SELF-INJURIOUS BEHAVIOUR IN PEOPLE WITH PRADER-WILLI SYNDROME

by

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Overview

There are two volumes in the thesis which is submitted for the Clinical Psychology Doctorate at the University of Birmingham by Leah Elizabeth Bull. Volume One consists of two research chapters. The first chapter is a systematic review of the phenomenology of skin picking in people with Prader-Willi Syndrome (PWS) and interventions for skin picking in this population. Skin picking phenomenology and interventions in this population are compared to the typically developing population. This chapter was written for submission to *Clinical Psychology Review*. The second chapter is an empirical chapter using a semi-structured interview to examine the phenomenology of skin picking in people with PWS, focusing specifically on the gaps in knowledge from previous research. This research also explores management strategies developed by families and people with PWS to try to reduce skin picking. This chapter was written for submission to *Journal of Intellectual Disability Research*.

Volume Two of the thesis contains five clinical practice reports (CPRs). The first CPR is a report of a 16 year old girl with a moderate learning disability who experienced anxiety. An assessment and formulation is presented using both a Cognitive-Behavioural and systemic perspective. The second CPR is a service evaluation assessing the accessibility of a Child and Adolescent Mental Health Service for people with a learning disability. The third CPR is a single case experimental design exploring the effectiveness of Mindfulness Cognitive Behavioural Therapy for symptoms of Generalised Anxiety Disorder experienced by an older adult. The fourth CPR is a case study detailing the assessment, formulation, intervention and outcome of adapted Dialectical Behaviour Therapy for an adult with Borderline Personality

Disorder. Finally, the fifth CPR is an abstract of an oral presentation, describing a case study of the use of Cognitive-Behavioural Therapy for an adolescent experiencing symptoms of low self-esteem, anxiety, depression, self-harm and suicidal thoughts.

Dedication

For my wonderful family and a special dedication to Grandad Bull, I miss you.

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Volume One: Chapter 1 - A systematic review of the phenomenology of skin picking and interventions for skin picking in people with Prader-Willi syndrome

1.1 ABSTRACT

This systematic review examines the phenomenology of skin picking in people with a neurodevelopmental disorder, Prader-Willi syndrome (PWS), and examines interventions to reduce skin picking in this group. A literature search was conducted using known derivatives of PWS and skin picking. Inclusion and exclusion criteria were applied to the search results in two stages: abstract and full text review. Established quality criteria were used to evaluate methodology. A number of phenomenological aspects of skin picking were described, highlighting areas with limited research, for example the frequency and duration of skin picking and type of skin picked. Intervention research was characterised by a higher number of pharmacological than behavioural interventions. The majority of interventions were reported to be effective, however the findings were based on case series and single case experimental designs. The similarities between skin picking in people with PWS and the typically developing population were compared using systematic and meta-analytic reviews of research in the typically developing population. There were more similarities than differences in skin picking in these two different populations. It is possible that interventions used for people who skin pick in the typically developing population could be effective for people with PWS. The clinical implications of the findings are discussed.

1.2 INTRODUCTION

Skin picking and trichotillomania (hair pulling) occur in the typically developing population with prevalence estimates of 1% to 4% (Odlaug & Grant, 2008; Roberts, O’Connor and Bélanger, 2013). Trichotillomania is defined by the removal of hair from any part of the body that is associated with relief from tension or pleasure (American Psychiatric Association, 2013). Skin picking has been characterised as a repetitive action that causes damage to the skin such as wounds, scars and infections (Van Ameringen et al., 2014). Trichotillomania and skin picking can cause significant distress physically, psychologically and socially (Grant, Stein, Woods & Keuthen, 2012; Roberts, O’Connor & Bélanger, 2013).

These behaviours are increasingly grouped together with other behaviours such as nail biting using the term body focussed repetitive behaviours (Teng, Woods, Twohig & Marcks, 2002) which refers to a group of behaviours that are directed towards the self and involves the removal of bodily material such as hair, skin or nails. They are thought of as habitual behaviours with shared topographies that can result in damage and can be seen as self-injurious (Teng et al., 2002, Grant et al., 2012; Roberts, O’Connor, Aardema & Bélanger, 2015).

Trichotillomania and skin picking typically involve the removal of an imperfection, either hair or skin with variable frequency (Roberts et al., 2013; Bohne, Keuthen & Wilhelm, 2005). They share a similar age of onset during adolescence (Odlaug & Grant, 2008; Lochner, Simeon, Niehaus & Stein, 2002). Common functions or triggers are shared such as emotion regulation, reduction of tension and removing body irregularities (Bohne et al. 2005; Roberts et al., 2013). Psychological models for understanding these behaviours are similar and include the presence of internal triggers (urges or thoughts), external triggers (setting, presence of imperfection), positive reinforcement through sensory stimulation and negative reinforcement through relief of negative emotions (Roberts et al., 2013). These behaviours are also comorbid. Snorrason, Belleau and Woods (2012) found that 83% of participants who engaged in skin picking also met criteria for at least one other body focussed repetitive behaviour such as trichotillomania. Furthermore, trichotillomania and skin picking show a familial incidence (Snorrason et al., 2012; Monzani et al., 2012; Bienvenu et al., 2009). The treatment approaches for trichotillomania and skin picking are also similar, for example habit reversal therapy (Flessner, Busch, Heideman & Woods, 2008).

A number of research studies report comorbidity of trichotillomania, skin picking and OCD (see Stein et al., 2010 for a review). The association between trichotillomania and skin picking is reflected in recent changes in *The Diagnostic and Statistical Manual of Mental Disorders* (5th ed.; DSM–5; American Psychiatric Association, 2013). DSM-5 locates skin picking and trichotillomania under Obsessive and Compulsive related behaviours implying shared features with Obsessive-Compulsive Disorder (OCD). Similarities have also been noted between OCD and body focussed repetitive behaviours as they are all repetitive and often function to reduce anxiety with acknowledgment that OCD is often more cognitively driven (Van Ameringen, Patterson & Simpson, 2014; Grant, Odlaug & Kim, 2010; Stein et al., 2010).

Body focused repetitive behaviours are common in people with an intellectual disability and those with genetic disorders (Barnard-Brak, Rojahn, Richman, Chestnut & Wei, 2015; Arron et al., 2011; Lang et al., 2010; Lang et al., 2009; Rojahn, Schroeder & Hoch, 2008). For example, biting, trichotillomania and skin picking are evident in people with Fragile-X syndrome (Symons, Byiers, Raspa, Bishop & Bailey Jr, 2010; Hall, Lightbody & Reiss, 2008) and biting is almost universal in Lesch-Nyhan syndrome (Robey, Reck, Giacomini, Barabas & Eddey, 2003; Anderson & Ernst, 1994). Biting, trichotillomania, skin picking and nail extraction is seen in people with Smith-Magenis syndrome (Taylor & Oliver, 2008; Martin, Wolters & Smith, 2006; Finucane, Haines Dirrigl & Simon, 2001; Dykens & Smith, 1998). However, there is limited research exploring the phenomenology of these behaviours collectively as they have been grouped in the typically developing population. This may be because these specific behaviours have not been well researched and therefore their phenomenology and topography are not well defined. In the typically developing population similarities between body focused repetitive behaviours have led to knowledge sharing and helped to inform future research and potential intervention strategies for these behaviours (Grant et al., 2012).

Prader-Willi syndrome (PWS) is one neurodevelopmental disorder in which one of these body focused repetitive behaviours, skin picking, has been researched and described frequently. PWS is caused by the loss of paternal information from chromosome 15q11.2 – 13. This information can be missing due to a paternal deletion, a maternal uniparental disomy (mUPD) or an imprinting error (Cassidy & Driscoll, 2009). A population study in the UK indicated that PWS was prevalent in approximately one in 29,000 live births (Whittington et al., 2001). Generally people with PWS have a mild to moderate intellectual disability (Whittington et al., 2004) with relative strengths in visual processing (Whittington et al., 2004) and difficulties in mathematics and short term memory (Bertella et al., 2005). Physically, PWS is associated with characteristic facial features and failure to thrive in infancy that later develops to hyperphagia in early childhood (Holm et al., 1993). There is also a well described behavioural phenotype with temper outbursts, repetitive behaviour and skin picking being common (Holm et al., 1993; Holland et al., 2003).

There has been a larger number of research studies exploring skin picking in people with PWS than other groups within the intellectual disability population, perhaps due to its high prevalence (Arron et al., 2011; Morgan et al., 2010). Prevalence rates reported vary from 52% to 96%, with the majority of studies reporting rates between 64% and 78% (Wigren & Heimann, 2010; Buono et al., 2005; Wigren & Hansen, 2005; Holland et al., 2003; Boer & Clarke, 1999; Feurer et al., 1998; Cassidy et al., 1997). The difference in these prevalence rates may be due to the methodologies used and the information people were provided with at recruitment. Some of the studies focused on describing skin picking, either using bespoke or established skin picking measures. There may have been a sample bias in those that used specific skin picking measures as people who had difficulties with skin picking or who engaged in skin picking were perhaps more likely to take part. Despite a number of studies describing the phenomenology of skin picking there has been no systematic review of this literature. Similarly, research has explored different interventions (Miller & Angulo, 2013; Wilson, Iwata & Bloom, 2012), although few studies have been published in this area. Therefore, it would be beneficial to explore the phenomenology of skin picking and interventions for skin picking in people with PWS systematically. It is also important to explore the similarities between these behaviours in the typically developing and intellectual disability populations. If there are similarities this could facilitate understanding of these behaviours and interventions.

1.2.1 Aims

The aims of this systematic review were to:

- evaluate the quality of the research literature exploring skin picking and interventions for skin picking in people with PWS
- describe the phenomenology of skin picking in people with PWS
- examine interventions used for skin picking in people with PWS and the efficacy of those interventions
- explore whether the conceptualisation of skin picking evident for the typically developing population would be helpful in this population by examining similarities and differences.

1.3 METHOD

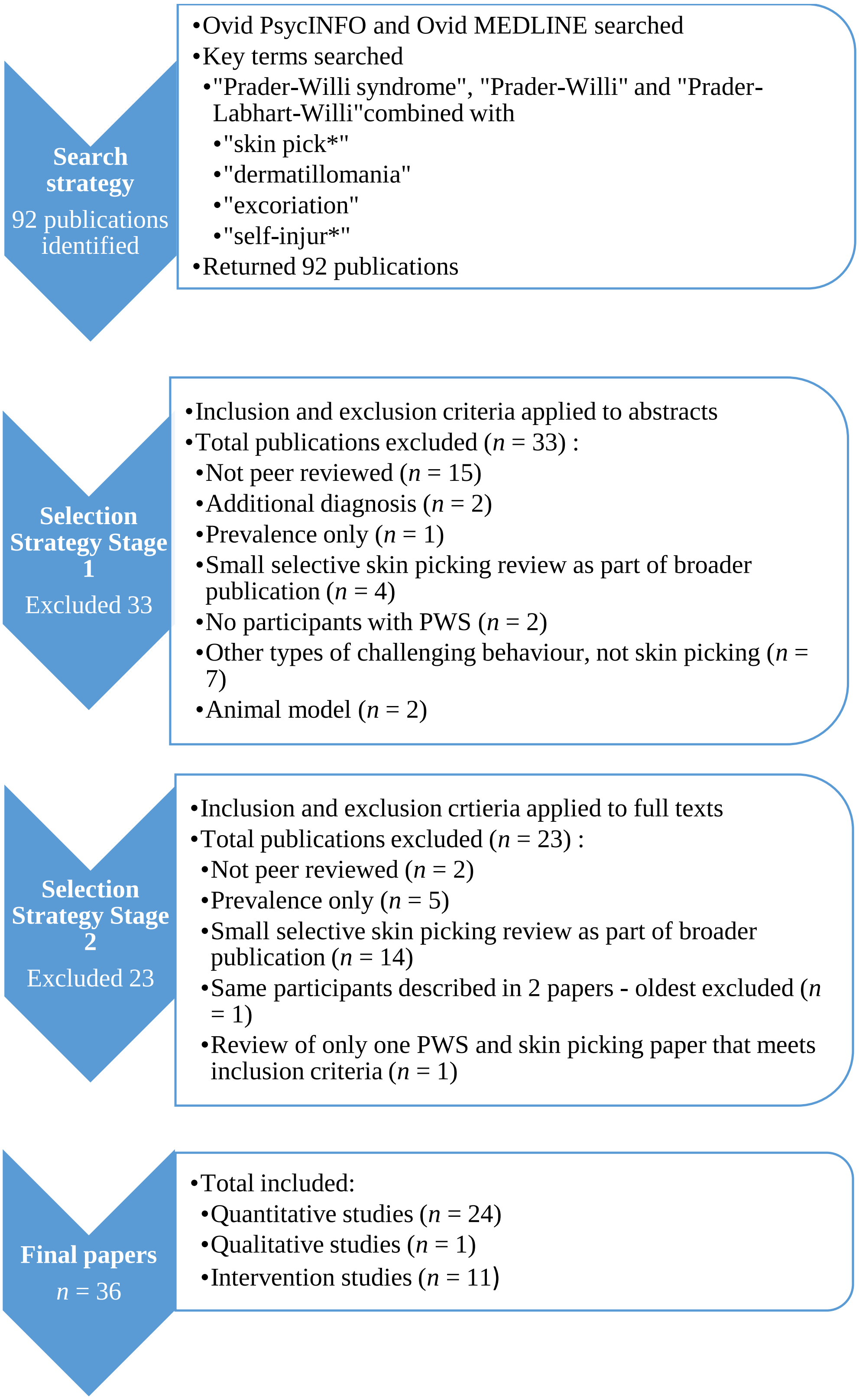
1.3.1 Search Strategy

Key databases searched were Ovid PsycINFO and Ovid MEDLINE. Literature searches were conducted by combining search terms for PWS with search terms for skin picking. The search terms used for PWS were “Prader-Willi syndrome”, “Prader-Willi” and “Prader-Labhart-Willi”. An initial search of skin picking was conducted to find out whether there were any different derivatives of skin picking to ensure a more inclusive search of skin picking publications, the following search terms were used, “skin pick*”, “dermatillomania”, “excoriation” and “self injur*”. Both databases were searched on 16th January 2015. The inclusion dates for the Ovid PsycINFO search were 1967 to January week two 2015 and for Ovid MEDLINE were 1946 to January week two 2015.

