Special Article

Weinberg's Syndrome: A Disorder of Attention and Behavior Problems Needing Further Research

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ABSTRACT

A subset of inattentive children have an underlying problem in sustaining wakefulness ("vigilance"). This disorder of vigilance, termed Weinberg's syndrome, is characterized by difficulty in maintaining wakefulness and alertness as evidenced by (among other symptoms) motor restlessness (fidgeting and moving about, yawning and stretching, talkativeness) and complaints of tiredness. During tasks requiring concentration (continuous mental activity) such as reading, children with Weinberg's syndrome will daydream, lose interest, complain of boredom, and become increasingly restless. Napping, while infrequent, usually is not refreshing. A distinct personality described by family members and friends as kind, affectionate, compassionate, or "angelic" also seems to characterize this condition. Weinberg's syndrome has a familial pattern suggesting autosomal-dominant inheritance. Additional neurophysiologic, pharmacotherapeutic, epidemiologic, and genetic studies will be necessary for a full understanding of Weinberg's syndrome. (*J Child Neurol* 2000;15:478–480).

While still a junior faculty member on the staff of the St Louis Children's Hospital, Warren Weinberg startled the pediatric, psychiatric, and neurologic communities in 1973 by publishing an article in *The Journal of Pediatrics*[1] in which he showed that depressive illness occurred frequently in children with school difficulties and that the diagnosis could be readily made by applying specific criteria during a structured evaluation session. Weinberg had taken a bold step: he regarded affective illness (depression being one form) as a neurobiologic condition that did not respect age barriers and he then tailored for children the already accepted criteria symptoms used to diagnose depression in adults.[2] Weinberg's article so contradicted entrenched establishment views[3] that Waldo E. Nelson, editor of *The Journal of Pediatrics,* found it necessary to include along with the article a disclaimer stating: "Although this paper has been recommended for publication (subject to revision of the original manuscript) by two selected reviewers, the Editor feels it necessary to stress extreme caution (1) in identifying any child as having a depressive illness and (2) in prescribing any medication for such a disorder."[1] Slowly, over the next decade, the medical community recognized the validity of Weinberg's observations on childhood affective illness[1,4,5] and accepted that depression indeed could occur in children, using what became termed the "Weinberg Criteria" to diagnose childhood depression.[6–8] Petti even incorporated the Weinberg Criteria into the Bellevue Index of Depression.[9,10] As the American Psychiatric Association progressed through its *Diagnostic and Statistical Manual of Mental Disorders*, 3rd edition (DSM-III),[11] *DSM-IIIR*,[12] and *DSM-IV*[13] nosologies, the diagnosis of depression in children by physicians and psychologists became acceptable and even commonplace,[14,15] such that the younger generation of clinicians now cannot even conceive of the controversy that surrounded the original descriptions by Weinberg and his colleagues.[1,4,5,16]

Not being satisfied that affective illness alone was sufficient to explain many of the behavioral problems in children referred to his offices, Weinberg continued his careful clinical analyses. This meticulous observation and documentation of neurobehavioral signs and symptoms permitted Weinberg to identify a group of inattentive children who previously had been classified under the rubric of attention-deficit disorder, but seemed unique. His careful study revealed that this subgroup of children labeled as

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having attention-deficit disorder actually appeared to have an underlying problem in sustaining wakefulness (or as Weinberg called it, "vigilance"). A constellation of readily recognizable signs and symptoms that were consistent from patient to patient were evident in these children with what Weinberg called a "vigilance disorder." The most obvious clinical feature was the difficulty in maintaining wakefulness and alertness as evidenced by motor restlessness (fidgeting and moving about, yawning and stretching, talkativeness) and complaints of tiredness. In tasks requiring concentration (continuous mental activity) such as reading, these children daydreamed, lost interest, complained of boredom, and became increasingly restless. Actual napping was infrequent, and such naps tended not to be refreshing. Interestingly, these children generally had a very distinct personality described by family members and friends as kind, affectionate, compassionate, or "angelic." Through documenting the family histories and often evaluating several generations of family members, Weinberg discovered a familial pattern that seemed to suggest autosomal-dominant inheritance of problems with vigilance.

Weinberg boldly proposed a new syndrome characterized by a disturbance in vigilance and again he gave *The Journal of Pediatrics* an opportunity to publish his findings.[17] As before, the editor of the journal (this time Joseph M. Garfunkel) felt compelled to add a disclaimer at the top of the article: "We suspect that this article will stir considerable controversy. Two experienced and critical reviewers recommended it for publication, and several of us concurred, although an overlap with attention-deficit hyperactivity disorder will occur to many readers."[17] Although a few concerns were subsequently raised by other investigators,[18,19] by and large there was no controversy but rather apathy surrounding the description of Weinberg's vigilance syndrome.[17,20–22] This was mainly the result of the blind faith shown by the medical and mental-health communities in the concept of attention-deficit disorder first introduced in *DSM-III*[11] (and later refined to attention-deficit hyperactivity disorder [ADHD] in *DSM-IV*[13]). Thus, as suggested by Garfunkel in his disclaimer,[17] since some children with Weinberg's syndrome just fulfilled various criteria for the inattentive form of ADHD, there was no need for any separate characterization. However, there is no reason to think that a syndrome such as ADHD, just as with any medical disorder, should not after careful observation be separable into distinct biologic disorders that initially appear to be phenotypically the same condition. This is what Weinberg accomplished with his description of problems with vigilance[17,20–22] and now it is up to the rest of the medical community to expand the characterization of **Weinberg's syndrome**.

Duane has started this effort by using pupillometry to demonstrate hypovigilance in some children originally labeled as having ADHD.[23] Such physiologic studies substantiate earlier investigations by Satterfield and colleagues[24,25] showing electroencephalographic, evoked response, and skin conductance changes indicating underarousal in some hyperactive children. Additional neurophysiologic studies with more modern technology are warranted in Weinberg's syndrome to help understand the pattern of altered wakefulness and the associated inattention and restlessness. Epidemiologic studies will also be important in determining how often Weinberg's syndrome is the cause of poor school performance or behavior problems. In addition, the familial occurrence of Weinberg's syndrome cries out for genetic investigations, which might not only identify a molecular biologic basis, but also could help to explain the neurobiology of a variety of sleep problems. Finally, detailed pharmacotherapeutic studies will be important to identify the best treatment approach for Weinberg's syndrome, particularly whether the methylphenidate commonly prescribed for ADHD is also optimal therapy for this condition. More thorough investigations of Weinberg's syndrome will help to characterize this condition such that presumably in the future the concept of a disturbance of vigilance in children will be as acceptable as the idea of childhood affective disorders is now.[14,26]

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Announcement

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