



Pica in Children With Sickle Cell Disease: Two Case Reports

Erin T. O'Callaghan PhD^{a,*}, Jeffrey I. Gold PhD^b

^aCalifornia School of Professional Psychology, Alliant International University, Los Angeles, Alhambra, CA

^bKeck School of Medicine, University of Southern California, Children's Hospital Los Angeles, Los Angeles, CA

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Children with sickle cell disease (SCD) are at greater risk for developing pica compared to other children. This comorbidity can result in harmful medical and nutritional, and neurodevelopmental consequences. This article will describe the medical, nutritional, and psychosocial functioning in two children with SCD and pica in order to illustrate the potential complications and correlates of this comorbidity. In addition, the clinical implications of pica in children with SCD will be discussed.

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AS DEFINED BY the *Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, Text Revision (DSM-IV TR)*, pica is the persistent eating of nonnutritive substances (American Psychiatric Association, 2000). A diagnosis of pica is made if eating nonnutritive substances persists for at least 1 month, the eating behavior is not part of a culturally sanctioned practice, and this behavior is not simply a symptom of another mental disorder. Pica is most common in young children (Chatoor, 2004), but it has also been reported in pregnant women (Corbett, Ryan, & Weinrich, 2003; Simpson, Mull, Longley, & East, 2000) individuals with intellectual and developmental disabilities (Danford & Huber, 1982; Matson & Bamburg, 1999; Williams, Kirkpatrick-Sanchez, Enzinna, Dunn, & Borden-Karasack, 2009), and individuals in lower socioeconomic groups (Rose, Porcerelli, & Neale, 2000). Ingestion of non-food substances can result in serious health complications, including intestinal obstruction (Anderson, Akmal, & Kittur, 1991; Chiu, Ciaccio, & West, 2005), development of gastric bezoars (Sprinkle & Hingsbergen, 1995; Stein-Wexler et al., 2006), lead poisoning (Issaivanan, Ahmed, Shekher, Esernio-Jenssen, & Manwani, 2009; Jones, 2009), dental injury (Baker, 2005), and electrolyte imbalance (Appel & Bleyer, 1999). Although the exact etiology of pica is unknown, pica has been linked to iron, zinc, and other mineral/nutritional

deficiencies (Khan & Tisman, 2010; Singhi, Ravishanker, Singhi, & Nath, 2003); dysfunctional eating patterns (Lemanek, Brown, Armstrong, Hood, Pegelow, & Woods, 2002); obsessive-compulsive disorder (Gundogar, Demir, & Eren, 2003); and psychosocial stressors (Singhi, Singhi, & Adwani, 1981).

Sickle cell disease (SCD) is an inherited red blood disorder that alters hemoglobin, which is the protein that transmits oxygen in the body. This results in the creation of abnormally shaped red blood cells that can obstruct blood flow. In the United States, SCD affects mostly African-Americans. It occurs in approximately 1 in 500 African-American births and 1 in every 1000 to 1400 Hispanic-American births. Obstruction of blood flow in individuals with SCD is associated with numerous medical complications including frequent pain episodes and chronic pain, damage to vital organs, cerebral infarctions, increased risk of infections, and respiratory problems (National Heart, Lung, and Blood Institute [NHLBI], 2010). These medical complications are associated with frequent hospitalizations. In addition, strokes are associated with neurological and neuropsychological deficits (NHLBI, 2010). In addition to the numerous medical complications associated with this disease, children with SCD are at higher risk for the development of emotional and behavioral problems including internalizing problems (i.e., depression and anxiety) and externalizing problems (i.e., oppositional behavior, inattention, hyperactivity) when compared to other children with

* Corresponding author: Erin T. O'Callaghan, PhD.

E-mail address: eocallaghan@alliant.edu (E.T. O'Callaghan).

chronic medical conditions and physically healthy youth (i.e., Benton, Boyd, Ifeagwu, Feldtmose, & Smith-Whitley, 2011; Benton, Ifeagwu, & Smith-Whitley, 2007; Hijmans et al., 2009; Kelch-Oliver, Oyeshiku Smith, Diaz, & Collins, 2007). It is clear that SCD is a complicated disorder that can be associated with numerous medical, psychosocial, and neurocognitive difficulties.