1.3.2 Selection Strategy

The two searches returned 92 publications. Inclusion and exclusion criteria were applied to each publication in order to select publications for review systematically. Publications were included if they reported on participants with PWS and if it was a peer reviewed publication. Publications were excluded that included participants with PWS and an additional diagnosis such as additional genetic syndromes, if the research only documented the prevalence rates of skin picking in people with PWS, if there was only a short selective review of skin picking included as part of a broader description of PWS and any animal model research.

There were two stages to the selection strategy. *Figure 1.1* shows a flow chart detailing the stages of selection and the publications extracted at each stage. The first stage involved reviewing titles and abstracts. Publications were only excluded at this stage if it was clear that they did not meet the inclusion criteria. Stage two involved reviewing full texts of the publications whilst applying the same inclusion and exclusion criteria. After completion of the selection strategy 36 studies were included in the current review. This included quantitative and qualitative papers and intervention studies.



Search strategy

92 publications identified

- Ovid PsycINFO and Ovid MEDLINE searched
- Key terms searched
  - "Prader-Willi syndrome", "Prader-Willi" and "Prader-Labhart-Willi"combined with
    - "skin pick*"
    - "dermatillomania"
    - "excoriation"
    - "self-injur*"
- Returned 92 publications

Selection Strategy Stage 1

Excluded 33

- Inclusion and exclusion criteria applied to abstracts
- Total publications excluded (*n* = 33) :
  - Not peer reviewed (*n* = 15)
  - Additional diagnosis (*n* = 2)
  - Prevalence only (*n* = 1)
  - Small selective skin picking review as part of broader publication (*n* = 4)
  - No participants with PWS (*n* = 2)
  - Other types of challenging behaviour, not skin picking (*n* = 7)
  - Animal model (*n* = 2)

Selection Strategy Stage 2

Excluded 23

- Inclusion and exclusion crtieria applied to full texts
- Total publications excluded (*n* = 23) :
  - Not peer reviewed (*n* = 2)
  - Prevalence only (*n* = 5)
  - Small selective skin picking review as part of broader publication (*n* = 14)
  - Same participants described in 2 papers - oldest excluded (*n* = 1)
  - Review of only one PWS and skin picking paper that meets inclusion criteria (*n* = 1)

Final papers

n = 36

- Total included:
  - Quantitative studies (*n* = 24)
  - Qualitative studies (*n* = 1)
  - Intervention studies (*n* = 11)

Figure 1.1: Process of selecting publications for review

1.3.3 Quality Review

Using quality criteria allows for an objective review of the literature (Downs & Black, 1998). The research studies selected for the current review include diverse methodologies, therefore, a number of different quality criteria were applied to ensure that each study was assessed for its quality using aspects pertinent to the particular research design. For quantitative studies criteria from Sale & Brazil (2004) were used that assess internal validity, external validity and reliability. Sale & Brazil (2004) also developed a similar set of quality criteria for qualitative research so this was used for the qualitative studies. This assesses the research credibility, transferability, dependability and confirmability. Finally, for intervention research the Downs & Black (1998) quality criteria were used. These assess the quality of reporting, external validity, internal validity and power.

For each criterion a study could score one or zero. A higher score reflected higher quality. A coding scheme was applied to identify the quality of research as either poor, satisfactory or good. All scores were converted to a percentage for each subscale within the criteria and for the overall score. Papers were then categorised for each subscale and for the overall score as poor, satisfactory or good (Poor - 0 - 33.33%, satisfactory - 33.34% - 66.67% and good 66.68% - 100%).

1.4 RESULTS

1.4.1 Phenomenology of skin picking in people with PWS

Table 1.1 shows a summary of the quantitative studies focusing upon the phenomenology of skin picking in people with PWS and *Table 1.2* shows a summary table of quantitative studies that considered associations between skin picking and other variables only and differences within people with PWS or between comparison groups. *Table 1.3* displays a summary of the qualitative research study. Finally, *Tables 1.4* and *1.5* provides an overview of the quality of the quantitative and qualitative research studies respectively. *Appendix A* shows the item by item scores for each quality criterion for each of the papers reviewed.

Table 1.1: Summary of the methodology and findings of quantitative studies assessing the phenomenology of skin picking in people with PWS

Authors	***Participants***						***Outcome measures***	***Key findings***												
	n	*Age*	*% male*	*Genetic subtype*	*Intellectual disability*	*Comparison group*		*Age of onset*	*Most common sites of skin picking*	*Type of skin*	*Method used*	*Frequency*	*Duration*	*Severity*	*Number of skin picking sites*	*Triggers/functions*	*Associations*	*Differences*	*Interventions*	*Other*
Hall, Hammond & Hustyi (2013)	1	12	100%	mUPD	Mild	No comparison group	Experimental functional analysis (Iwata et al., 1994), Heart rate and activity. Behaviour observation with high inter-rater reliability (skin picking)	-	Face, chest/stomach, arms, fingers, legs, and feet	-	Fingernails or fingers	Daily	-	Broken skin, bleeding, previous hospitalisations	-	Automatic reinforcement	-	-	First aid, stress ball	Heart rate increased during skin picking but activity did not also increase. Heart rate increased the most when damage was caused.
Hustyi et al. (2013)	55	6-25	45%	51% deletion 42% mUPD 7% unknown	-	No comparison group	Self-Injury Trauma Scale (Iwata et al., 1990), Functional Analysis Screening Tool (Iwata et al., 2013)	-	Finger, toes, leg and arms	-	-	-	-	Average severity was superficial break in the skin.	Number of sites were an average 5-8 sites.	Most common sensory stimulation	No link between skin picking and age or gender	No genetic subtype differences	Most common interventions used behavioural or basic first aid	-
Wigren & Heimann (2010)	37	12-30 (mean 20.6)	43%	-	Estimated average to severe	No comparison group	Bespoke questionnaire - phenomenology of skin picking, compulsions, aggression	73% started skin picking before age 7	-	-	-	Once a week-daily or once a month-once a year	-	-	-	-	Participants with more severe skin picking showed more frequent temper outbursts	-	-	70% reported to do it privately, 61% hide skin picking, co morbidity with hair pulling reported for 13% and clothes pulling 38%
Buono, Scannella & Palmigiano (2010)	10	1-47 (mean 14.3)	60%	-	Mild to moderate	25 participants with DS (64% male), 49 participants with ASD (59% male)	Self-Injurious Behavior Schedule (Buono et al., 2006)	90% of the PWS engaged in SIB before 7 years old	Hands, arms, and legs (significantly different to both DS and ASD groups)	-	-	-	-	-	-	-	-	Significantly more skin picking than both other groups	-	30% also engaged in hair pulling

- Not reported or none conducted

Authors	*Participants*						*Outcome measures*	*Key findings*												
	n	*Age*	*% male*	*Genetic subtype*	*Intellectual disability*	*Comparison group*		*Age of onset*	*Most common sites of skin picking*	*Type of skin*	*Method used*	*Frequency*	*Duration*	*Severity*	*Number of skin picking sites*	*Triggers/functions*	*Associations*	*Differences*	*Interventions*	*Other*
Morgan et al. (2010)	67	-	54%	31% deletion 31% mUPD 3% imprinting defect 9% unknown	-	No comparison group	Bespoke questionnaire - psychiatric and medical history, phenomenology of skin picking, Skinpicking Scale (Keuthen et al., 2001) modified, Milwaukee Inventory for the Dimensions of Adult Skin-picking (Walther et al., 2009) modified, Spence Child Anxiety Scale for Parents (Nauta et al., 2004), Swanson, Nolan and Pelham Questionnaire: Fourth Edition (Swanson, 1992), Paediatric Quality of Life Inventory-Parent Proxy (Varni et al., 2001)	-	Hands, legs, face and arms	Most common type of skin picked was scabs, itchy skin, insect bites, inflamed skin, healthy skin, scars and pimples	Fingernails or fingers	-	Less than a minute up to 90 minutes. Majority reported 1-5mins (28%)	42% reporting symptoms within a clinically significant range. Most common damage reported was minor sores, bleeding, scars and infections.	-	Most common antecedents were at school, waiting, watching television, in the car, in bed, reading.	Skin picking positively correlated with anxiety, inattention and oppositionality and was associated with focused, compulsive skin picking. Problems in health related quality of life also correlated positively with skin picking severity.	No difference in skin picking severity between genetic subtypes.	-	5% also engaged in hair pulling
Didden, Korziliu s & Curfs (2007)	119	4-49 (mean 22)	53%	30% deletion 11% mUPD 4% imprinting defect 31% unknown	Mild to moderate	No comparison group	Self-Injury Checklist (Bodfish et al., 1995) items added, Compulsive Behavior Checklist (Gedye, 1992), Questions About Behavioral Function (Matson & Vollmer, 1995)	Mean age of onset was 6 years.	Arms, legs and face	-	-	-	-	Mild to moderate severity reported by 73% of the sample	-	Most common function was either positive sensory reinforcement or negative reinforcement by escape from arousal or discomfort.	Significant positive correlation between skin picking and severity and number of compulsive behaviours. Age of onset of skin picking and compulsive behaviour correlated.	No ID differences in skin picking. No genetic subtype differences.	4% reported that they had received an intervention	Other SIB reported - 9% hitting, 9% hair pulling, 8% head banging, 8% eye gouging
Buono et al. (2005)	15	1-29. Separate age groups - 1-12years (*n*=7) and 13+ (*n*=8)	47%	-	Mild to moderate	No comparison group	Bespoke questionnaire - phenomenology of skin picking	Majority reported onset before 6 years old.	Hands, arms and legs	All reported picking when cut or insect bite.	-	Majority of sample reported "frequent" skin picking	-	"High intensity"	-	-	-	No ID or age differences	-	-

- Not reported or none conducted

Authors	*Participants*						*Outcome measures*	*Key findings*												
	n	*Age*	*% male*	*Genetic subtype*	*Intellectual disability*	*Comparison group*		*Age of onset*	*Most common sites of skin picking*	*Type of skin*	*Method used*	*Frequency*	*Duration*	*Severity*	*Number of skin picking sites*	*Triggers/functions*	*Associations*	*Differences*	*Interventions*	*Other*
Symons et al. (1999)	62	3-44 (mean 18)	39%	61% deletion 18% mUPD 21% unknown	Mild to moderate	Comparison group 29 school children with ID/ASD/Other developmental disabilities, 76% male, majority severe ID	Bespoke questionnaire - self-injury body grid	-	Legs, head and arms	-	-	-	-	-	-	-	-	Significant positive correlation between age and number of sites of skin picking for deletion subtype. Genetic subtype difference that deletion subtype picked more sites than mUPD group. Compared to control group the PWS group engaged in skin picking whereas comparison group more likely to bang, hit or bite.	-	Other SIB reported - 14% head banging, 9% hair pulling
Feurer et al. (1998)	75	4-41 (mean 11.4)	41%	71% deletion 16% mUPD 13% unknown	Mean IQ 61	No comparison group	Compulsive Behavior Checklist (Gedye, 1992)	-	-	-	-	-	-	Ranged from mild-severe. Majority mild or moderate	-	-	-	-	-	17% engaged in hair pulling

- Not reported or none conducted

Table 1.2: Summary of the methodology and findings of quantitative studies assessing associations and differences observed in skin picking in people with PWS

Authors	***Participants***						***Outcome measures***	***Key findings***		
	n	*Age*	*% male*	*Genetic subtype*	*Intellectual disability*	*Comparison group*		*Associations*	*Differences*	*Other*
Dykens (2014)	123	4-48 (mean 14.35)	54%	21% Type I deletion 30% Type II deletion 35% mUPD 14% other causes	Mean IQ 66	No comparison group	Yale-Brown Obsessive Compulsive Scale (Goodman et al., 1989) with skin picking separated, Recreation and Leisure Questionnaire (Sellinger et al., 2006).	Significant negative correlation between skin picking and physical activity. Significant positive correlation between skin picking and puzzles, games and watching television	-	-
Arron et al. (2011)	-	Mean 17.04	53%	-	96% verbal, 73% mobile, 96% able or partly able to feed, dress and wash	Angelman, Cri du Chat, Cornelia de Lange, Fragile X, Lowe and Smith-Magenis syndrome. Group with ID of heterogenous aetiology	Wessex Scale (Kuschlick et al., 1973), The Challenging Behaviour Questionnaire (Hyman et al., 2002).	-	PWS were more likely than group with ID to show self-injury and more likely than all the groups to show rubbing/scratching. No age, gender differences between self-injury	-
Hiraiwa et al. (2007)	165	2-31 Separate age groups - Young children *n*=34 Primary school children *n*=57 Adolescents *n*=45 Young adults *n*=29	-	-	-	42 participants with ID matched for age, gender and IQ	Bespoke questionnaire	-	PWS showed significantly more self-injury than the comparison group. Prevalence of skin picking appeared to increase with age - rates 35% in young child group, 56% in primary school group, 62% in adolescent group and 59% in young adult group	-

