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Commentary

Impact of pediatric chronic pain on siblings: Evidence and theory

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Pediatric chronic pain is a complex health problem with far reaching effects on the whole family unit, including siblings. Having a brother or sister with a chronic pain condition renders siblings not only more likely to develop a pain condition themselves (Champion, 2013), but has also been found to have significant effects on the sibling's own functioning (Guite et al., 2007). The current article will describe the nature of the impact of chronic pain on siblings. The mechanism and theories that have been proposed to account for these associations will then be overviewed, giving consideration to both environmental and genetic influences.

Familial risks of pediatric chronic pain

A child who has a brother or sister with a chronic pain condition has been found to have a threefold increased risk of also experiencing a chronic pain condition during childhood or adolescence (Champion, 2013). Both genetic and environmental factors have been implicated, with the relative contribution of genetic and environmental influences varying for different pain conditions and across age (Hestbaek et al., 2004). Heritability estimates typically range from 30% to 50% for common pediatric recurrent and chronic pain conditions (Champion, 2013).

Over the past decade there has been considerable attention given to describing parental and familial factors in the context of pediatric chronic pain (Palermo & Chambers, 2005; Palermo & Lewandowski-Holley, 2013). A systematic

review of 16 articles examining family functioning in the context of pediatric chronic pain included 7 studies comparing the family functioning of children with chronic pain with the family functioning of healthy control children (Lewandowski et al., 2010). Four of the 7 studies revealed significantly poorer familial functioning in the context of chronic pain relative to healthy control families, with case families reporting poorer family cohesion and greater conflict. Interestingly, 1 study found families of pain patients reported significantly less conflict than normative samples (Schanberg et al., 1998), suggesting the possibility that in some contexts having a family member with pain may strengthen and unify families. Two of the smaller studies did not find any group differences in family functioning. Although the causal direction of child or adolescent pain and various familial factors is difficult to disentangle, the potential impact of these familial factors on siblings should not be overlooked.

Functioning of siblings

Despite the siblings of pediatric pain patients being exposed to many of the same parental and familial influences as the child with pain, far less research has been carried out to investigate the functioning of siblings. Compared to siblings of healthy children without pain, siblings of children with chronic pain have been found to have more emotional and behavioral symptoms, higher physiological anxiety, and lower self-esteem (Engström, 1992; Guite et al., 2007). Siblings of

pain patients have been described as commonly competing for attention, and jealous of their unwell brother or sister (Engström, 1992; Britton & Moore, 2002). Moreover, siblings of pain patients have been reported as having poorer quality peer relationships than siblings of healthy children without pain (Engström, 1992; Britton & Moore, 2002). In light of the increased demands and responsibilities that parents face in caring for a child with chronic pain (e.g. attending appointments, providing physical and emotional support), it is not surprising that they have been found to report having less energy and time for leisure activities with siblings (Britton & Moore, 2002).

Given the limited research with siblings of chronic pain patients, it is of interest to also consider the literature on siblings of children with other chronic conditions, including asthma, cancer, cardiac disease, cystic fibrosis and diabetes. Although the nature of these conditions differs, each may require use of medications, medical appointments, and may confer a range of possible impacts on daily functioning. A meta-analysis of 43 studies, published from 1970 to 1995, found 60% of studies reported increased sibling risk of poorer functioning (Williams, 1997). A second meta-analysis of 51 studies, published from 1976 to 2000, also found a statistically significant and overall negative effect on the functioning of siblings of children with a chronic illness (Sharpe & Rossiter, 2002). These reported risks and negative effects on siblings included higher internalizing and externalizing behavioral problems, lower self-esteem, withdrawal, poorer peer relations, feelings of loneliness, anxiety, fear, depression, anger, guilt and worry, and school problems (Williams, 1997; Sharpe & Rossiter, 2002; Williams et al., 2009; Alderfer et al., 2010).

