



Caregiver survey of pharmacotherapy to treat attention deficit/hyperactivity disorder in individuals with Williams syndrome



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ABSTRACT

Williams syndrome (WS) is a genetic condition characterized by a unique neurocognitive and behavioral profile, including increased incidence of attention deficit/hyperactivity disorder (ADHD). The purpose of the present study was to examine the perceived helpfulness and side effects of medications used to treat ADHD (methylphenidate class, amphetamine class, atomoxetine) in individuals with WS. This was accomplished with a survey of parents/caregivers of individuals with WS through the Williams Syndrome Association. Five-hundred twelve (512) parents/caregivers responded to the survey regarding their child's/adult child's use of ADHD medications. Twenty-seven percent (27%) of the individuals had been prescribed a medication for ADHD, most commonly a methylphenidate class medication. OROS-methylphenidate was reported as the most helpful methylphenidate class formulation, with 74% reporting it at least somewhat helpful. Survey participants reported similar side effects as typically developing controls, but to a greater degree. Irritability was the most commonly endorsed side effect of an ADHD medication (38%). Individuals reported use of stimulant medications in the presence and absence of underlying cardiac conditions, with 56% of ADHD medication users reporting supra-aortic stenosis, 36% pulmonary artery stenosis, and 25% systemic hypertension. Individuals taking ADHD medications were more likely to report dental problems ($p = 0.004$). Additional studies are needed to further investigate these findings and examine short-versus long-acting stimulant medications and dosage effects.

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1. Introduction

Williams syndrome (WS) is a genetic condition with an estimated prevalence of 1 in 7500 (Strømme, Bjørnstad, & Ramstad, 2002). It is caused by a deletion at chromosome 7q11.23 of approximately 1.6 million base (Mb) pairs (Hillier et al.,

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2003), including the gene for elastin (ELN). WS is characterized by certain specific phenotypic features, including characteristic facies, cardiac defects (most notably supravalvular aortic stenosis), hypercalcemia, friendly personality, and intellectual disability (Waxler, Levine, & Pober, 2009). It is estimated that at least 75% of individuals with WS have IQ and adaptive behavior scores that identify them as having an intellectual disability (Mervis & Klein-Tasman, 2000) and their average IQ score ranges from 42 to 68, which is within the Mildly to Moderately Delayed range (Martens, Wilson, & Reutens, 2008). The behavioral phenotype of WS includes an increased incidence of certain psychiatric disorders, such as attention deficit/hyperactivity disorder (ADHD), anxiety disorder, and specific phobias (Einfeld, Tonge, & Florio, 1997; Einfeld, Tonge, & Rees, 2001; Greer, Pai, Choudry, & Klein, 1997; Leyfer, Woodruff-Borden, Klein-Tasman, Fricke, & Mervis, 2006; Rhodes, Riby, Park, Fraser, & Campbell, 2010).

A 2006 study of 128 children with WS found that 64.7% of the sample population met DSM-IV criteria for ADHD (Leyfer et al., 2006). Of those who met criteria, 69% were in the Inattentive subtype, 27% were in the Combined subtype (Inattention & Hyperactivity-Impulsivity), and 4% were in the Hyperactive-Impulsive subtype. Short attention span was endorsed in over 95% of children with WS on a parent-report developmental checklist in a 1997 study (Einfeld et al., 1997). A recent study used the Conners Parent Rating Scale to examine attention deficit/hyperactivity symptoms in 11 children; subjects scored within the abnormal range in the categories of Cognitive Problems/Inattention (100%), Hyperactivity (90.9%), and the ADHD index (100%) (Rhodes et al., 2010). Up to 85% of adults with WS are reported to have difficulties with distractibility (Elison, Stinton, & Howlin, 2010).

In the general population, prevalence studies have shown varying rates of ADHD, ranging from 2.6% to 10.8% in male adolescents and 0.54% to 3.9% in female adolescents (Barbaresi et al., 2002; Cuffe et al., 2001; Neuman et al., 2005). Studies have shown increased incidence of ADHD in children with an intellectual disability (ID). A recent study compared children who have ADHD with and without ID, finding that 35–50% of 5–8-year-olds with ID met criteria for ADHD versus 8–12% of typically developing (TD) 5–8 year-olds (Neece, Baker, Blacher, & Crnic, 2011). When compared to TD children, other studies have shown a 2.5 to over 3.5 times increase in the incidence of hyperactivity and/or attention problems in children with developmental delays and/or intellectual disability (Dekker, Koot, Ende, & Verhulst, 2002; Emerson & Einfeld, 2010).

Although up to 64% of individuals with WS meet criteria for ADHD, with an even greater percentage reportedly showing evidence of inattention and distractibility, few studies have evaluated the use of stimulant medications in this population. Four studies completed in the last 15 years have examined a total of 46 children. The only medication reported in these studies was methylphenidate. The largest and most recent study, including 38 children and adolescents, found that 72% of the 30 children with ADHD treated with methylphenidate experienced significant improvement (Green et al., 2011). Two previous reports also concluded methylphenidate was useful in treating ADHD symptoms in WS (Bawden, MacDonald, & Shea, 1997; Power, Blum, Jones, & Kaplan, 1997), while a third report determined methylphenidate showed no benefit over placebo in treating ADHD symptoms in two children with WS (Zwaigenbaum, Dick, Handley-Derry, Malone, & Jacobson, 2006). Given previous studies have had relatively small samples and have studied only methylphenidate, the purpose of this survey was to gather more information on the use of ADHD medications in WS. We utilized a large sample, included several ADHD medications, examined degree of helpfulness, and described reported side effects. It is our intent that these results may be used to help direct future studies in this area.

2. Methods

2.1. Participants

A survey about psychotropic medication use was distributed to 2846 members of the Williams Syndrome Association. Participants completed the survey either on-site at a Williams Syndrome Association National Conference or online from home. The survey was also available in paper format, but the vast majority of survey responses were online. We collected data from 512 caregivers of individuals with WS. Of the 358 who reported age of the WS individual at the time of the survey, 145 were adults (41%) and 213 were children under 18 years of age (59%).

2.2. Survey design

This survey of caregivers of children and adults with WS included questions regarding ADHD medication use, including Ritalin[®] (methylphenidate), Methylin[®] (methylphenidate), Focalin[®] (dexmethylphenidate), Concerta[®] (osmotic release oral system [OROS]-methylphenidate), Metadate[®] (modified-release methylphenidate), Daytrana[®] (transdermal methylphenidate), Dexedrine[®] (dextroamphetamine), Dextrostat[®] (dextroamphetamine), Adderall[®] (mixed amphetamine salts), Vyvanse[®] (lisdexamphetamine), and Strattera[®] (atomoxetine). Both the trade names and the generic names were listed in the survey, given the similarity of the generic names of many of the stimulants and greater public familiarity with trade names; however, for the purpose of this paper we report using generic names. Survey participants were allowed to write in other medications taken for ADHD symptoms. The survey gathered information on age initiated, purpose, duration of use, perceived effectiveness (Helpful, Somewhat Helpful, Not Helpful), and side effects (tremor/shaking, anxiety, headache, sleep problems, irritability, stomach ache, weight loss, muscle tics, fast heart rate, high blood pressure, aggression/fighting, hallucinations, or appearing “zoned out”). Respondents were able to indicate additional side effects not listed.

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