- Not reported or none conducted

Authors	***Participants***						***Outcome measures***	***Key findings***		
	n	*Age*	*% male*	*Genetic subtype*	*Intellectual disability*	*Comparison group*		*Associations*	*Differences*	*Other*
Hartley et al. (2005)	65	12-45 (mean 23.81)	45%	62% deletion 35% mUPD 3% imprinting defect	IQ range 45-83	No comparison group	The Reiss Screen for maladaptive behaviour (Reiss, 1988)	Significant positive correlation between self-injury and IQ	Higher self-injury score for the deletion subtype compared to mUPD	-
Wigren & Hansen (2005)	58	5-18 (mean 11.6)	53%	-	-	No comparison group	Conners Parent Rating Scale-48 (Connors, 1989), The Childhood Routines Inventory (Evans et al., 1997)	No correlation with ADHD, impulsive-hyperactivity. Significantly positively correlated to insistence on sameness and conduct problems.	-	-
Dykens (2004)	240	3-50 (mean 20.78). Separated into age groups - Young children *n*=56, Adolescent *n*=80, Young adult *n*=59 and Older adult *n*=45.	49%	47% deletion 10% mUPD 1% other causes 42% unknown	Mean IQ 64	No comparison group	Yale-Brown Obsessive Compulsive Scale (Goodman et al., 1989) Child Behavior Checklist (Achenbach, 1991)	-	For children and adolescents females showed more skin picking. Prevalence of skin picking differed across age groups. Young children (54%) and the older adults (43%) showed similar prevalence rates. The adolescent group (72%) showed the highest prevalence followed by young adults (65%)	-

- Not reported or none conducted

Authors	***Participants***						***Outcome measures***	***Key findings***		
	n	*Age*	*% male*	*Genetic subtype*	*Intellectual disability*	*Comparison group*		*Associations*	*Differences*	*Other*
Holland et al. (2003)	65	Mean 20.8	-	-	Mean IQ 63	42 participants with ID	Diagnostic checklist rated for frequency and severity, Developmental Behaviour Checklist (Einfeld & Tonge, 1989), the Aberrant Behavior Checklist (Aman et al. 1986)	In PWS group there was a significant positive correlation between skin picking and severity of eating disorder.	Prevalence rate of skin picking in PWS group (78%) was significantly higher than the ID group (35%).	Skin picking loaded together with mood swings, stubbornness and argumentativeness. Authors suggest that skin picking possibly related to low mood
Dimitropoulos et al. (2001)	105	2-6	46%	74% deletion 15% mUPD 11% unknown	-	Down syndrome group - *n*=56 (63% male). Typically developing group - *n*=76 (50% male)	Compulsive Behavior Checklist (Gedye, 1992), bespoke questionnaire asking about temper tantrums and food related behaviours	-	No age differences. Ages 2-3 = 27% Ages 4-5 = 40%	-
Akefeldt & Gillberg (1999)	44	0-36	64%	-	Mild to moderate	16 participants with ID (56% male), borderline-moderate ID	Clinical diagnosis interview based on Holm et al. (1993), Modified Greenswag Interview (Greenswag, 1987), SSBPPQ (O'Brian, 1992), the Rutter Parent Questionnaire (Rutter et al., 1970)	-	PWS had higher scores for skin picking than the comparison group. Lower mean skin picking score in the younger compared to older age group	-

- Not reported or none conducted

Authors	***Participants***						***Outcome measures***	***Key findings***		
	n	*Age*	*% male*	*Genetic subtype*	*Intellectual disability*	*Comparison group*		*Associations*	*Differences*	*Other*
Boer & Clarke (1999)	210	3-51 (mean 17.1)	-	-	-	No comparison group	Society for the Study of Behavioural Phenotypes Postal Questionnaire (O'Brian, 1995)	-	No significant difference between children and adults in the prevalence of skin picking but higher	-
Dykens, Cassidy & King (1999)	46	6-42 (mean 17)	39%	50% deletion 50% mUPD	Mean IQ 67	No comparison group	Child Behavior Checklist, Yale-Brown Obsessive-Compulsive Scale	-	Higher scores for skin picking in the deletion group compared to mUPD	-
Dykens & Smith (1998)	35	Mean 9	43%	-	-	35 participants with Smith-Magenis (43% male) syndrome and 35 participants with ID (43% male)	Child Behavior Checklist (Achenbach, 1991)	-	Higher skin picking than participants with Smith-Magenis syndrome and participants with ID	-
Cassidy et al. (1997)	54	0-57	56%	70% deletion 30% mUPD	Mean IQ 67	No comparison group	Diagnostic criteria assessment based on Holm et al., 1993)	-	Significantly higher prevalence of skin picking in deletion compared to mUPD group	-
Clarke et al. (1996)	30	16-44	50%	70% deletion 7% mUPD 3% other cause 20% not reported	-	30 participants matched for age, gender and ID	Aberrant Behavior Checklist (Aman et al., 1985a, 1985b, 1987)	-	Significantly higher mean self-injury score compared to matched comparison group	-

- Not reported or none conducted

Authors	***Participants***						***Outcome measures***	***Key findings***		
	n	*Age*	*% male*	*Genetic subtype*	*Intellectual disability*	*Comparison group*		*Associations*	*Differences*	*Other*
Dykens et al. (1992)	21	13-46 (mean 25) Separated into age groups – 13-19 (*n*=6), 20-29 group (*n*=9), 30-46 group (*n*=6)	86%	-	Mean IQ 62	No comparison group	Child Behavior Checklist (Achenbach & Edelbrook, 1983)	-	Highest skin picking score evident in adolescent group	-

- Not reported or none conducted

Table 1.3: Summary of the methodology and findings of qualitative studies assessing the phenomenology of skin picking in people with PWS

Authors	***Participants***						***Outcome measures***	***Key findings***												
	n	*Age*	*% male*	*Genetic subtype*	*Intellectual disability*	*Comparison group*		*Age of onset*	*Most common sites of skin picking*	*Type of skin*	*Method used*	*Frequency*	*Duration*	*Severity*	*Number of skin picking sites*	*Triggers/functions*	*Correlations*	*Differences*	*Interventions*	*Other*
Didden et al. (2008)	10	29-54 (mean 41)	50%	40% deletion 20% mUPD 40% unknown	IQ range 57-77	No comparison group	Bespoke semi-structured interview	-	Face, legs, arms and feet	-	-	Varied	-	Varied	-	7 said picked due to skin itching. Some reported picking when feeling anxious or bored.	-	-	8 stated that nothing could help stop it but 2 said short nails, being busy and rewards for not picking helped	Reported a negative view of their skin picking.

- Not reported or none conducted

Table 1.4: Quality review of quantitative studies using Sale and Brazil (2004) quality criteria

Authors	**Internal validity**	**External validity**	**Reliability**	**Overall**
Dykens (2014)	Poor	Satisfactory	Good	Satisfactory
Hall, Hammond & Hustyi (2013)	Good	Satisfactory	Good	Satisfactory
Hustyi et al. (2013)	Satisfactory	Good	Good	Good
Arron et al. (2011)	Satisfactory	Satisfactory	Good	Good
Wigren & Heimann (2010)	Satisfactory	Good	Poor	Satisfactory
Buono, Scannella & Palmigiano (2010)	Poor	Satisfactory	Good	Satisfactory
Morgan et al. (2010)	Satisfactory	Good	Good	Good
Didden, Korziius & Curfs (2007)	Satisfactory	Good	Good	Good
Hiraiwa et al. (2007)	Poor	Good	Poor	Satisfactory
Buono et al. (2005)	Satisfactory	Satisfactory	Poor	Satisfactory
Hartley et al. (2005)	Satisfactory	Satisfactory	Good	Satisfactory
Wigren & Hansen (2005)	Poor	Good	Good	Satisfactory
Dykens (2004)	Poor	Satisfactory	Good	Satisfactory
Holland et al. (2003)	Satisfactory	Satisfactory	Good	Satisfactory
Dimitropoulos et al. (2001)	Satisfactory	Good	Good	Good
Akefeldt & Gillberg (1999)	Satisfactory	Satisfactory	Good	Satisfactory
Dykens, Cassidy & King (1999)	Poor	Good	Good	Satisfactory
Boer & Clarke (1999)	Poor	Good	Good	Satisfactory
Symons et al. (1999)	Poor	Good	Poor	Satisfactory
Dykens & Smith (1998)	Satisfactory	Satisfactory	Good	Satisfactory
Feurer et al. (1998)	Satisfactory	Satisfactory	Good	Satisfactory
Cassidy et al. (1997)	Poor	Good	Poor	Satisfactory
Clarke et al. (1996)	Satisfactory	Satisfactory	Good	Satisfactory
Dykens et al. (1992)	Poor	Satisfactory	Poor	Satisfactory

Table 1.5: Quality review of qualitative studies using Sale and Brazil (2004) quality criteria

Authors	**Credibility**	**Transferability**	**Consistency**	**Confirmability**	**Overall**
Didden et al. (2008)	Satisfactory	Satisfactory	Poor	Poor	Satisfactory

Overall, the quality of the majority of studies was satisfactory with five studies falling within the good range overall. Areas that fell within the poor range were the internal validity and reliability subscales. Ten studies fell within the poor category for internal validity which was usually due to confounding variables not being discussed or controlled for. For reliability, six studies fell within the poor range and this was often because the studies used non standardised measures and did not comment or attempt to establish the reliability of this measurement tool. Only one study used qualitative methods and was the only study to use participants with PWS as informants rather than parents or carers. All remaining studies were quantitative.

1.4.1.1 Age of onset

Four studies reported ages at which skin picking first began. Age of onset was reported to be early for participants with the majority reporting onset before ages six or seven (Wigren & Heimann, 2010; Buono, Scannella & Palmigiano, 2010; Buono et al., 2005). One study reported that the mean age of onset was six years old (Didden, Korzilius & Curfs, 2007). This study had a good overall quality rating and was the only study within this group to use well-established and validated measures.

1.4.1.2 Sites

Eight studies examined the common sites of skin picking in people with PWS, see *Table 1.1 and 1.3.* Multiple sites were identified with the most common being head, arms, hands and legs and was reasonably consistently reported by the eight studies. The one self-report study also supported these results (Didden et al., 2008). Only one study documented the number of skin picking sites which was five to eight sites (Hustyi et al., 2013). This study fell within the good range based on quality criteria, however the number of sites was reported as a mean but this may not have been an appropriate measure of central tendency as there were outliers skewing the data.

1.4.1.3 Type of skin

Only two studies documented the type of skin that participants picked, this included both healthy skin and skin with an imperfection such as a spot, scab and insect bites (Morgan et al., 2010; Buono et al., 2005). One of these studies was rated as good overall (Morgan et al., 2010).

1.4.1.4 Method used

Two studies explored the method used by participants to skin pick and both found that participants used fingers or fingernails to skin pick (Hall, Hammond & Hustyi, 2013; Morgan et al., 2010). One of these studies was a single case study that operationally defined skin picking for an observer and only included the use of fingers as the method (Hall et al., 2013). The other study had a sample size of 67 and was rated as good (Morgan et al., 2010). This study assessed method used by giving a set of answers to select. Some respondents in this study reported biting and some object use such as scissors but this was only demonstrated by a small percentage of the sample (17% and 1.5% respectively).

1.4.1.5 Frequency

Three studies reported the frequency of skin picking (Hall et al., 2013; Wigren & Heimann, 2010; Didden et al., 2008). This was reported to vary widely from daily to once a year with no studies identifying the modal frequency. One study reported skin picking to be “frequent” but the authors did not provide a clear definition of this (Buono et al., 2005).

1.4.1.6 Duration

Only one study examined the duration of skin picking (Morgan et al., 2010) and was a good quality study. The study reported wide ranging durations from less than a minute to around an hour and a half. The most common duration appeared to be between one and five minutes.

1.4.1.7 Severity

Severity of skin picking was reported in two different ways, by the damage to the skin and by “severity” or “intensity” although these terms were not always operationally defined. Damage reported included bleeding, scars and infections (Hall et al., 2013; Hustyi et al., 2013; Morgan et al., 2010). Severity ranged from mild to severe (Didden et al., 2007) and a “high intensity” was reported by Buono et al. (2005). One high quality study identified that over 40% of their sample showed skin picking within a clinically significant range (Morgan et al., 2010) according to a standardised measure of skin picking (The Skin Picking Scale; Keuthen et al., 2001).

1.4.1.8 Triggers or functions

Four informant report studies commented on the triggers or functions to skin picking. Events that seem to make skin picking more likely were watching television, waiting for something, being in the car or in bed (Morgan et al., 2010). Common functions appeared to be either positive reinforcement from sensory stimulation or negative reinforcement by providing relief from arousal or discomfort (Hall et al., 2013; Hustyi et al., 2013; Didden et al., 2007). All three of these studies used standardised measures for assessing the function of challenging behaviour. Self-reports indicated that participants engaged in skin picking when they felt an itch and that being anxious or bored could also trigger skin picking (Didden et al., 2008).

1.4.1.9 Related variables

Some of the research studies examined whether skin picking was associated with other variables. Providing supporting information for the research findings of triggers or functions to skin picking, Dykens (2014) found that there was a negative correlation between skin picking and exercise or physical activity. Furthermore, skin picking and sedentary activities such as watching the television were positively correlated. Good quality research has also shown that the higher the score on an anxiety scale the more skin picking shown (Morgan et al., 2010).

Skin picking is associated with other aspects of the phenotype of PWS such as temper outbursts, insistence on sameness and severity of problems with eating (Wigren & Heimann, 2010; Wigren & Hansen, 2005; Holland et al., 2003).