Research investigating the experience of a child or adolescent who has a brother or sister with a chronic health condition is still in its infancy and has largely been exploratory in nature (e.g. Engström, 1992; Britton & Moore, 2002; Guite et al., 2007). There is also a trend in contemporary medical research to search for risk factors associated with the development of health conditions (Ware, 2006) such as chronic pain. Such research commonly assesses large numbers of

variables, typically without clear theoretical conceptualizations about how these variables are interrelated and without shedding light on causal processes. Parental and familial factors have often been considered in such a context, typically in the absence of a sound conceptual framework (Palermo & Chambers, 2005). For the field to progress, greater consideration needs to be given to key environmental mechanisms, genetic mechanisms, as well as the complex interaction of the two, in accounting for functional impairments in siblings. Where possible, these factors should be considered within a broader theoretical conceptualization.

Environmental mechanisms

Broadly, two classes of theories have been drawn upon when accounting for the common reductions in sibling functioning, namely, (a) social learning theory, and (b) family systems theories. Social learning theory holds that individuals learn by observing others (Bandura, 1977). Individuals may be more likely to imitate behaviors displayed by their parents or other individuals significant in their lives, such as older siblings (Ardelt & Day, 2002). For example, given that parental attention is highly valued, siblings may learn and copy behaviors from their brother or sister that are seen to result in increased parental attention.

According to the family systems theory, the family may be considered as an interacting unit with a common purpose (Bavelas & Segal, 1982). Members affect each other, and cannot be understood in isolation (Bavelas & Segal, 1982). Family roles are interconnected and therefore a change in one role affects everyone else's roles. When a child experiences chronic pain, family members typically take on different roles to compensate for changes in the individual's functioning. For example, siblings may take on additional roles to compensate for things that their brother or sister is unable to do; parents may spend more time and emotional energy on the child with pain, leaving less time and energy for other siblings. Generally, family members are motivated by returning the affected individual to a pain-free state, and maintaining a desired level of family functioning. Families must keep shifting to suit the changing needs of the child in pain. Occasionally,

the ongoing nature of the individual's pain may serve some benefit within the family system, for example a parent enjoying a sense of being needed by the child, or a parent staying home with their child and not attending a workplace they don't enjoy. The family systems theory has predominantly been applied to research in a descriptive way, rather than as a model for hypothesis generation. Traditionally, the family systems theory does not seek to identify cause and effect, but rather to describe an ongoing, fluid process (Bavelas & Segal, 1982).

Genetic mechanisms

Some children are evidently vulnerable to chronic or recurrent common pain disorders which are frequently multiple (von Baeyer & Champion, 2011). This vulnerability and associated psychological vulnerability is probably significantly genetically influenced, but published evidence is very limited. Research with adult chronic pain populations has found a complex interaction between environmental triggers and genetic vulnerabilities (Mogil, 2012). In a pediatric twin family case-control study, if a parent had a history of chronic pain, the odds of each child having chronic pain increased twofold (Champion, 2013). Similarly a parent having functional abdominal pain or migraine conferred a one and a half fold increase in odds of a child and sibling having chronic pain. Moreover, siblings were at significantly increased risk of migraine, nonmigraine headaches and functional abdominal pain when one of their siblings had those conditions. Not all of such increased risk can be assumed to be genetic.

The importance of identifying genotype-environment correlations has also recently been highlighted as a promising avenue for future research (Avinum & Knafo, 2013; Ludeke et al., 2013). Given that environmental factors such as parent-child interactions are underpinned by genetically-influenced traits such as personality, mental health and intelligence, a more complex examination of environmental and genetic factors is needed than hitherto achieved.

Conclusion

Siblings of chronic pediatric pain patients are not only at increased risk of developing a pain condition themselves, but they are also at risk of poorer functioning. These effects need to be understood within the context of environmental and genetic influences. The social learning theory and the family systems theory have been drawn on to describe these associations. Further theoretically-driven research is needed to increase understanding of both the impact and mechanisms associated with pediatric chronic pain and sibling functioning. Specifically, more research is needed to investigate factors that may adversely impact sibling functioning, as well as factors that may mitigate such influences. Little is currently known about why some siblings experience poorer outcomes than other siblings within the same family. A greater understanding of risk factors and protective factors may help guide the development and use of targeted sibling screening tools to assess those at greatest risk of poor outcomes. This assessment process then needs to be complemented by the development of theoretically-driven clinical interventions for this at-risk sibling population and their families.

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