Skin Picking in Prader-Willi Syndrome: A Pilot Study of Clinical Differences and Comorbid Symptoms

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Wigren, M. & Heimann, M. Skin Picking in Prader-Willi Syndrome: A Pilot Study of Clinical Differences and Comorbid Symptoms. Göteborg Psychological Reports, 1999, 29, No. 6. Skin picking constitutes a minor diagnostic criterion for Prader-Willi syndrome and is considered to be a syndrome specific behavior and part of a behavioral phenotype. This study surveyed different aspects and patterns of skin picking as well as related grooming behaviors and compulsive and impulsive aggressive symptoms. Parents of 37 PWS subjects, aged 12 to 30 years, participated by completing questionnaires, specially designed for the purpose of capturing specific features relevant to the clinical PWS picture. Two-thirds of the subjects displayed skin picking with a frequency ranging from chronic to transient episodic symptoms. Many subjects with skin picking also exhibited comorbid grooming behaviors and subjects with excessive skin picking also had additional problems with frequent tantrums and violent outbursts.

Key words: Prader-Willi syndrome, skin picking, trichotillomania, compulsions, impulsive aggression

Prader-Willi syndrome (PWS) is a congenital disorder associated with abnormalities in chromosome 15. It is characterized by difficulties in gross motor skills, delayed sexual development, eating disorders, sleep abnormalities and cognitive delays or dysfunctions. Hypotonia at birth is a characteristic feature that

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often causes severe feeding problems in early infancy. These problems abate, and between two to six years of age the children develop a preoccupation with food and hyperphagia, which can result in serious obesity if not controlled. Many clinical features of PWS symptomatology are related to hypothalamic dysfunctions (Butler, 1990; Holm et al., 1993; Swaab, 1997). In addition, many affected individuals suffer from emotional problems, psychopathological disorders (Whitman & Accardo 1987; Clarke, 1993; Dykens, Leckman & Cassidy, 1996) and maladaptive behaviors such as temper tantrums, impulsivity, perseverations and self-mutilating behaviors, of which skin picking is the most common (Dykens & Kasari, 1997).

The skin picking behavior constitutes a minor diagnostic criterion for the Prader-Willi syndrome (Holm et al., 1993). It is considered to be a syndrome specific behavior (Dykens & Kasari, 1997) and part of a behavioral phenotype (Dykens, 1996). Skin picking is also classified as an obsessive compulsive symptom (Hellings & Warnock, 1994) that, in some cases, causes serious damage and medical complications (Hanchett, 1998). To summarize, skin picking is a recurrent and problematic symptom in the PWS clinical picture and findings which may facilitate early clinical identification and intervention are of importance. This paper presents results from a preliminary survey on differences in clinical manifestations of skin picking and the association with other behavior problems of PWS symptomatology, which might provide additional information on this syndrome specific behavior.

Previous reports on skin picking have revealed a stability over time (Dykens, Hodapp, Walsh & Nash, 1992) and a prevalence that seems to range from 75 to 95 percent among child and adult populations (Greenswag, 1987; Clarke, Waters & Corbett, 1989; Stein, Keating, Zar & Hollander, 1994). Incidence of skin picking seems to be independent of obesity, gender and level of mental retardation. (Stein, et al., 1994; Dykens & Cassidy, 1995). Less research has focused directly on frequency of skin picking, however, generic descriptions ranging from "excessive" to "occasionally present" indicate significant within-group variability. This variability seems to cover a symptomatology ranging from chronic to transient episodic skin picking with relatively long periods of remission. Findings from some case studies indicate onset before school age (Dech & Budow, 1991; Benjamin & Bout-Smith, 1993), but according to clinical consensus, onset during late childhood is much more typical (Holm et al., 1993).