Additionally, skin picking and compulsive behaviours were positively correlated and the age of onset of skin picking and compulsivity also correlated (Didden et al., 2007).

Finally, in one study that used a quality of life measure (Paediatric Quality of Life Inventory- Parent Proxy; Varni et al., 2001) participants with a high skin picking score had a higher score on the health problems subscale of this measure suggesting that people who showed more skin picking had more physical health difficulties (Morgan et al., 2010).

1.4.1.10 Genetic subtype and age differences

Three studies, rated good, found that there were no differences in skin picking between genetic subtypes (Hustyi et al., 2013; Morgan et al., 2010; Didden et al., 2007). However, two studies showed that there was a higher prevalence of skin picking in those with a deletion subtype of PWS compared to a mUPD subtype (Dykens. Cassidy & King, 1999; Cassidy et al., 1997) and two studies showed that those with a deletion subtype pick more sites (Symons, Butler, Sanders, Feurer & Thompson, 1999) and show more problematic skin picking (Hartley et al., 2005). These four studies were not rated as highly according to the quality criteria.

Three studies reported that there were no differences between age and skin picking for younger children, older children, adolescents and adults (Arron et al., 2011; Buono et al., 2005; Dimitropoulos et al., 2001). However, one study found that younger participants showed less skin picking (Akefeldt & Gillberg, 1999) and a second found that adolescents showed the highest rate of skin picking compared to children and adults (Dykens et al., 1992). However, both of these studies had a small sample. Finally, one study noted a higher prevalence of skin picking in adults compared to children but that this was not significant (Boer & Clarke, 1999). This study had the largest number of participants and used a standardised measure. All studies examining age and skin picking were cross sectional so individual differences between the age groups may account for some of the differences observed.

1.4.1.11 Other aspects of skin picking reported

One study reported that the majority of participants who skin pick do so when others are not present and try to hide it (Wigren & Heimann, 2010). Hall et al. (2013) recorded a participant's physiology during skin picking and found that heart rate increased whilst the participant was engaged in skin picking. One study found that the lower a participant's Body Mass Index the more skin picking someone showed (Dykens, 2004). Skin picking has also been shown to have an association with mood changes, "stubbornness" and "argumentativeness" (Holland et al., 2003).

Other types of self-injury in people with PWS have been noted with six studies documenting the prevalence of trichotillomania ranging from 5% to 30% (Buono et al., 2010; Morgan et al., 2010; Wigren & Heimann, 2010; Didden et al., 2007; Symons et al., 1999; Feurer et al., 1998).

Finally, a qualitative self-report study documented that some adults with PWS have negative feelings associated with their skin picking such as guilt (Didden et al., 2008).

1.4.1.12 Interim summary

In summary research has found that skin picking typically starts before the age of six or seven years old with legs, arms, hands and head being the most common sites picked using fingers or fingernails to pick with. The frequency and duration was reported to vary and skin damage reported. The most common functions appeared to be sensory stimulation or escape from high arousal.

1.4.2 Interventions for skin picking in people with PWS

1.4.2.1 Behavioural interventions

Table 1.6 shows a summary of studies researching behavioural interventions for skin picking in people with PWS. *Table 1.7* provides an overview of the quality of the behavioural intervention research based on Downs and Black (1998).

Table 1.6 Summary of behavioural intervention studies for skin picking in people with PWS

Authors	**Participants**						**Design**	**Outcome measures**	**Intervention**	**Key findings**	
	n	*Age*	*Gender*	*Genetic subtype*	*Intellectual disability*	*Comparison group*				*Triggers/functions*	*Intervention outcome*
Wilson, Iwata & Bloom (2012)	1	31	Female	-	-	No comparison group	Single case experimental design – ABA	Functional analysis. Direct observation with high inter-rater reliability. Measurement of wounds	Differential reinforcement of other behaviour (DRO)	Participant's skin picking maintained by automatic reinforcement.	DRO reduced skin picking and decreased wound size
Radstaake et al. (2011)	1	16	Female	mUPD	IQ = 52	No comparison group	Single case experimental design - AB	Functional behavioural assessment (Didden, Duker & Korzilius, 1997). Photographs of wounds rated by researchers.	Differential reinforcement of incompatible behaviour (DRI) and differential reinforcement of alternative behaviour (DRA) and sessions with researchers discussing skin picking	Skin picking maintained by positive reinforcement (self-stimulation and negative reinforcement (escape from emotional arousal).	Photographs rated correctly as beginning or end of intervention. Qualitative feedback - decrease in frequency, severity and number of wounds at follow-up

- Not reported or none conducted

Table 1.7: Quality review of behavioural intervention studies using Downs and Black (1998) criteria

Authors	**Reporting**	**External validity**	**Internal validity – bias**	**Internal validity - confounding**	**Power**	**Overall**
Wilson, Iwata & Bloom (2012)	Good	Poor	Good	Satisfactory	Poor	Satisfactory
Radstaake et al. (2011)	Good	Poor	Good	Satisfactory	Poor	Satisfactory

Two studies examined whether behavioural interventions could reduce skin picking in people with PWS (Wilson, Iwata & Bloom, 2012; Radstaake et al., 2011), see *Table 1.6.* Both were single case experimental designs generating baseline and intervention data and one study used a reversal design (Wilson et al., 2012) that offers more control for confounding variables (Elmes et al., 2006; Perdices & Tate, 2009). Both studies fell within the satisfactory range according to quality criteria (see *Table 1.7*). The function of skin picking was reported to be positive reinforcement through self-stimulation and for one participant negative reinforcement through escape from high arousal. Both studies reported that the behavioural intervention used reduced skin picking. The studies used differential reinforcement of other, alternative and incompatible behaviours as intervention strategies.

1.4.2.2 Pharmacological interventions

Table 1.8 shows a summary of the studies researching pharmacological interventions. None of the pharmacological intervention studies reported what the triggers or functions of skin picking were for participants so this column is not included in the summary table. *Table 1.9* provides an overview of the quality of the pharmacological intervention research based on Downs and Black (1998).

Table 1.8: Summary of pharmacological intervention studies for skin picking in people with PWS

Authors	***Participants***						***Design***	***Outcome measures***	***Intervention***	***Intervention Outcome***
	n	*Age*	*% Male*	*Genetic subtype*	*Intellectual disability*	*Comparison group*				
Miller & Angulo (2013)	35	5-39	34%	67% deletion 31% mUPD	-	No comparison group	Single case series	Number of skin picking wounds and size of wounds. Parental report of frequency and severity.	N-Acetylcysteine	71% stopped skin picking, 29% had less skin picking wounds
Ye, Bawa & Lippmann (2013)	1	16	0%	-	-	No comparison group	Single case	Frequency and severity of skin picking, mood and aggression	Topiramate	Reduction in skin picking, aggression and anxiety. Increase in mood.
Banga & Connor (2012)	1	15	100%	-	IQ = 65	No comparison group	Single case	Frequency of skin picking	Naltrexone	Skin picking reduced
Shapira et al. (2004)	8	19-38 (mean = 29.5)	38%	75% deletion 25% mUPD	-	No comparison group	Experimental group design	Aberrant Behavior Checklist (Aman et al., 1985), Repetitive Behavior Scale, Yale-Brown Obsessive-Compulsive Scale Checklist (Goodman et al., 1989), Clinical Global Impression Scale, Controlled Oral Word Association Test (Lezak, 1996), Semantic Category Naming Test (Lezak, 1996), Vigilance and Delay Task of the Gordon Diagnostic System task (Gordon, 1983). Appetite assessed with free access to food and hunger analogue scale. Behavioural records from residential home.	Topiramate	Appetite did not reduce. No changes in cognition as measured by tests. Behaviour improved and skin picking reduced
Shapira et al. (2002)	3	19-32	33%	100% deletion	-	No comparison group	Single case series	Weight, some photographs of skin picking wounds and number of wounds	Topiramate	Skin picking reduced
Yaryura-Tobias et al. (1998)	4	38-45	50%	-	Average to moderate	No comparison group	Single case series	Clinical observation	Partial or SSRIs - Thioridazine/ Fluxoxetine/ Clomipramine/ Valproate	Skin picking reduced for all participants

- Not reported or none conducted

Authors	***Participants***						***Design***	***Outcome measures***	***Intervention***	***Intervention Outcome***
	n	*Age*	*% Male*	*Genetic subtype*	*Intellectual disability*	*Comparison group*				
Hellings & Warnock (1994)	2	35 and 44	50%	-	-	No comparison group	Singe case series	Clinical observation	Fluoxetine	Skin picking reduced
Benjamin & Buot-Smith (1993)	1	9	100%	-	Mild	No comparison group	Single case	Clinical observation at hospital and school	Fluoxetine and Naltrexone	Skin picking reduced and wounds healed
Selikowitz et al. (1990)	15	5-27 (mean = 14.2)	-	33% deletion 67% not reported	Average to moderate	No comparison group	Double blind placebo controlled trial	Weight, observations of food related behaviour, aggression and skin picking	Fenfluramine	Lost weight, aggression reduced and negative food related behaviours reduced. No change in skin picking

- Not reported or none conducted

Table 1.9: Quality review of pharmacological intervention studies using Downs and Black (1998) criteria

Authors	**Reporting**	**External validity**	**Internal validity – bias**	**Internal validity - confounding**	**Power**	**Overall**
Miller & Angulo (2013)	Satisfactory	Poor	Good	Poor	Poor	Satisfactory
Ye, Bawa & Lippmann (2013)	Poor	Poor	Satisfactory	Poor	Poor	Poor
Banga & Connor (2012)	Satisfactory	Poor	Satisfactory	Poor	Poor	Poor
Shapira et al. (2004)	Satisfactory	Poor	Satisfactory	Poor	Poor	Satisfactory
Shapira et al. (2002)	Satisfactory	Poor	Satisfactory	Poor	Poor	Satisfactory
Yaryura-Tobias et al. (1998)	Satisfactory	Poor	Satisfactory	Poor	Poor	Satisfactory
Hellings & Warnock (1994)	Poor	Poor	Satisfactory	Poor	Poor	Satisfactory
Benjamin & Buot-Smith (1993)	Poor	Poor	Satisfactory	Poor	Poor	Poor
Selikowitz et al. (1990)	Good	Poor	Good	Good	Good	Good

Nine studies reported upon the use of psychopharmacological interventions. The majority were case series using pre-post design, therefore limiting the external validity of the research and statistical power. Furthermore, due to the case study design, the influence of confounding variables was often not controlled for, thereby, limiting the internal validity of the studies. All studies were rated poorly for external validity. One study used a group design but had no comparison group and a small *n* (Shapira et al., 2004). One study was a double blind placebo controlled trial (Seikowitz et al., 1990) which was the only study to fall within the good quality range according to criteria (see *Table 1.8*). Three studies fell within the poor range (Ye, Bawa & Lippmann, 2013; Banga & Connor, 2012; Benjamin & Buot-Smith, 1993) as they had gaps in the reporting of information and threats to both internal and external validity. All studies with the exception of the double blind placebo controlled study (Seikowtiz et al., 1990) scored poorly for power due to the design being case studies or having small samples.

The high quality research study used fenfluramine, an appetite suppressant, and found no reduction in skin picking (Seikowitz et al., 1990). All other studies reported successful interventions. Topiramate, an anticonvulsant medication, was the drug used by most of the studies (Ye et al., 2013; Shapira et al., 2002; Shapira et al., 2002). Another used a different anticonvulsant medication, valproate (Yaryura-Tobias Grunes, Bayles & Neziroglu, 1998). A wide variety of different pharmacological medication has been tried including tricyclic antidepressants, Selective Serotonin Re-uptake Inhibitors (SSRIs) and atypical antipsychotics (Yaryura-Tobias et al., 1998; Hellings & Warnock, 1994; Benjamin & Buot-Smith, 1993).

1.4.3 Comparison between skin picking in the typically developing population and skin picking in people with PWS

1.4.3.1 Phenomenology of skin picking

The final aim of this systematic review was to examine similarities and differences between the phenomenology of skin picking seen in the typically developing population and in the intellectual disability population. The literature reviewed above on the phenomenology of skin picking was compared to a recent systematic review of the phenomenology of skin picking in the typically developing population (Odlaug & Grant, 2012).

Similarities and differences were noted in a number of different aspects of skin picking in each population. For the aspects of skin picking that have research findings described in both populations *Table 1.10* summarises key findings. Odlaug and Grant (2012) documented that the most common age of onset was either during childhood (before 10 years old) or during adolescence. This differs to people with PWS as age of onset is fairly consistently reported to be around six or seven years old or earlier (Wigren & Heimann, 2010; Buono et al., 2010; Didden et al., 2007; Buono et al., 2005). Sites of skin picking between the two populations show some similarities, however Odlaug and Grant (2012) stated that the face was the most common site compared to head, arms, hands and legs for people with PWS. Both groups appear to pick multiple sites and the type of skin picked tends to be imperfections of the skin such as spots and also healthy skin (Hustyi et al., 2013; Odlaug & Grant, 2012; Buono et al., 2010; Morgan et al., 2010).

