Rodney would have had the opportunity to express his feelings of distress and frustration and he may have learned how to better manage these feelings. Both individual and family therapy could have targeted conflict between Rodney and his parents, as Rodney attempts to increase independence and

control over his life and his parents attempt to shift their child-rearing to be congruent with Rodney's adolescent socio-emotional development. Improved functioning and symptom relief for Rodney, his mother, and the family unit would likely each influence the other and contribute to enhanced adjustment and functioning for everyone involved. This example highlights the need for available psychological services for adolescents with limb deficiencies.

The availability and delivery of psychological services for adolescents with limb deficiency disorders may be best achieved through multidisciplinary collaboration with orthopedic surgeons, nurses, and physical therapists. Physicians and physical therapists may be best able to identify problems with adherence to rehabilitation regimens and missed appointments, which may be indicative of problems in parent, family, and child functioning. A psychologist would work in concert with a physical therapist to address issues around resistance to rehabilitation regimens and pain management, for example. The availability and role of the psychologist in orthopedic clinics likely varies from occasional use of consult-liaison services to more integrated and routine involvement. However, seemingly subtle changes in family and individual adjustment in caregivers and their children may not be easily identified by medical professionals, and, as a result, these cases would not receive the psychological attention that might maximize adjustment. The inclusion of a psychologist on the orthopedic team and the presence of the psychologist in orthopedic clinics would increase visibility and afford the clinician the opportunity to meet families, and, through observation and interaction with the medical team, initially identify families who might require services. In addition, patients and their parents may be more amenable to psychological support and intervention if the psychologist is

presented and perceived as an integral and essential participant on the orthopedic team. Ultimately, the provision of psychological services, particularly targeting parent, family, and adolescent adjustment, would optimize treatment and overall patient care. Moreover, future research, as discussed above may help identify additional targets of intervention and more finely tune the role of the psychologist on the orthopedic team.

Conclusions

The present study investigated differences in psychological adjustment and functional independence in adolescents with limb deficiency disorders. Adolescents in this sample were generally resilient and did not experience more psychological problems than a normative sample. In addition, this study yielded comparable psychological and functional outcomes for adolescents who receive EFT and adolescents who receive other treatments for limb deficiencies. Most notably, variability in psychological adjustment and functional independence in adolescents with limb deficiencies was generally explained by socio-ecological factors, while time since surgery accounted for additional variability in functional independence. These results suggest important implications for clinical intervention and future research, particularly in the context of adolescence and the unique challenges that this developmental period presents for youth with limb deficiencies and their families.

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APPENDIX A: PATIENT MEDICAL AND TREATMENT INFORMATION

Please answer the following questions to the best of your ability. Feel free to use the sides and back of this form if you need additional space. There are four pages to this questionnaire.

1) Patient's age: _____
2) Date of Birth: _____

Please check the box next to your response:
3) Race:

- ❑ Caucasian
- ❑ African American
- ❑ Hispanic American
- ❑ Asian American
- ❑ Native American
- ❑ Other _____

4)Which limbs are affected?

- ❑ Left Leg
- ❑ Right Leg
- ❑ Both Legs
- ❑ Left Arm
- ❑ Right Arm
- ❑ Both Arms
- ❑ Both Arms and Legs
- ❑ Other_____

5)What is the diagnosis causing the limb deficiency(ies):

- ❑ Blounts
- ❑ Rickets
- ❑ Hypoplasia
- ❑ Leg length discrepancy
- ❑ Fracture
- ❑ Bow Leg
- ❑ Clubfoot
- ❑ Polio
- ❑ Other_____

6) Please answer the questions that apply to your child.
a. What is the degree of limb length discrepancy (for example, how much shorter is one leg than the other)? _____ centimeters (cm)

b. What is the degree if rotation/ angulation discrepancy (for example, how much is the leg bent)? _____ degrees

7) If your limb deficiency is the result of a fracture or amputation, please indicate when the injury occurred. _____

8) Is part of the limb(s) missing or has part of the limb(s) been amputated? Please describe._____
_____

9) Has external fixator treatment been discussed with you as a treatment option for your child?

YES NO

10) Was external fixator treatment offered to you as a treatment option for you child?

YES NO

- **If patient has undergone external fixator treatment, please proceed to question 13**

11) If NO: Can you please describe the reason(s) why external fixator treatment was not offered?
_____
_____
_____
_____

12) If external fixator treatment was offered to you but you chose not to get the procedure, can you please describe your reason(s)?
_____
_____
_____
_____

- **If patient has not undergone external fixator treatment, please proceed to question 18**

13) How many times has your child had external fixator treatment

- ❑ 1
- ❑ 2
- ❑ 3
- ❑ 4
- ❑ 5

14) Age at the time of external fixator treatment:

at Time 1_____
Time 2_____
Time 3_____
Time 4_____
Time 5_____

15) Number of external fixators that your child has had on each time:

at Time 1_____
Time 2_____
Time 3_____
Time 4_____
Time 5_____

16) Anatomical site of fixation:

- ❑ Thigh
- ❑ Lower Leg (below knee)
- ❑ Lower Leg and foot
- ❑ Foot
- ❑ Upper Arm
- ❑ Forearm
- ❑ Forearm and hand
- ❑ Hand
- ❑ Other_____

17) Type of fixator:

- ❑ Ilizarov
- ❑ EBI
- ❑ Garche
- ❑ Other_____

18) Number and type of all surgeries other than external fixator treatment for the limb deficiency (ies):

_____ _____
_____ _____
_____ _____
_____ _____

19) Number and type of complications from surgery or treatment

_____ Pin Infections
_____ Fractures after External Fixator Treatment
_____ Contractures
_____ Others_____

20) Number and length of hospital admissions:

_____ _____
_____ _____
_____ _____
_____ _____

APPENDIX B: DEMOGRAPHIC INFORMATION

Please answer the following questions as completely as possible.

1) Highest grade of schooling completed by the patient _____

2) Highest grade of schooling completed by patients' mother or mother figure

_____

3) Highest grade of schooling completed by patient's father or father figure

_____

4) Family income _____

VITA

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Presentations and Publications

Hitelman, J, Barakat, L., Sheslow, D. and Boyer, B. *Psychological and Orthopedic Outcomes of Treatment for Adolescents with Limb Deficiency Disorders: A Focus on the Family.* Poster to be presented at the National Conference on Child Health Psychology, April 15-17, 2004, Charlston, SC.

Boyer, B., Hitelman, J., Knolls, M., and Kafkalas, C. (2001) Posttraumatic Stress and Family Functioning in Pediatric Spinal Cord Injuries: Moderation or Mediation? *American Journal of Family Therapy, 31(1),* 23-37.

Ricardo B. Eiraldi, PhD, Laurie Mazzuca, MA, Julia Keleher, MA, and Jennifer Hitelman, MA. *School-Based Prevention Program for Minority Children With Externalizing Disorders.* Poster Presented at the American Psychological Association Convention, August 4-8, 2000, Washington, D.C.

Hitelman, J.S, McGuffin, P.W., Bricketto, K.M., Nezu, A.M., and Nezu, C.M. *Practical and Theoretical Correlations between the Adaptive Behavior Scale- Residential and Community, Second Edition and the Wechsler Adult Intelligence Scale- Third Edition.* Poster Presented at the Association for the Advancement of Behavior Therapy Convention, November 5-8, 1998, Washington D.C.

Honors and Awards

Fonds pour la Formation de Chercheurs et l'Aide a la Recherche (FCAR-1999-2000)
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