Besides skin picking, various additional forms of self-mutilating behaviors have been observed among individuals with PWS (Hanchett, 1998). Many of these behaviors show characteristics that belong to a spectrum of pathological grooming behaviors. Because phenotype behaviors sometimes display substantial variation within a syndrome specific group (Dykens, 1995), the comorbidity between different forms of grooming and self-mutilating behaviors in the PWS symptomatology should be highlighted in the future. In order to survey comorbidity in this study, hair pulling and clothes pulling were selected for both clinical and methodological reasons. These behaviors have a relatively high prevalence in the syndrome population and they also share many similar
characteristics with skin picking (e.g., behavioral topography and motor movements required). Furthermore, both hair pulling and clothes pulling sometimes result in nonfood pica behaviors that might cause medical complications due to hair or thread ingestion. (Hamdan-Ellen, 1991; Dech & Budow, 1991; Benjamin & Bout-Smith, 1993). Keeping these medical complications and the supposed high pain threshold among persons with PWS (Holm, et al., 1993; Wharton & Levine, 1998) in mind, descriptive information regarding the emergence of hair and clothes pulling among subjects with PWS remains to be addressed.

Dykens et al. (1996) reported an increased risk for obsessive-compulsive disorder in persons with PWS. Compulsions are also positively associated with self-mutilating behaviors among subjects with mental retardation. (King, 1993; Bodfish, Crawford, Powell, Golden & Lewis, 1995). According to Hellings and Warnock (1994), self-injurious behaviors included in the PWS symptomatology should be seen as obsessive-compulsive symptoms. This is also in line with what is known for the general population in which these types of grooming behaviors are classified as belonging to the spectrum of obsessive-compulsive disorders (Lenane, Swedo, Rapoport, Scerrey & Guroff, 1992; Stein, Hutt, Spitz & Hollander, 1993).

According to DSM IV (APA, 1994), compulsive and self-mutilating behaviors provide release of tension and are usually recognized as deviant by the individuals themselves, although adolescents and adults tend to deny and hide their compulsive activities (Swedo & Rapoport, 1989). However, children and adults with dysfunctions within the cognitive and social domains sometimes display repetitive compulsiveness independent of social context (Bodfish et al., 1995). Our knowledge of phenomenological aspects of skin picking - of how the behavior is exhibited and handled by the affected individuals - is still limited.

Other behavioral problems such as temper tantrums and violent outbursts are also prevalent clinical features among children and adults with PWS (Holm, et al., 1993; Dykens & Cassidy, 1995), although the relation between self-mutilating behaviors and impulsive aggression (Stein, Hollander & Liebowitz, 1993; Herpertz, Sass & Favazza, 1997) is not well-understood. This is an association that needs further exploration.

This study has three specific aims: (1) To survey the prevalence, frequency, age of onset, patterns of stability and change over time, as well as phenomenological aspects of the skin picking behavior in an adolescent and adult Swedish population; (2) To search for comorbidity between skin picking, clothes pulling and hair pulling; and (3) To relate differences in patterns of skin picking to prevalence of compulsive and impulsive aggressive behaviors.
Method

Participants

Information on 37 persons (16 males and 21 females) with PWS was gathered through parental questionnaires. The mean chronological age was 20.6 (SD = 5.9; range = 12-30). All subjects were clinically diagnosed as having PWS, and the mean age of diagnosis was 6.2 (SD = 5.0 ; range = 0-19). Parental estimates (n=35) indicate that eight of the participants had severe mental retardation, twenty moderate mental retardation, five mild mental retardation, and two subjects were judged to have an intelligence within the normal range. Parental report on level of mental retardation is consistent with the level of cognitive functioning, as has been reported in previous research (Greenswag, 1987).

At the beginning of this study, twenty-three individuals (62%) were residing at home. All of the subjects younger than 20 lived at home, and, among the adults, all but one had lived with their parents until 15 years of age, fourteen (70%) until 18 years, and eleven persons (55%) until 21 years of age. Of the nine subjects older than 26 years, five resided at home until 25 years of age. None of the subjects had any experience of living in large institutional settings.