Table 1.10: A summary of the skin picking research findings in the typically developing population compared to people with PWS

Phenomenology of skin picking	**Typically developing population**	**PWS**
Age of onset	Before 10 years or Adolescence	Age 6 or 7
Sites of skin picking	Face	Head, arms, hands and legs
Number of sites	Multiple	Multiple
Type of skin	Skin imperfections	Spot, insect bites, scabs and healthy skin
Method used	Fingers/fingernails and some object use e.g. tweezers	Fingers/fingernails, some biting and some object use e.g. scissors
Duration	Most commonly hours but varied	Less than a minute to 1 ½ hours
Severity	Bleeding, scars, infections	Mild to severe, bleeding, scars and infections
Triggers/functions	Positive reinforcement (sensory stimulation), Negative reinforcement (escape from emotional arousal), emotion regulation, removing imperfections	Positive reinforcement (sensory stimulation), Negative reinforcement (escape from emotional arousal)
Comorbidities	Trichotillomania and nail biting, mood disorders, OCD and body dysmorphic disorder (BDD)	Trichotillomania, head banging, eye gouging

The method used to pick skin has been reported in both areas of research to be most commonly fingers or fingernails. However, Odlaug and Grant (2012) documented that people in the typically developing population also pick at their skin using objects such as tweezers and Morgan et al. (2010) noted that a small percentage of participants with PWS used biting or implements such as scissors. Duration varies in both groups but in the typically developing population engaging in skin picking for hours per day appears to be most common (Odlaug & Grant, 2012; Morgan et al., 2010). Severity has been reported in both populations according to the damage to the skin that is incurred as a consequence of skin picking and has been reported to be similar across populations (Hall et al., 2013, Hustyi et al., 2013; Odlaug & Grant, 2012; Morgan et al., 2010).

The triggers and functions to skin picking in the two different populations show similarities and differences. Both report that skin picking can be triggered by periods of boredom, which in people with PWS has been described as sensory stimulation, and both can also be triggered by emotional arousal such as stress or anxiety and that engaging in skin picking can in some way reduce emotional arousal (Hall et al., 2013; Hustyi et al., 2013; Odlaug & Grant, 2012; Morgan et al., 2010; Didden et al., 2007). However, although these triggers or functions are the same, the research literature uses different terminology to describe them, for example in the typically developing population the term “reduction of tension” is used but the same function is described using operant terminology in research exploring PWS (“negative reinforcement as escape from emotional arousal”). In *Table 1.10* the same terminology has been used for triggers or functions for both populations to demonstrate more clearly the similarities and differences. One difference in the triggers or functions of skin picking is that in the typically developing research it has been found that skin picking possibly serves a broader emotional regulation function and can also be triggered by a desire to remove imperfections (Odlaug & Grant, 2012).

Finally, both areas of research have explored comorbidities within people who engage in skin picking. Trichotillomania, a body focused repetitive behaviour, is one disorder that has been found to be prevalent in both populations. However, other comorbid disorders or behaviours are different between the groups. In people with PWS it has been found that some people who engage in skin picking also engage in head banging and eye gouging, although the prevalence rates for these behaviours are not as high as has been found for trichotillomania (Wigren & Heimann, 2010; Buono et al., 2010, Morgan et al., 2010, Didden et al., 2007; Symons et al., 1999; Feurer et al., 1998). However, mood disorders, OCD and Body Dysmorphic Disorder (BDD) has also been found to be highly prevalent amongst those in the typically developing population (Odlaug & Grant, 2012).

The review by Odlaug and Grant (2012) of skin picking in the typically developing population did not comment on the frequency of skin picking in this population although this has been examined in people with PWS (Hall et al., 2013; Wigren & Heimann, 2010; Didden et al., 2008; Buono et al., 2005). However, Odlaug and Grant (2012) in their systematic review describe additional research findings that have not been explored in people with PWS, such as emotion and cognition. Research has shown that after engaging in a period of skin picking people most commonly report feeling relieved and some report a feeling of pleasure. Furthermore, it has been found that people who engage in skin picking often have thoughts about wanting to pick and experience an urge to pick. An additional finding related to cognition is that typically developing people who engage in skin picking have deficits in inhibition (Odlaug & Grant, 2012; Odlaug, Chamberlain & Grant, 2010).

1.4.3.2 Interventions for skin picking

To compare interventions for skin picking in people with PWS to those for the typically developing population, a meta-analytic review study of interventions was used for comparison (Gelinas & Gagnon, 2013). Gelinas and Gagnon (2013) applied exclusion criteria to ensure that studies that included participants with an intellectual disability were not reviewed. The review also did not include case study designs or studies where there was insufficient data to complete the meta-analysis.

Gelinas and Gagnon (2013) reviewed six pharmacological intervention studies that met criteria. These studies used different pharmacological agents but all were either an SSRI or an anticonvulsant and all were effective at improving skin picking as measured by severity of picking. However, SSRIs appeared to be more effective than the anticonvulsant medication. This is comparable to the pharmacological treatments tried for people who have PWS (Ye, Bawa & Lippmann, 2013; Shapira et al., 2004, 2002; Yaryura-Tobias et al., 1998, Hellings & Warnock, 1994; Benjamin & Buot-Smith, 1993), however, other pharmacological agents have also been researched in this population but SSRIs and anticonvulsants have been more commonly assessed for their efficacy.

In comparison to interventions researched for people with PWS compared to the typically developing population there are clear differences in the non-pharmacological treatments researched. For people with PWS only two studies have investigated non-pharmacological treatments and have assessed whether behavioural interventions are effective at reducing skin picking (Wilson et al., 2012; Radstaake et al., 2011). However, no similar purely behavioural interventions have been assessed in the typically developing population (Gelinas & Gagnin, 2013). Other psychological interventions have been researched that include a more cognitive component to the intervention. Gelinas and Gagnon (2013) reviewed six studies examining the effectiveness of psychological interventions for reducing skin picking in the typically developing population. Again, different interventions have been tried along with different modes of delivery, particularly for how many sessions are offered, although all showed a successful reduction in skin picking severity.

The different psychological interventions for skin picking in the typically developing population reviewed in this recent meta-analysis include Cognitive-behaviour therapy (CBT), Habit Reversal Therapy and Acceptance and Commitment therapy (ACT; Gelinas & Gagnon, 2013). CBT for skin picking includes psycho-education, identifying and challenging thoughts associated with skin picking and some behavioural strategies such as using distraction when experiencing the urge to pick (Schuck, Keijsers & Rinck, 2011). Habit Reversal Therapy involves assisting an individual to recognise the triggers to their skin picking and noticing when the triggers occur. Once a person has noticed them, a “competing response” is taught where a person engages in something that means they are unable to engage in skin picking at the same time (Teng, Woods & Twohig, 2006). This component of Habit Reversal Therapy is comparable to differential reinforcement of an incompatible behaviour which has been found to be a successful behavioural intervention for reducing skin picking in people with PWS (Radstaake et al., 2011). Finally, ACT focuses more upon the urge to engage in skin picking by encouraging people to see this as less of an aversive event and to become more accepting of experiencing the urge (Twohig, Hayes & Masuda, 2006).

1.5 DISCUSSION

The first aim of this systematic review was to examine the research exploring the phenomenology of skin picking in people with PWS. All but one of these studies utilised informant report methodology, often using questionnaires or structured interviews. The age at which skin picking appears to start was consistently reported across studies to be around six to seven years old, however all studies were retrospective.

In terms of sites of skin picking, studies reported similar sites with the most common being head, arms, hands and legs, however the type of skin picked at these sites varied from healthy to imperfect skin and was only examined by two studies. Two studies also documented how participants picked at their skin which was by using fingers or fingernails. One of these studies was a case study. Frequency and duration were reported to vary widely and only a few studies assessed frequency (Hall et al., 2013; Wigren & Heimann, 2010; Didden et al., 2008; Buoni et al., 2005) and duration was only assessed by Morgan et al. (2010). Furthermore, not all of these studies were clear about what was meant by “frequent” (Buono et al., 2005). This could be explored in more detail in future research.

The severity of skin picking in people with PWS was not well defined although Morgan et al. (2010) found that 41% of their sample met clinical significance for skin picking. Studies reported that participants often caused damage to their skin. Some studies reported that skin picking had resulted in hospitalisation. Both the severity of skin picking and damage caused highlights the physical implications of these behaviours. Only one study briefly mentioned the possible psychological impact of skin picking for people with PWS when a qualitative self-report study documented that participants had negative views about their skin picking (Didden et al., 2008).

Finally, studies documented the function of skin picking as measured by standardised measures. These studies found that skin picking appeared to be positively reinforced by providing sensory stimulation and negatively reinforced by providing escape from emotional arousal such as anxiety (Hall et al., 2013; Hustyi et al., 2013, Didden et al., 2007, 2008). However, it is important to mention that negative reinforcement through escape from high arousal or discomfort is often a default category used in functional analysis when behaviour does not show differentiation across conditions (Patel, Carr, Kim, Robles & Eastridge, 2000; Piazza, Adelinis, Hanley, Goh & Delia, 2000). Other triggers to skin picking have been mentioned such as positive reinforcement through social attention (Didden et al., 2007) and triggers to skin picking have only been examined in four studies, one of which was a single case experimental design (Hall et al., 2013). Future research may benefit from exploring triggers and functions of skin picking by assessing antecedents and consequences to the behaviour. Additionally, it would be interesting to explore setting events which are situations or conditions that make a behaviour more likely (Wahler & Fox, 1981). It is important to be clear about triggers and functions of skin picking to ensure appropriate interventions can be applied.

All of the studies that used informants to report on skin picking behaviour in people with PWS used questionnaires or structured interviews. Research may benefit from using a more open questioning style or a semi structured interview schedule to help to further define skin picking in people with PWS. Furthermore, only one study has been conducted that has directly asked people with PWS to report on their own behaviour and experience of skin picking. This approach to collecting data may be particularly beneficial for assessing aspects of skin picking that are more internal for example thoughts or emotions.

The second aim was to critically review the research documenting the efficacy of interventions for skin picking in people with PWS. There have been few studies researching the effectiveness of interventions for skin picking in this population. Two different behavioural interventions that fell within the satisfactory range of the quality criteria reported that these interventions reduced skin picking. A range of pharmacological interventions using different drugs have also been reported to be effective at reducing skin picking, however, some of these studies were of poor quality.

The final aim was to explore the similarities and differences between skin picking in people with PWS and skin picking in the typically developing population to see whether this research could help inform understanding of skin picking in people with PWS or identify gaps in current knowledge. Similarities included the body sites most commonly picked, that multiple sites were often picked, the type of skin that was picked and that fingers or fingernails were used to pick at the skin. Furthermore, similarities in the damage to the skin caused by skin picking were found and triggers such as boredom and the function of skin picking to reduce emotional arousal were similar. Finally, both showed comorbidity with another body focused repetitive behaviour, trichotillomania. One difference identified between the two populations was the age at which skin picking typically began.

Given the similarities in skin picking between the typically developing population and people with PWS this could have important clinical implications. Due to the recent introduction of skin picking disorder in DSM-5 (American Psychiatric Association, 2013) it would be interesting to see how many people would meet criteria for such a diagnosis. Furthermore, skin picking is conceptualised as a body focused repetitive behaviour and often compared to other body focused repetitive behaviours. According to the similarities identified in this review it may be helpful to compare across similar behaviours in the intellectual disability literature and this may inform and develop interventions for these behaviours in this population.

Research describing skin picking in the typically developing population highlighted areas that could be useful to explore in people with PWS. For example, research in the typically developing population has explored emotions connected to skin picking and how emotions may change before, during or after skin picking (Odlaug & Grant, 2012). However, emotions have not been explored in the same way in people with PWS. This is perhaps due to all but one study asking informants about skin picking rather than asking people with PWS directly. The one study that did use self-reports mentioned emotions connected to skin picking but mainly guilt and shame associated with engaging in it (Didden et al., 2008). Therefore, it would be interesting to explore how a person’s emotions may change over the course of a period of skin picking and this may also lend some support for the finding that some people appear to engage in skin picking as a way of reducing emotional arousal such as stress or anxiety (Didden et al., 2007).

In the typically developing literature there has also been much more research exploring an emotional regulation hypothesis of skin picking (Roberts et al., 2013). This hypothesis states that skin picking functions to reduce negative emotions that people may otherwise have difficulty reducing, therefore it is negatively reinforcing (Roberts et al., 2013). In people who engage in skin picking compared to a group of people who do not, Snorrason, Smári & Ólafsson (2010) found that those who engaged in skin picking had more difficulties with regulating their emotions. It would seem pertinent to explore this possibility within people with PWS given the function identified of skin picking reducing emotional arousal (Didden et al., 2007, 2008). Temper outbursts are also common in people with PWS (Holland et al., 2003; Walz & Benson, 2002) and have been shown to involve high levels of emotion and appear to be difficult for the person to control or manage (Tunnicliffe, Woodcock, Bull, Oliver & Penhallow, 2014) suggesting that perhaps people with PWS also have difficulties regulating their emotions. Therefore, this may contribute to the likelihood of skin picking occurring and to the function of skin picking for this group.

Additional aspects of skin picking explored in the typically developing population but not in people with PWS have been the cognitions associated (Odlaug & Grant, 2012). For example, it has been reported that individuals have thoughts or urges to engage in skin picking before they do so (Odlaug & Grant, 2012) and this has been a focus for some interventions such as CBT and ACT (Gelinas & Gagnon, 2013). This has not been explored in people with PWS but it would be helpful to explore to develop understanding to see whether similar psychological interventions as used in the typically developing population for skin picking could be helpful. Furthermore, research has shown that for some people who skin pick, there appears to be a cognitive deficit in inhibition (Odlaug & Grant, 2012; Odlaug et al., 2010). Inhibition is the ability to stop a response when necessary and is part of a broader range of cognitions called executive functions which refers to a set of cognitions that help to control and manage behaviour (Miyake et al., 2000). It would be interesting to explore whether the same cognitive deficit may underlie skin picking in people with PWS as research has shown some executive functioning deficits in people with PWS (Jauregi et al., 2007; Woodcock, Oliver & Humphreys, 2009a).