Clinicians and researchers alike have long noted an increased incidence of pica in children with SCD (i.e., Ivascu et al., 2001; Lemanek et al., 2002). However, only two empirically-based research studies have examined the prevalence rates and correlates of this comorbidity. One study conducted by Ivascu et al. (2001) noted an overall prevalence rate of 33.9% in 395 children with sickle cell disease. More specifically, approximately 36% of children homozygous for Hb S (HbSS) had pica; whereas 25.5% of children with Hb SC, Hb SD, and Hb S β thalassemia had pica (Ivascu et al., 2001). Issaivanan et al. (2009) noted in their review of the literature on children with SCD and lead poisoning, that 76% of the cases reported were positive for a history of pica. Additional research is needed to further investigate the prevalence of pica in children with SCD.

Other than the prevalence study by Ivascu et al. (2001), only one other empirical research study has specifically examined pica in children with SCD. Lemanek et al. (2002) analyzed dysfunctional eating patterns, pica, and disease severity in 139 children with SCD. Based on caregiver report, 51.8% of the children with SCD demonstrated clinically significant dysfunctional eating patterns including pica, and 62.2% of the children with SCD showed symptoms of pica (Lemanek et al., 2002). In addition, these researchers reported that dysfunctional eating patterns were associated with a greater number of pain episodes and hospitalizations. Despite these important findings, no further research on this topic has been conducted.

In addition to the abovementioned studies, there are three published case reports describing pica in children and adolescents with SCD (Altepeter, Annes, & Meller, 2011; Hackworth & Williams, 2003; Stein-Wexler et al., 2006). Altepeter et al. reported on the surgical resection of a gastric bezoar in an adolescent girl with SCD who was hospitalized with abdominal pain after ingesting material/fabric from a couch and car seat cushions. Informed by a biopsychosocial perspective, Stein-Wexler et al. published a single case study of a boy who ate hair and carpet and developed a trichobezoar and Hackworth and Williams described three children who ate foam rubber. Although both of these case reports proposed that the children's pica symptoms were related to anxiety or other psychosocial stressors, the authors did not utilize any formal measures of emotional and/or behavioral functioning.

The purpose of the current case report is to describe the medical, nutritional, and psychosocial functioning of two children with SCD and pica from a biopsychosocial perspective, in order to illustrate the potential complications

and correlates of this co-morbidity. The clinical implications of pica in children with SCD will also be discussed.

Method

A case study approach was used to present two cases of children with SCD and pica (ages 7 and 10 years). As part of standard of care, these children received medical and behavioral health care to treat both SCD and pica. To formulate the following two case reports, a retrospective electronic medical chart review was conducted to obtain relevant medical information. All laboratory and medical information was confirmed via consultation with medical personnel. Behavioral health information was obtained from a review of behavioral health records. Before participation in this case study, the children's legal guardians provided authorization under the Health Insurance Portability and Accountability Act (HIPAA) as instructed by the Institutional Review Board.

Reviews of medical and behavioral health records provided information about the children's demographic information, disease status, history of pica symptoms, nutritional history, academic performance, and overall psychosocial history and functioning. As part of their behavioral health care, the specific emotional and behavioral functioning of the two children with SCD and pica was assessed using the *Behavior Assessment System for Children, Second Edition: Parent Report Scale and Self Report of Personality* (BASC-2 PRS and SRP; Reynolds & Kamphaus, 2004). The BASC-2 is an empirically validated rating scale of children's behavioral and emotional functioning with high rates of internal consistency and test-retest reliability (Reynolds & Kamphaus, 2004). The BASC-2 assesses for symptoms associated with childhood disorders in the *DSM-IV* TR and has been used to aid in screening and diagnosis in pediatric primary care settings (Simonian, 2006). Further, the BASC-2 has also been used extensively for research purposes with numerous populations, including the report of behavioral and emotional functioning in children with chronic medical illnesses such as cancer and sickle cell disease (i.e., Wolfe-Christensen, Mullins, Stinnett, Carpenter, & Fedele, 2009; Ziadni, Patterson, Pulgaron, Robinson, & Barakat, 2011).

Teacher-, parent-, and self-report measures are available to provide an assessment of a child's behavioral and emotional functioning in multiple settings. Scores on the BASC-2 are converted to *T*-scores and are classified within either the clinical or adaptive scales. The clinical scales measure behaviors that are maladaptive (aggression, anxiety, attention problems, atypicality, conduct problems, depression, hyperactivity, learning problems, somatization, and withdrawal). The adaptive scales measure positive behaviors (activities of daily living, adaptability, functional communication, leadership, social skills, and study skills). Clinical scale *T*-scores in the *average* range (ranging from

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