Procedure

In cooperation with the Swedish National Association for Disabled Children and Young People all (n=54) parents to children with PWS within the age range of 12 to 30 years were selected from the Swedish Prader-Willi Syndrome Association’s membership list. Based on an estimated incidence of 1 in 10,000 live births (Holm et al., 1993), this sample probably constitutes about 30 percent of the total Swedish PWS population within the age range of 12 to 30 years. The upper age limit in this sample was selected because, up to this age, persons with impairments demanding special needs have the legal right to attend local schools and to work and reside in the local home community. This fact increased the probability that the parents reached would have reliable information about their children’s past as well as current symptomatology. The rationale behind the lower age limit was based on the assumption that maladaptive and self-mutilating behaviors are more likely to emerge during late childhood and early adolescence (Holm et al., 1993).

Instrument

A questionnaire specially designed for the purpose of capturing specific features relevant to the clinical PWS picture was constructed. It consisted of 115 questions covering the following areas: self-mutilating behaviors, compulsions, impulsive
aggressive symptoms, family setting and living arrangement. The questionnaire also included items regarding level of mental retardation, age and year of clinical diagnosis and diagnostic uncertainties (a full version of the questionnaire can be obtained from the first author).

For the purpose of this study, the following definitions were used: Skin picking: inflicting sores by picking and scratching the skin; Hair pulling: pulling hair from scalp, eyelashes or eyebrows; Clothes pulling: picking and pulling own clothes into pieces. To investigate the association between skin picking and compulsiveness, three compulsive behaviors were selected: insistence on sameness, checking and washing, compulsive symptoms that are prevalent among subjects with PWS as well as in OCD populations (Swedo & Rapoport, 1989; Hanna, 1995; Dykens, et al., 1996). Based on previous research (see Clarke et al., 1989), questions regarding frequencies of self-mutilating and tantrum behaviors allowed five different response alternatives: once a week, once a month, six times a year, once a year, and not present. Questions concerning the prevalence at different ages used the following age periods: 0-6, 7-10, 11-14, 15-18, 19-22, 23-26 and 27-30 years. These age intervals also cover common age-dependent transitions related to changes in school and/or work situations. The parents were asked to rate the frequency of skin picking, hair pulling, clothes pulling and tantrums separately for each age interval up to the current age. This procedure made it possible to gather information about age of onset, prevalence and frequency at different ages, as well as individual developmental profiles.

Information concerning phenomenological aspects of skin picking was collected through structured questions focusing on the parents opinion about the context in which the behavior was usually displayed. Did their child have a tendency to hide the behavior? Was it mostly displayed in a social or private context? In addition, the parents were asked to judge whether the exhibited behavior seemed to have an anxiety reducing effect. Information regarding behavior manifestations during tantrums was gathered through structured questions, including items concerning verbal aggressiveness and physical aggressive behaviors toward persons and/or property. The parents were also encouraged to add further descriptive information to the structured questions.

The questionnaire was tested on parents to two subjects with PWS (age of child: 12 and 19 years) before it was used in the main study. For reasons of secrecy and anonymity, the questionnaires and one reminder were distributed by the National Association for Disabled Children and Young People. Of the 54 questionnaires sent out, 40 were returned (= 74%). An additional three subjects were excluded since parental information revealed that the PWS diagnosis was uncertain or unconfirmed. On parental permission, follow-up phone calls were made to about 20% of the sample. This interview made it possible to verify questionnaire information and to get access to additional descriptive information.

Results
Skin picking was present in 24 individuals (65%) at the current age (see Table 1). A similar prevalence was observed for adults (68%; 20 years and older) and younger subjects (65%; 12 to 20 years old) as well as for men and women. Within-group variability in both the incidence level and frequency of the skin picking behavior was evident in our sample. One third of the sample (35%) did not exhibit skin picking. Eleven subjects (30%) displayed frequent skin picking (once a week to daily picking), which seems to be a more progressive, chronic-like behavioral pattern leading to persistent unhealed sores on diverse body sites. Episodic skin picking (once a year to once a month) was present in thirteen cases (35%). Among subjects with the episodic form, some persons were reported to display skin picking only once or twice a year. Additional information from telephone interviews with parents indicated that these infrequent skin picking episodes, in some cases, included serious damage to one specific body site, causing scar formation and dermatological problems that needed special care.