This is the first review to systematically examine skin picking in people with an intellectual disability and PWS. It has summarised the findings describing skin picking and interventions and highlighted gaps in the research. Furthermore, it has extended the research to consider whether it is possible to learn about important aspects of skin picking researched within the typically developing population, identifying new avenues for future research and potentially effective interventions for skin picking in people with PWS and possibly more broadly for people with an intellectual disability.

This review has also used already established quality criteria designed for specific research studies in order to evaluate the research (Sale & Brazil, 2004; Downs & Black, 1998). Downs and Black (1998) created criteria for evaluating the quality of a variety of intervention studies both randomised controlled trials and non-randomised controlled trials. These criteria have high reliability, both inter-rater reliability and test re-test reliability. Sale and Brazil (2004) created separate quality criteria to evaluate quantitative and qualitative research, both were inclusive lists, however, there are limitations to these criteria. Sale and Brazil (2004) acknowledged that some items within the criteria may be more important than others and this importance or weighting was not described or included with the scoring. Furthermore, the criteria were listed but not described in detail so it is not clear what level of detail was required in order for a criterion could be judged to be met.

A further limitation was that no inter-rater reliability was established for the quality criteria ratings. This would have been helpful particularly for the studies evaluated using the Sale & Brazil (2004) criteria as these criteria were not as well defined and assessing its reliability would have added to the quality of this systematic review. However, the rater applied the same conditions to each of the criteria and for all studies to ensure consistent ratings and comparability across studies. There were limitations in addressing aim three of the systematic review as there was a reliance on already published systematic and meta-analytical reviews that have their own limitations (Gelinas & Gagnon, 2013; Odlaug & Grant, 2012). Finally, it should be acknowledged that we cannot be certain that all relevant studies were included within the systematic review. The search terms selected for finding relevant studies aimed to be as inclusive as possible by including known derivatives of the key themes such as skin picking.

This review has described the phenomenology of skin picking in a population of people with an intellectual disability, specifically people with PWS where there is a high prevalence of skin picking. Skin picking in people with PWS is well defined but there are some gaps in the knowledge such as the frequency of skin picking and emotions and cognitions associated with skin picking. Both behavioural and pharmacological interventions have been reported to be effective at reducing skin picking in people with PWS. More similarities than differences appear to be present between skin picking in this population and the typically developing population. Therefore, it may be useful to draw on interventions for skin picking in the typically developing population. These interventions have included CBT, Habit Reversal Therapy and ACT. Finally, given the recent inclusion of skin picking disorder as a diagnostic term it will be interesting to see how research and interventions develop for both populations.

Volume One: Chapter 2 - Skin picking in people with Prader-Willi syndrome: Phenomenology and management strategies.

2.1 ABSTRACT

Prader-Willi syndrome (PWS) is a neurodevelopmental disorder with a high prevalence of skin picking. Research has clearly documented some aspects of skin picking such as age of onset and sites of injury. However, other aspects have not been as clearly described, for example, the frequency and duration of skin picking, the type of skin picked and strategies that people have developed to try to reduce skin picking. This study explores the less well defined aspects of skin picking in PWS by using a semi-structured interview with informants (parents or carers) who observe skin picking shown by the person they care for. Nineteen participants (nine male) aged 7-40 years old took part. The results show that participants most commonly picked at least monthly and episodes of skin picking typically lasted between fifteen minutes and an hour. The majority of participants picked skin with an imperfection (n = 18) compared to healthy skin (n= 1) and were reported to be happy and content whilst picking. The majority of informants reported antecedents to be anxiety, boredom and unoccupied hands. The most common management strategy used by parents and carers was distraction and the majority of participants with PWS did not have their own management strategy to try to reduce skin picking. Results are discussed within the context of previous research and implications for intervention considered.

2.2 INTRODUCTION

Prader-Willi syndrome (PWS) is a rare neurodevelopmental disorder with a distinct behavioural phenotype and is associated with a mild to moderate intellectual disability (*Section 1.2*). In *Volume One, Chapter 1* a systematic review explored skin picking in people with PWS. A summary of the findings of this review can be found in *Section 1.5*.

The systematic review highlighted a number key aspects of the phenomenology of skin picking in people with PWS that were not well described (see *Volume One, Chapter 1*). For example, the type of skin typically picked by individuals with PWS comprises both healthy and imperfect skin e.g. skin with a spot or scab (Morgan et al., 2010; Buono et al., 2005). However, this has only been assessed by two studies and the studies did not address whether people were more likely to pick one type of skin than the other and whether people start picking more often at areas that are damaged compared to those that are not. Only two studies considered the way that people with PWS pick at their skin, both reported that fingers or fingernails were used but one of these studies was a case study design (Hall, et al., 2013; Morgan et al., 2010). Furthermore, studies have reported wide ranging frequency and duration of skin picking with no studies clearly defining the frequency categories used, for example "frequent" (Hall et al., 2013; Morgan et al., 2010; Wigren & Heimann, 2010; Didden et al., 2008; Buono et al., 2005). Only one study assessed the duration of skin picking (Morgan et al., 2010). Another aspect of skin picking in people with PWS that is unclear is the function (an operant term referring to the maintenance of a behaviour through positive or negative reinforcement) or antecedents to skin picking. Studies have reported a wide range of functions, however, the majority have found skin picking to function as positively reinforcing by providing sensory stimulation or negatively reinforcing by providing relief from high emotional arousal (Hall et al., 2013; Hustyi et al., 2013; Morgan et al., 2010; Didden et al., 2008; Didden et al., 2007).

Skin picking can have both short term and long term consequences. Physical consequences include damage such as reddening and abrasions, bleeding and infections and long term skin damage in the form of scarring has been reported with implications for physical health (Hall et al., 2013; Hustyi et al., 2013; Morgan et al., 2010). Furthermore, the psychological consequences of engaging in skin picking have been highlighted by one study that interviewed people with PWS and found that participants had negative opinions and emotions about their skin picking behaviours, such as viewing it as “filthy” and feeling angry and guilty about it (Didden et al., 2008). Finally, studies have asserted that skin picking can be triggered by periods of high emotional arousal and that skin picking can function to reduce that anxiety (Hustyi et al., 2013; Didden et al., 2007). Although, this was assessed by questionnaire measure of behavioural function which has limitations of relying on informant report and is often retrospective compared to observational methods (Toogood & Timlin, 1996). Therefore, skin picking in people with PWS has implications for quality of life and appears to be closely linked to psychological wellbeing. Establishing successful interventions for skin picking is clearly important.

Various pharmacological interventions have been evaluated with most reporting improvements (Miller & Angulo, 2013; Ye et al., 2013; Banga & Connor, 2012; Shapira et al., 2004; Benjamin & Buot-Smith, 1993). However, the majority of this research employed case series studies and often potentially confounding variables were not controlled for resulting in poor external and internal reliability. Only one study used a double blind placebo controlled research design (Seikowitz et al., 1990; see *Volume One, Chapter 1*). Research has recently started to explore behavioural interventions for skin picking in people with PWS (Wilson et al., 2012; Radstaake et al., 2011). Both studies are single case reports and reported the interventions to be successful. Finally, no studies have assessed any interventions that parents, carers or people with PWS themselves might use.

When first describing aspects of phenotypic behaviours (a set of behaviours that are associated with a particular population; Dykens, 1995) research often begins at the descriptive and phenomenological level. For example, early studies investigating self-injurious behaviour focused on describing the behaviour before moving towards observational and experimental research and then management and reduction (Hyman, Oliver & Hall, 2002; Oliver, 1995). It is important to have an accurate description of behaviour prior to identifying the most appropriate types of interventions to try to reduce such behaviours.

One way of beginning this descriptive process is by using interviews or questionnaires to gather information. Previous research examining the phenomenology of skin picking in people with PWS has used these techniques. Most of these studies (e.g. Morgan et al., 2010; Didden et al., 2007) used standardised questionnaires such as the Skin Picking Scale (Keuthen et al., 2001). Whilst some of the questionnaires have good reliability and validity, none have been designed specifically for PWS so these may miss key aspects of the phenotype of PWS that may be important to consider. For example, PWS has been associated with a high pain threshold that may interact with skin picking (Cassidy et al., 1997) but no standardised skin picking measures assess pain perception. Other methods adopted to assess the phenomenology of skin picking in people with PWS have included bespoke questionnaires that may overcome some of the limitations of using more standardised measures. However, questionnaires can limit the freedom of the respondent and can be constrained by the design of the questionnaire with more close-ended questions (Oppenheim, 2005). No studies have used a more open methodology with parents or carers of people with PWS. A semi-structured interview is one way to elicit information using some open ended questions and also enables the researcher to question further on answers that are ambiguous or not anticipated (Barriball & White, 1994).

Semi-structured interviews have been used previously to examine other aspects of the behavioural phenotype of PWS (Tunnicliffe et al., 2014; Woodcock, Oliver & Humphreys, 2009b). Both of these studies used operational definitions and Tunnicliffe et al. (2014) used a coding scheme to quantify the data within a framework that defines for each question how to classify participants’ responses (Oppenheim, 2005). Tunnicliffe et al. (2014) demonstrated the utility of the semi-structured interview method for describing and clearly defining temper outbursts. This study advanced the research literature about temper outbursts in PWS using semi-structured interviews to demonstrate clear behavioural and emotional sequences to temper outbursts. Furthermore, the semi-structured interview study conducted by Woodcock et al. (2009b) demonstrated how this methodology could make clear distinctions between similar repetitive behaviours (preference for routine) in two different neurodevelopmental disorders, PWS and Fragile-X syndrome, that more standardised measures may not have been able to differentiate.

The objective of this research was to develop and employ a semi-structured interview to gather further information about the phenomenology of skin picking in people with PWS. The general aim was to describe aspects of skin picking in people with PWS where there has been little previous research or where there are gaps in the previous research identified from a systematic review of the literature (*Volume One, Chapter 1*). The specific aims were to explore the type of skin that people with PWS tend to pick, i.e. healthy or imperfect skin, to describe the method that people with PWS used to skin pick, to describe the frequency and duration of skin picking, to consider the influence of pain and to explore what strategies parents, carers and people with PWS might be using to try to prevent or reduce skin picking.

2.3 METHOD

2.3.1 Participants

Participant inclusion criteria were people with a diagnosis of PWS and who were currently engaging in skin picking. The ethical review committee approved this research and participants who were under sixteen years old could take part by providing their own consent or a parent or carer could consent on their behalf. Participants who were over sixteen years old were required to provide their own informed consent and a parent or carer could not provide consent on their behalf.

Participants were recruited from an international conference for PWS that was based in the UK in July 2013 and from an already established database of participants who had previously taken part in research at the Cerebra Centre for Neurodevelopmental Disorders. Participants were contacted from the database who had taken part in a recent questionnaire study (between July 2011 and February 2012) and had indicated that the person was engaging in skin picking according to responses on the Challenging Behaviour Questionnaire (CBQ; Hyman, Oliver & Hall, 2002) and who had indicated that they would like to be contacted about future research (*n* = 61).

Four participants were recruited from the international conference and fifteen participants from the database. Of the 61 participants available for contact from the database 25 (41%) participants were uncontactable, one (2%) participant was deceased, five (8%) participants were no longer currently engaging in skin picking, three (5%) adult participants were not able to provide their informed consent, three (5%) participants were not interested, one (2%) participant was unable to take part due to a stressful life event and eight (13%) participants did not return a signed consent form.

Participant demographics are shown in *Table 2.1*. No formal measure of level of intellectual disability was included in this research study so for some participants this was unknown. However, for those participants who were on the database and who had taken part in a questionnaire study between July 2011 and February 2012 an indication of their level of ability could be provided by informant report answers on the Wessex Scale (Kushlick, Blunden & Cox, 1973) which assess motor skills, verbal abilities and daily living skills. For motor skills the scale categorises participants as mobile or not mobile and for verbal abilities categorises participants as verbal or non-verbal. According to this scale a participant is considered to be verbal if they can say odd words. For daily living skills participants are given an overall score from three to nine based on their washing, dressing and toileting abilities, a low score indicates poorer daily living skills. Information from The Wessex Scale (Kushlick et al., 1973) was unavailable for five participants. All participants were verbal. Three participants were partly mobile and the remaining participants were fully mobile. Daily living scores ranged from five to nine with a mean score of 7.37.

Table 2.1: Participant demographic information

Participant	Age (years:months)	Gender	Genetic subtype
1	30:0	Female	Unknown
2	26:9	Female	Deletion
3	30:7	Male	Unknown
4	24:6	Male	Deletion
5	7:2	Female	Deletion
6	38:0	Male	Unknown
7	14:4	Female	Deletion
8	15:5	Male	Deletion
9	39:3	Female	Deletion
10	13:8	Male	mUPD
11	24:0	Male	Deletion
12	10:4	Female	mUPD
13	21:5	Female	Deletion
14	40:4	Female	Deletion
15	19:9	Male	Deletion
16	8:11	Female	Deletion
17	15:11	Female	Deletion
18	14:7	Male	mUPD
19	14:5	Female	Deletion

2.3.2 Measures

2.3.2.1 Semi-structured interview schedule

A semi-structured interview schedule was developed with a series of open and fixed choice questions (see *Appendix B*). The design was based on a previous semi-structured interview for assessing temper outbursts in people with PWS (Tunnicliffe et al., 2014). This interview had good inter-rater reliability and showed convergent validity with another measure of temper outbursts, a behaviour diary (Bull et al., 2015). Open questions were designed to allow free response from informants whilst minimising bias from the interviewer. Written prompts were included for these questions if informants were finding questions difficult to answer. Fixed choice questions included those questions asking for a “yes”, “no” or “don’t know” response. Additional fixed choice questions were used for assessing the frequency and duration of skin picking. The fixed choice question and answers were taken from the Challenging Behaviour Interview (CBI; Oliver et al., 2003) which addressed the frequency and duration of challenging behaviour. These items in the CBI (Oliver et al., 2003) had good inter-rater reliability (.69) and test retest reliability (Pearson’s $r = .90$).

Content of the questions was based on previous research and included the frequency and duration of skin picking, the age at which skin picking first began, body sites targeted, the type of skin picked, method used to pick and damage to the skin. Informants were also asked about precursor behaviours to skin picking (behaviours that occur directly before a target behaviour; Najdowski, Wallace, Ellsworth, MacAleese & Cleveland, 2008), emotion during and after skin picking and what happens after skin picking (e.g. whether participant seeks help or talks about it). Antecedents to skin picking were ascertained by asking informants to think about the last time their son, daughter or person they cared for showed skin picking, (similar to Tunnicliffe et al., 2014). Informants were also asked about any intervention strategies that they or the person with PWS use, both preventative and reactive, and how successful those interventions were. Additional questions were constructed based on findings of previous research such as the desire of the person with PWS to stop skin picking, ability of the person to stop skin picking, what a person with PWS might say about their skin picking and if the person shows any signs of pain whilst skin picking.

2.3.3 Procedure

After participants had returned signed consent forms giving their informed consent to take part, informants were contacted by telephone or email to arrange a convenient date and time for the interview. Participants were then contacted by telephone at the arranged time to conduct the interview. Interviews lasted between 15 minutes and 52 seconds and 55 minutes 15 seconds with a mean duration of 25 minutes 35 seconds. All interviews were conducted by the author.

This study had ethical approval from the Science, Technology, Engineering and Mathematics Review Committee at the University of Birmingham (Reference ERN_12-0018AP11). See *Appendix C* for a copy of the approval

2.3.4 Coding

A coding scheme quantified responses, see *Appendix D.* This template minimises researcher bias and ensures that answers are comparable across participants. The coding template was created once all interviews had been conducted which is the process recommended to ensure representativeness and minimise any amendments to the coding scheme that may need to be made and therefore minimises errors in coding (Oppenheim, 2005).

2.3.5 Inter-rater reliability

A second researcher independently coded eight (42%) of the interviews using the coding scheme. The percentage agreement at an item by item level for each overall interview was calculated. Agreement for items ranged from 68.75% to 100.00%. The mean item agreement between researchers was 88.51%.

2.4 RESULTS

2.4.1 Phenomenology of skin picking

2.4.1.1 Age of onset

Informants were asked to recall how old their child or person they care for was when they started to skin pick, *Table 2.2* shows these results. Three informants were not able to answer this question as they had only known the participant as an adult and two informants could not remember the age at which it began but recalled that it had started before the person was 10 years old. The most commonly reported age was between 3-5 years old (n = 7/19) but also between 6-8 years old (n = 6/19). One informant reported skin picking to start at 18 months old and one informant reported it to begin at 13 years old.

Table 2.2: Frequency table of the phenomenology of skin picking, form, frequency, duration and injury (n = 19)

Age of onset	**0-2 years**	**3-5 years**	**6-8 years**	**9-11 years**	**12-14 years**	**Don't know**		
	1	7	6	0	1	4		
Sites of injury*	**Arms**	**Hands**	**Legs**	**Face**	**Torso**	**Site of any skin imperfection**	**Feet**	**Neck**
Most common site	9	9	6	5	3	1	0	0
Other sites	3	4	3	4	3	7	4	1
Type of skin picked*	**Skin imperfection**	**Healthy skin**						
	18	9						
Method used to pick*	**Fingers / fingernails**	**Teeth**	**Objects**					
	19	6	2					
Frequency	**In the next 15 minutes**	**In the next hour**	**By this time tomorrow**	**By this time next week**	**By this time next month**	**Don't know**		
	1	3	3	4	6	2		
Duration	**Less than a minute**	**Less than 5 minutes**	**Less than 15 minutes**	**Less than an hour**	**More than an hour**	**Don't know**		
Longest duration	0	1	4	6	1	7		
Typical duration	0	2	4	6	0	7		
Damage to skin*	**Sore red skin**	**Bleeding**	**Shallow wounds**	**Deep wound**	**Infection**	**Scarring**		
	8	18	8	7	7	7		

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*Participants could provide multiple responses to answer these questions

2.4.1.2 Sites of injury

Informants reported multiple sites of injury for each participant. Informants were asked to report the common sites followed by other sites they were aware of. Sites were grouped according to body area, for example “around fingernails” was grouped under hand. *Table 2.2* shows the grouped sites reported and the frequency of informants reporting each site. *Appendix E* shows the non-grouped body sites reported by informants. The majority of informants reported the legs (n = 9), hand (n = 13), arms (n = 12) and face (n = 9) as the most common sites of injury. Eight informants stated that the child or person they care for would pick anywhere on their body if there was a skin imperfection present.

2.4.1.3 Type of skin

The majority of informants (n = 16/19) reported that the type of skin picked most commonly picked was skin where there was an imperfection compared to otherwise healthy skin. Examples of imperfections reported included sore skin, dry skin, cut/scratch/graze, spot, insect bites, sunburn, eczema, itchy skin and scarring. Eight informants reported that both healthy and imperfect skin were picked. One informant said that anxiety influenced the type of skin picked with healthy skin being picked when feeling particularly anxious. Only one informant reported healthy skin to be the only type of skin picked. See *Table 2.2.*

2.4.1.4 Method used

All participants used their fingers or fingernails to pick their skin with. An additional six informants reported that their child or person they care for used their teeth as well to bite at the skin. Two informants reported that in the past their child or person they cared for used objects to pick with (scissors and a safety pin). See *Table 2.2*.

2.4.1.5 Frequency

Informants were asked to comment upon when skin picking would be likely to happen again based upon its frequency within the last month. *Table 2.2* shows the number of informants reporting each frequency. The majority of informants reported that skin picking occurred at least monthly (*n* = 6/19).

Nine informants reported that skin picking was not regular in frequency but seemed to occur more sporadically, the majority of informants attributed change in frequency and particularly increases in frequency to periods of higher anxiety (*n* = 7/9). The remaining informants were not sure why frequency appeared to change. However, eight informants said that skin picking frequency did not change and was fairly constant over time. Two informants were not sure whether the frequency changed over time.

2.4.1.6 Duration

Informants reported that the duration of each episode of skin picking had typically lasted less than an hour but more than 15 minutes for the majority of participants (*n* = 6/19). When asked about the longest episode in the last month, all but three informants reported that no episode had exceeded the typical duration. One informant was not sure how long the longest duration would have been over the last month as this participant would sometimes pick when alone. Two informants rated skin picking occurring for longer than typical on at least one occasion. For one participant skin picking was reported to last for over an hour because the participant was not interrupted from doing it. This informant reported that they thought that their child or person they care for would continue to pick unless interrupted or stopped by someone else. *Table 2.2* displays the longest duration and typical duration for each participant.

2.4.1.7 Damage

All but one informant reported that skin damage due to skin picking had occurred in the past year (n = 18). Of those who reported damage, all reported multiple types of skin damage and all reported that bleeding had occurred. *Table 2.2* shows the type of damage reported and the number of participants reporting each type.

2.4.1.8 Antecedents

Informants reported a variety of different antecedents, each antecedent reported and the number of informants reporting each antecedent is shown in *Table 2.3*, informants could report multiple antecedents. The most common antecedents reported were anxiety (n = 9), a skin imperfection combined with unoccupied hands or boredom (n = 6), boredom (n = 4) and hands not being occupied (n = 4).

*Table 2.3: Antecedents reported and number of informants reporting each antecedent (n = 19)**

Antecedent	Most common antecedents	Other antecedents	Total
Anxiety	6	3	9
Skin imperfection and hands not occupied/boredom	5	1	6
Boredom	4	1	5
Hands not occupied	4	0	4
Hungry	1	1	2
Tired	1	1	2
Inactive	1	0	1
Being alone	1	0	1
Feeling hot	0	2	2
Being told “no”	0	1	1
Feeling ill	0	1	1
Anger	0	1	1
Skin imperfection	0	1	1
When person wants to	0	1	1
Not sure	2	0	2

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*Participants could answer these questions with multiple responses

2.4.1.9 Precursors

Informants were asked if there were any precursors to skin picking. Informants found this question difficult to answer and some did not appear to differentiate between an antecedent and a precursor behaviour. Therefore, data from informants who reported the same antecedent and precursor were not included (*n* = 9/19). The number of informants reporting each precursor (*n* = 10) is shown in *Table 2.4*. Precursors reported were participant being quiet (*n* = 1/10), participant struggling to concentrate (*n* = 1/10) and rubbing skin or scab (*n* = 2/10).

Table 2.4: Precursors reported and number of informants reporting each precursor (n = 10)

Precursor	Frequency
No precursors	5
Don't know	1
Quiet	1
Rubbing skin/scab	2
Difficulty concentrating	1

2.4.1.10 Emotions associated with skin picking

Informants described the emotion that the person they care for can sometimes show whilst skin picking and after skin picking. Some informants described more than one emotion that could occur, *Table 2.5* shows each emotion reported and the number of informants reporting each emotion during and after skin picking. The most common emotion reported whilst picking and after picking was feeling relaxed/content/happy (n = 19). Anger and guilt were emotions only reported to occur after skin picking, anger seemed to occur if a person was interrupted or reprimanded about picking. One informant identified that the person that they care for would usually feel anxious when they start a new wound but normally happy and content when picking an already established skin picking wound. Additionally, five informants commented that during skin picking the person they care for appear to be "in the zone" or extremely focussed upon what they are doing.

*Table 2.5: Emotion reported whilst skin picking and after picking and number of participants reporting each emotion (n = 19)**

Emotion	Whilst skin picking	After skin picking
Relaxed/content/happy	10	9
Anxious/tense	4	3
Bored	2	0
Sad/upset	3	5
Emotionless	1	0
Angry	0	3
Guilty	0	2

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*Participants could answer these questions with multiple responses

2.4.1.11 Pain

All informants said that their son, daughter or person they care for showed no signs of pain whilst skin picking except for two informants who said that they were not sure as skin picking was done in private.

2.4.1.12 Events subsequent to skin picking

Some informants reported multiple events to occur subsequent to skin picking. The majority of participants try to hide their skin picking (*n* = 11). Some participants request a plaster/bandage or help (*n* = 6) whereas five participants do not talk about skin picking and two participants were reported to deny having done it. Other participants were reported to apologise for doing it (*n* = 4) or to say negative statements about having done it, for example “I was silly”, “I shouldn’t have done it” (*n* = 3). Five participants were reported to engage in conversations about their skin picking wounds and how they are healing. Finally, two participants were reported to often say “I can’t help it” after having picked.

2.4.1.13 Time spent looking at skin

Informants were also asked to comment on the percentage of time participants paid attention to their skin picking. Most participants spent 100% of the time looking at the skin they were picking (*n* = 10/19). *Table 2.6* shows each percentage reported and the number of informants reporting each percentage.

Table 2.6: The percentage of time that each participant was reported to look at the skin whilst picking (n = 19)

Percentage of time spent looking at skin picking	**Frequency**
0%	3
10%	2
20-30%	1
80-90%	1
100%	10
Not sure	2

2.4.1.14 Ability to stop and desire to engage in skin picking

The majority of informants said that their son, daughter or person they care for can stop when asked to (*n* = 15/19). Informants were also asked about participants' desire to skin pick or to not skin pick. Most reported that each time a person engaged in skin picking they wanted to do it (*n* = 10/19), however, two informants reported that they thought the person they care for engaged in skin picking without thinking or being consciously aware. One informant thought that the person could not help doing it and two informants thought that sometimes the person they care for engaged in skin picking even though they did not want to. Four informants were unsure about the desire of the person they care for to skin pick.

2.4.2 Management strategies

2.4.2.1 Preventative

Informants were asked if they used any preventative strategies, preventative strategies reported are shown in *Table 2.7* and the number of informants who reported each intervention. The most common strategy reported was distraction (n = 9), however, others were more practical such as keeping nails short and skin moisturised. Of those who reported using a preventative strategy nobody reported preventative interventions to be successful all of the time. Six informants did not report any preventative strategies.

Table 2.7: Frequency of preventative, reactive and self-management strategies reported (n = 19)

Prevention strategies*	**Distraction**	**Reduce anxiety by talking**	**Keep skin moisturised**	**Keep nails short**	**Teach consequences**	**Draw attention to rubbing of skin**	**Plasters**	**Active schedule**	**No strategy reported**			
	9	2	2	1	1	1	2	1	6			
Reactive strategies*	**Distraction**	**Reprimand/ ask to stop**	**Reward chart for not picking**	**Cover with plasters and distraction**	**Teach consequences**	**Cover with plasters**	**Hold hands**	**Put hand on arm**	**Act horrified**	**Discuss skin picking**	**Draw attenti on to it**	**No strategy**
Main reactive strategy	6	1	3	1	2	0	1	0	0	0	1	4
Additional reactive strategy	2	2	1	0	0	4	1	1	2	2	0	8
Self-management strategy	**Keeping busy**	**Flicking fingers together**	**Self-talk**	**No strategy**	**Not sure**							
	2	1	1	14	1							

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*Participants could answer these questions with multiple responses

2.4.2.2 Reactive

Other strategies reported were more reactive which were used once skin picking had begun. Informants often used multiple strategies (*Table 2.7* shows all reactive interventions reported) but the most common strategy identified as the main or principal strategy used was distraction (n = 6). Other strategies included rewarding for evidence of no picking (n = 4), covering areas with plasters (n = 4) or reprimanding the person or asking them to stop (n = 3). Informants reported reasons why they would use different strategies the most common being whether the person was actively engaging in skin picking or not (n = 4). Other reasons were the severity of skin picking or damage (n = 3), if the principal intervention did not work (n = 1), being in a public place (n = 1), the person's mood (n = 1) and the availability of the informant to implement a strategy (n = 1). Four informants reported that they didn't use reactive strategies either because they did not work (n = 2) or because the person was an adult so it was hard to use reactive strategies (n = 2).