Table 1
Descriptive Information: Sex, Chronological Age (CA), and Reported Mental Retardation (MR)

<table>
<thead>
<tr>
<th></th>
<th>Frequent Sp¹</th>
<th>Episodic Sp</th>
<th>No symptoms</th>
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</thead>
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<tr>
<td></td>
<td>N=11</td>
<td>n=13</td>
<td>n=13</td>
</tr>
<tr>
<td>Gender</td>
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<td></td>
<td></td>
</tr>
<tr>
<td>Female (21)</td>
<td>6</td>
<td>9</td>
<td>6</td>
</tr>
<tr>
<td>Male (16)</td>
<td>5</td>
<td>4</td>
<td>7</td>
</tr>
<tr>
<td>Chronological age</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mean</td>
<td>19.9</td>
<td>22.2</td>
<td>19.8</td>
</tr>
<tr>
<td>SD</td>
<td>7.5</td>
<td>4.8</td>
<td>5.8</td>
</tr>
<tr>
<td>Range</td>
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<td>12-30</td>
<td>13-30</td>
</tr>
<tr>
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<tr>
<td>No MR (2)</td>
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<td>0</td>
<td>2</td>
</tr>
<tr>
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<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Moderate MR (20)</td>
<td>6</td>
<td>9</td>
<td>5</td>
</tr>
<tr>
<td>Severe MR (8)</td>
<td>4</td>
<td>2</td>
<td>2</td>
</tr>
</tbody>
</table>

¹ Frequent= at least once a week. Episodic=once a year – once a month

The within-group variability in both the incidence level and the reported frequency of skin picking behavior made it possible to isolate three subgroups, with similar age and gender distribution, displaying different skin picking behavioral
patterns (Table 1). One such group displayed a frequent and chronic-like pattern of skin picking (n = 11, five males; mean age = 19.9, SD = 7.5; range = 12-30), a second group displayed episodic skin picking (n = 13, four males; mean age = 22.2; SD = 4.8; range = 12-30), and a final group did not show any skin picking symptoms at all (n = 13, seven males; mean age = 19.8; SD = 5.8; range = 13-30). Although the majority of the subjects with frequent skin picking are reported to have moderate or severe cognitive dysfunctions, these levels of cognitive dysfunctions are prevalent among all groups. Normal intelligence is reported only among subjects without skin picking. Two parents to subjects without skin picking gave unspecified data concerning level of mental retardation.

Onset and developmental pattern of skin picking. The majority of the subjects with skin picking (73%) had started to show the symptom before seven years, and all but one had displayed skin picking before eleven years of age. All subjects but one with frequent skin picking did show an early start, while both early and late onset were more often observed among subjects with episodic skin picking. Two parents to adult subjects were uncertain about data concerning age of onset. Considering the recall bias for information on age of onset and symptom development among subjects within an age range of 12 to 30 years of age and our results indicating an earlier onset than expected, we chose to focus on the four youngest subjects in order to minimize recall bias. This breakdown revealed that three of the twelve-year-old children displaying frequent skin picking at current age were reported to have an onset before seven years of age, whereas the fourth child displayed episodic skin picking with an onset after ten years of age. The same pattern of onset was reported by parents to adult subjects. Information from follow-up phone calls also revealed that excessive picking was a problematic behavior that needed special care and attention from nursery school teachers.

Observations concerning the developmental pattern of skin picking revealed that, once having started, no subject ever gave up this behavior and that most of the subjects displayed a stability over time in reported frequency from onset to current age. This pattern of stability was evident among all subjects with an early onset of frequent forms while subjects with episodic skin picking displayed a more variable pattern over time. Transition from episodic to frequent skin picking was reported for only one subject in our sample.

Phenomenological aspects. The majority (70%) of the subjects involved in skin picking exhibited the behavior in private contexts (e.g., during bedtime or bathroom routines). According to parental opinion, 61 percent of the subjects tried to hide both the execution of the behavior and the inflicted damages. Consequently, it was not possible for the parents to form an opinion about the anxiety reducing effects of the behavior. Seven subjects (30%) with frequent and episodic skin picking displayed the symptoms independent of social context and, according to the parents, these subjects did not show any tendency to hide or deny the behavior or the inflicted sores. Furthermore, parents reported that skin picking displayed in social contexts seemed to have an anxiety reducing effect.

Comorbidity of self-mutilating behaviors. The prevalence and frequency of clothes pulling and hair pulling at current age is depicted in Table 2. Fourteen
subjects (38%) exhibited clothes pulling and five individuals were involved in hair pulling (13%). Age of onset for both behaviors varied from less than seven to eighteen years of age. Further analysis revealed that subjects displaying both frequent clothes pulling plus an early onset (four cases) exhibited a distinct stability in reported frequency from onset to current age, whereas subjects with episodic forms of clothes pulling displayed fluctuations in frequency over time. Furthermore, two subjects ceased to show clothes pulling during adolescence. The low prevalence of hair pulling in this population does not allow any firm conclusions, but the results indicate a wide age of onset range (before seven years of age to late adolescent) and individual changes over time, including transitions between frequent and episodic patterns.

Fourteen (58%) of the twenty-four subjects with skin picking also displayed additional self-mutilating symptoms. Two subjects were reported to exhibit all three of the self-mutilating behaviors focused on in the study and twelve persons displayed two symptoms. The most common comorbidity constellation, comprising skin picking and clothes pulling, was exhibited by ten subjects (27%). Skin picking was reported as a single symptom in nine cases (24%) and very few persons were reported to show only hair pulling (n=1) or clothes pulling (n=2). Only two subjects with frequent skin picking displayed additional high frequency comorbid symptoms. The majority of the subjects with comorbid symptoms usually exhibited one high frequency behavior in co-occurrence with an additional episodic symptom.

Prevalence of temper tantrums and compulsive behaviors. All subjects in our study were reported to show temper tantrums including impulsive-aggressive symptoms. However, within-group variability in frequency and severity was evident (Table 2). Fourteen subjects (38%) had frequent tantrums (once a week to daily tantrums), whereas 23 persons (62%) exhibited more sporadic tantrum behaviors (once a year to once a month). About two-thirds of the subjects with temper tantrums exhibited impulsive-aggressive symptoms, including physical aggression toward persons or property. Tantrums including violent acts against persons, e.g., hitting, kicking, spitting or pinching, were exhibited by eighteen subjects (49%), whereas impulsive-aggressive symptoms restricted to verbal aggression were present in twelve cases (32%).
Table 2
Skin Picking and Other Maladaptive Behaviors: Reported Comorbidity for Clothes Pulling (Cp), Hair Pulling (Hp), Temper Tantrums (Tt) and Compulsions

<table>
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<th>Frequent Sp</th>
<th>Episodic SP</th>
<th>No symptoms</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>n=11</td>
<td>n=13</td>
<td>n=13</td>
</tr>
<tr>
<td>Clothes pulling</td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>Frequent Cp (4)</td>
<td>1</td>
<td>2</td>
<td>1</td>
</tr>
<tr>
<td>Episodic Cp (10)</td>
<td>6</td>
<td>3</td>
<td>1</td>
</tr>
<tr>
<td>Hair pulling</td>
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<tr>
<td>Frequent Hp (1)</td>
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<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Episodic Hp (4)</td>
<td>0</td>
<td>3</td>
<td>1</td>
</tr>
<tr>
<td>Temper tantrums</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Frequent Tt (14)</td>
<td>10</td>
<td>3</td>
<td>1</td>
</tr>
<tr>
<td>Episodic Tt (23)</td>
<td>1</td>
<td>10</td>
<td>12</td>
</tr>
<tr>
<td>Physical Aggression (25)</td>
<td>10</td>
<td>10</td>
<td>5</td>
</tr>
<tr>
<td>Verbal Aggression (12)</td>
<td>1</td>
<td>3</td>
<td>8</td>
</tr>
<tr>
<td>Compulsions</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sameness (31)</td>
<td>10</td>
<td>11</td>
<td>10</td>
</tr>
<tr>
<td>Checking (23)</td>
<td>8</td>
<td>8</td>
<td>7</td>
</tr>
<tr>
<td>Washing (4)</td>
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</table>

The majority of the participants (84%) were reported to show one or more compulsive behaviors. Insistence on sameness, being the most prevalent compulsive symptom (see Table 2), and checking constituted a common comorbidity constellation among our subjects (57%). Four persons (all women) exhibited compulsive washing and in three cases, this compulsive behavior was part of a comorbid compulsive pattern.

Another aim of the present study was to investigate to what extent subjects with different clinical manifestations of skin picking also display a different clinical picture in terms of other related behavioral problems. Prevalence of temper tantrums and compulsive behaviors among the subjects in the three subgroups is depicted in Table 2. The majority of the subjects with a high observed frequency of skin picking were also reported to have a high frequency of temper tantrums including physical violence, whereas episodic temper tantrums were more common among individuals with episodic or no skin picking. Our results indicate that
subjects who did not show skin picking displayed a different clinical picture, including less similar grooming self-mutilating symptoms and less frequent temper tantrums with physical aggression, compared to individuals with episodic and frequent skin picking. In contrast, compulsive symptoms are common among all groups and the prevalence of insistence on sameness or checking seems to be independent of both incidence and frequency of skin picking.

Discussion

This study has shown that skin picking is a very common symptom among children and adults with Prader-Willi syndrome. Skin picking seems to be a symptom with an early onset, a stable developmental course and considerable within syndrome variability in terms of incidence and frequency. Moreover, a high degree of comorbidity with hair and clothes pulling is also evident in our sample. Furthermore, our results indicate that subjects with chronic patterns of skin picking also display high frequency impulsive aggressive symptoms.

Even though the majority of the subjects displayed skin picking, the observed prevalence among our group of adult and adolescent subjects is lower than results reported in previous studies (Greenswag, 1987; Clarke et al., 1989; Stein et al., 1994). The more stringent definition of skin picking, e.g., inflicting sores, used in this study may explain a part of the relatively low prevalence. The differences might also be due to a sample bias in this study, to the methodology employed or they may reflect differences in cultural or environmental backgrounds. Another explanation for the low prevalence may be related to the age range used in our study. Although our findings indicate an early onset, some of the youngest children may still be at risk for developing skin picking symptoms at a later date. In accordance with previous research (Stein et al., 1994; Dykens & Cassidy, 1995), incidence and frequency of skin picking among our subjects seem to be independent of both gender and level of mental retardation.

Our results indicate that skin picking may have a more early onset than previously suggested (Holm, et al., 1993). This is especially true for frequent forms of skin picking, in which case our findings are in accordance with data from some clinical descriptions and recent research revealing an onset during preschool years (Dech & Budow, 1991; Benjamin & Bout-Smith, 1993; Roof, Dimitropoulos, Thompson, Butler & Stone, 1997). Our findings regarding later onset of episodic skin picking are more in accordance with consensus (Holm et al., 1993). The subjects in our study also displayed symptom stability over time, indicating that frequency of skin picking in childhood might predict the long-term course of the behavior into adulthood. These findings have clinical implications and indicate the importance of early symptom identification and intervention.

Our findings also revealed within-group variability in terms of the phenomenological aspects of skin picking behavior. Many subjects actually exhibited coping strategies, including selecting privacy, hiding and denying,
similar to those displayed by persons with obsessive-compulsive disorder. This
concealing of symptoms has clinical implications and should be taken into account
in the clinical assessment. On the other hand, about one third of the subjects in
this study were reported to display skin picking independent of social context and
without adjustment strategies, a behavioral pattern that might be better
understood in the context of dysfunctions in cognitive and social domains.

Comorbidity of very similar grooming self-mutilating symptoms is common
among our subjects with skin picking. This comorbid behavioral pattern highlights
the importance of expanding the clinical assessment so that it also incorporates
information regarding the existence of comorbid self-mutilating behaviors. One
must also be aware of the fact that the absence of skin picking or hair and clothes
pulling among our subjects does not preclude the presence of other forms of self-
mutilating behaviors that have been observed among individuals with PWS
(Hanchett, 1998). The prevalence of hair pulling, although based on only five
subjects, is consistent with the data reported previously by Stein et al. (1994), and
reveals both a chronic and remitting course and a relatively wide range of age of
onset. This pattern has also been observed in studies on hair pulling among
children and adolescents with obsessive-compulsive disorder (Hamdan-Ellen, 1991,
Reeve, Bernstein, Christenson, 1992, Swedo, 1993). Clothes pulling also shows a
prevalence comparable to that of other self-mutilating behaviors reported by Stein
et al. (1994).

Our results indicate that the majority of subjects with excessive skin picking
also seem to have more problems with frequent tantrums and impulsive aggressive
behaviors, compared to subjects with episodic or no skin picking. These findings
imply an association between impulsive affective aggression and autoagression
due to underlying impulse control dysfunctions. Contrary to expectations,
prevalence of compulsions in our sample seems to be independent of incidence and
frequency of skin picking. Actually, time consuming and intense insistence on
sameness, washing, checking of time and personal properties was displayed by
persons with no skin picking.

Another challenging issue in the PWS symptomatology is the relation between
skin picking and the eating disorder which has an onset during preschool years.
Hyperfagia and ideation with food constitute major diagnostic criteria and, like
skin picking, they are supposed to be syndrome specific behaviors (Holm, et al.
1993; Dykens & Cassidy, 1996). Case studies also indicate that skin picking, hair
pulling and clothes pulling in some cases have oral dimensions and can result in
pica behaviors. Whether or not and how hyperfagia and ideation with food at an
early age are related to early onset of chronic patterns of skin picking are
questions for future study.

The results of this study should be considered in relation to the methodology
employed. Our findings are limited in that we relied on parental reports.
Furthermore, the assessment of onset and developmental course of the symptoms
was dependent on retrospective data and is therefore subject to recall bias which
can only be ameliorated through prospective and longitudinal research. Measuring
syndrome specific behaviors is also a problematic endeavor (Dykens, 1995).
Although reliable and valid standard psychometric instruments do have specific advantages for comparative research on populations with different etiologies, these instruments might not be sensitive enough to capture phenomenological aspects and qualitative differences within specific syndromes. In this pilot study, data were collected using a questionnaire, specially designed for the purpose of capturing syndrome specific features relevant to the clinical PWS picture - features that can not easily be obtained using general assessment tools. However, although our measures are specialized in content and focused on observable behaviors, they do have limitations in terms of reliability and validity and the hypotheses emanating from this pilot study need to be more substantially investigated. Further research, including clinical interviews and behavioral observations, is greatly needed in order to validate the estimations our instrument has yielded.

Recent findings on clinical differences among subjects with PWS indicate a possible association between symptom severity and different PWS genetic subtypes. (Cassidy et al., 1997). Consequently, our findings argue for the need to further examine the relationship between genetic status and the within-group variability, observed in the incidence, frequency and phenomenology of skin picking. Despite these limitations, the results of this pilot study indicate that excessive skin picking can, in most cases, be identified at an early age, that childhood symptomatology might predict long-term development and, thirdly, comorbid grooming behaviors and impulsive aggression are common symptoms among PWS subjects with skin picking.

References