2.4.2.3 Self-management strategies

When asked whether the person with PWS had their own management strategy for trying to stop skin picking, the majority of informants said no (n = 14), and one informant was uncertain. The remaining participants were reported to have their own management strategies which were keeping busy (n = 2), flicking fingers together (n = 1), and self-talk such as "don't pick" (n = 1). Of these four participants who used a strategy only one consistently used the strategy. *See Table 2.7.*

2.5 DISCUSSION

This is the first study to have explored the phenomenology of skin picking in people with PWS using a reliable, semi-structured interview. It is also the first to explore a large number of the phenomenological aspects of skin picking as well as strategies to reduce to skin picking. Attention was paid to the type of skin picked, the method used, frequency and duration and the possible influence of pain on skin picking in people with PWS. Strategies that parents or carers or that people with PWS might use themselves were also examined.

Age of onset was reported to be between three and eight years old which is comparable to previous research in this area (Wigren & Heimann, 2010; Buono et al., 2010; Didden et al., 2007; Buono et al., 2005). However, of interest is one participant who was reported to start skin picking at thirteen years old. This is much older than other participants in this study and older than has been reported in previous research. This may reflect differences within the syndrome, for example, this participant had the mUPD subtype of PWS and skin picking in this genetic subtype of PWS has been reported to be less severe than skin picking seen in people with the deletion subtype of PWS (Hartley et al., 2005; Dykens et al., 1999; Symons et al., 1999). However, in the typically developing population research has documented a bimodal age of onset, with onset occurring either in childhood or adolescence (Odlaug & Grant, 2012) which contrasts with the age of onset typically seen in people with PWS (Hartley et al., 2005; Dykens et al., 1999; Symons et al., 1999). The most common sites of skin picking reported were legs, hand, arms and face and participants appeared to pick multiple sites which supports what has been found in previous research (Hustyi et al., 2013;

Buono et al., 2010; Morgan et al., 2010; Didden et al., 2008; Didden et al., 2007) The method used in this study (semi-structured interview) contrasts with that of previous research, therefore providing additional support.

The type of skin picked found in previous research included both healthy skin and skin with imperfections (Morgan et al., 2010; Buono et al., 2005) but previous research did not comment on what type of skin was more commonly picked. This study found that both healthy and imperfect skin was picked but that imperfect skin was most common. Some participants picked both healthy and imperfect skin. It would be interesting to explore whether there are any variables that influence the type of skin picked and when certain types of skin are picked. For example, one informant reported that the person they care for seemed to pick healthy skin when feeling anxious but imperfect skin when bored as a form of sensory stimulation.

All participants picked their skin using their fingers or fingernails. This provides support for research that had briefly documented the method of skin picking used (Hall et al., 2013; Morgan et al., 2010). The use of objects to pick skin was reported by two informants and one of these linked object use to a time of high anxiety.

Frequency of skin picking varied across participants as has been found previously (Wigren & Heimann, 2010; Buono et al., 2005). However, this study found that skin picking most commonly occurred on a weekly or monthly basis. The duration of skin picking episodes for the majority of participants lasted for more than fifteen minutes but less than an hour. This is in contrast to the one study that explored the duration of skin picking as this study found skin picking episodes to most commonly last for less than five minutes (Morgan et al., 2010). However, Morgan et al., (2010) used a questionnaire with different response options asking respondents to select time windows defined by minutes between one and ninety minutes, whereas this study used time windows in minutes and hours with fewer response categories.

A further aim of the research was to examine the potential influence of pain on skin picking as a high pain threshold has been documented in people with PWS (Cassidy et al., 1997). No participants showed any signs of pain whilst engaging in skin picking and no informant reported that the person they care for reported feeling pain. It is possible that if people with PWS are not receiving feedback in the form of pain when skin is damaged by picking, there is no immediately aversive cost. A recent review of self-injury in people with an intellectual disability highlighted that a high pain threshold may be important to consider when assessing and intervening to reduce self-injurious behaviour (Oliver & Richards, 2015). A cognitive model of pain has shown that pain functions to direct attention towards the painful stimuli and motivates a person to respond by escaping from the painful stimuli (Eccleston & Crombez, 1999). Without a pain signal skin picking may continue for long periods of time. It would be interesting to see whether in the typically developing population skin picking duration is less than for people with PWS. However, there has been no research documenting clearly the most common amount of time that people spend skin picking in the typically developing population. Although, research has shown that the severity of skin picking, as measured by the damage caused to skin, has been reported to be similar for both people with PWS and the typically developing population (Hall et al., 2013, Hustyi et al., 2013; Odlaug & Grant, 2012; Morgan et al., 2010).

A further finding was that the majority of participants tended to look at the skin they were picking. This is similar to the finding reported by Morgan et al. (2010) who found that more severe skin picking was associated with focussed picking which is picking that the person is aware of and paying attention to. The authors of this paper argued that this demonstrated that skin picking was a compulsive behaviour and that it is not done automatically. Research into skin picking in the typically developing population conceptualise it as a compulsive behaviour due to high comorbidity of OCD, the experience of urges to pick and that picking is done with conscious awareness (Van Ameringen et al., 2014; Grant, et al., 2010) and the recent changes to DSM-5 (American Psychological Association, 2013) now incorporates skin picking under a broader category of “obsessive-compulsive and related symptoms”. It is possible that in people with PWS there is an association with more compulsive behaviour but further research would be needed to be able to draw any conclusions about this. One way this could be explored would be to assess whether people experience urges to engage in the behaviour by using self-report methodology and to assess any other thoughts related to picking that people with PWS may have.

When exploring the antecedents to skin picking in this sample of participants with PWS, two main antecedents were identified; feeling anxious and being bored combined with unoccupied hands. This supports the findings of previous studies that have found the function of skin picking for people with PWS as negatively reinforcing by reducing high emotional arousal or positively reinforcing by providing sensory stimulation at times of little activity or boredom (Hall et al., 2013; Hustyi et al., 2013; Didden et al., 2007). This has also been found in the typically developing population (Odlaug & Grant, 2012). Therefore, it seems that for some people skin picking appears to function as a regulatory mechanism to reduce anxiety. It seems possible that for people with PWS there may be a broader emotional regulation difficulty as temper outbursts can be quite common in people with PWS and are associated with a high expression of emotion (Holland et al., 2003; Tunnicliffe et al., 2014). An additional finding was that some informants stated that the antecedent of hands not being occupied or boredom did not always result in skin picking and that it would usually happen only if there was the presence of a skin imperfection such as a spot. This may suggest that skin picking in these cases could be related to grooming behaviour driven by a compulsion to remove the imperfect skin. This hypothesis of skin picking in people with PWS as a deviant grooming behaviour has been suggested by Dykens, Cassidy & King (1999).

This study examined whether there were any precursor behaviours which refers to any behavioural signs that occur prior to a specific behaviour, in this case skin picking. Only four precursors were identified with rubbing skin or scab being shown by two participants. Informants did not appear to separate antecedents and precursors. This may have been due to the capacity of the interview to differentiate between these. The question included a description of a precursor, however, this question appeared before the question about antecedents. It may have been easier for informants to identify precursors once antecedents had been identified.

In summary, this study has found a number of common characteristics of skin picking in people with PWS. Based on the results of this research skin picking appears to most commonly start between three to eight years old. Preventing the onset of skin picking via proactive intervention during this period could be one strategy, particularly considering the high prevalence of skin picking in people with PWS (Wigren & Heimann, 2010; Buono et al., 2005; Wigren & Hansen, 2005; Holland et al., 2003; Boer & Clarke, 1999; Feurer et al., 1998; Cassidy et al., 1997). Furthermore, this research and previous research has shown that the sites of injury tend to be legs, arms, face and hands (Buono et al., 2010; Morgan et al., 2010; Didden et al., 2008; Didden et al., 2007; Buono et al., 2005; Symons et al., 1999), that people pick with their fingers or fingernails and that it more commonly occurs on a weekly or monthly basis with each episode most commonly lasting between fifteen minutes and an hour. Intervening to try to prevent skin picking from developing may consist of monitoring skin in the key injury sites and trying to keep skin as imperfection free as possible (e.g. keeping skin moisturised) during the most common onset period. Times when there are a skin imperfection appear to be times when skin picking is more likely to occur as participants in this study most commonly picked imperfect skin. When imperfections appear other preventative strategies may be helpful such as covering the imperfection with plasters or keeping the person occupied. Further research exploring the possibility of early intervention would be important.

Some informants reported using preventative strategies to try to stop skin picking from occurring. For participants where skin picking appeared to be triggered by anxiety, parents or carers may try to engage in a conversation with that person to find out about what is worrying them and try to help them problem solve and therefore reduce their anxiety. This supports the hypothesis that perhaps for some people with PWS there is an emotion regulation difficulty, particularly for anxiety. If emotion regulation is a difficulty it would be important to try to put in place other more functional regulatory skills. It may be appropriate to explore interventions that help to teach emotion regulation skills such as Dialectical Behaviour Therapy (DBT; Linehan, 1993) or relaxation techniques often used in Cognitive-Behavioural Therapy (CBT; Beck, 1967).

Informants also reported using more reactive strategies for times when somebody had already begun skin picking. The most common strategy reported was using distraction by engaging the person in an activity or more specifically giving them something to do with their hands. This is the first study to ask parents or carers what strategies they had developed and used themselves. The use of distraction corresponds to behavioural techniques or differential reinforcement of incompatible behaviour or alternative behaviour that has been explored in two single case experimental designs (Wilson et al., 2012; Radstaake et al., 2011). However not all parents or carers reported using techniques to reinforce the alternative or incompatible behaviours. Future research could explore the wider utility of such interventions particularly for those episodes of skin picking that appear to be triggered by periods of inactivity or boredom. In the typically developing literature CBT, Habit Reversal Therapy and ACT (Gelinas & Gagnon, 2013) have been reported to be effective at reducing skin picking, see *Section 1.4.3.2.* It is possible that these interventions may be useful for people with PWS particular if the interventions are tailored to the needs of people with an intellectual disability.

Finally, five participants were reported to have their own strategy to try to reduce their skin picking with only one consistently using the strategy. Furthermore, informants reported that the majority of participants appeared to be happy, content or relaxed whilst skin picking and after skin picking. This suggests that there are some people with PWS who engage in skin picking but do not want to and are motivated to try to stop and there are some people who appear to be happy to engage in skin picking and are perhaps not motivated to stop. This raises an interesting ethical question of whether intervening is appropriate if somebody does not want to stop skin picking. It would be important to consider on a case by case basis a person's understanding of their skin picking but also to assess any negative consequences to skin picking, particularly with regard to health and the damage that their skin picking may be causing. Furthermore, if a person does not have motivation to change then any intervention will be susceptible to failure (Prochaska, & DiClemente, 1982). Motivational interviewing for some people may be an important first step as this is a way of exploring a person's motivation to change and can help elicit change (Rollnick & Miller, 1995). Further research may benefit from asking people with PWS directly about their understanding of skin picking and the impact that it has. Only one study has explored this to date finding that participants did hold negative views of their skin picking behaviour (Didden et al., 2008).

Overall, the interview demonstrated good inter-rater reliability with forty two percent of interviews being rated by a second rater and agreement between two raters was above 80% for all items on the interview. Frequency and duration of skin picking was explored using clearly defined terminology. A further strength was the use of a structured coding scheme to ensure the minimisation of researcher bias in quantifying the results and ensuring comparability across participants.

One limitation of this research is that the interview was only able to explore skin picking that was observed by informants. Informants in this interview reported that sometimes people try to hide their skin picking or skin picking is only done when alone, therefore, this method of exploring skin picking is limited to only skin picking observable to informants. This area of research may be further enhanced by self-report interviews. A further limitation of this research is that some of the questions asked informants to recall past events. This retrospective approach can be a potential source of error in measurement as informants recall may be affected by forgetting or biases. It would be interesting to be able to explore skin picking longitudinally in a larger sample of people with PWS to avoid some of these limitations. Another approach would be to ask participants to record skin picking on a daily basis to establish a more natural recording of skin picking without the threat to validity of relying upon informants' recall. Furthermore, the semi-structured interview was only assessed for inter-rater reliability, although it had good inter-rater reliability, its validity was not established. Interviews can also be subject to interviewer bias or influence, however, this was minimised by using a clear interview schedule with some structured prompts and by using a coding scheme in order to analyse the data.

With more participants it may have been possible to examine some differences within the groups, for example research has documented some genetic subtype differences in skin picking where a higher severity of skin picking has sometimes been reported in people with the deletion subtype of PWS (Hartley et al., 2005; Dykens et al., 1999; Symons et al., 1999). Furthermore, it would have been interesting to explore changes with age as some research has documented higher skin picking in adolescence (Dykens et al., 1992; Bull; 2014) and appears to be lowest during adulthood (Akefeldt & Gillberg, 1999; Bull; 2014). During the interviews at least three informants reported changes with age in skin picking, although this was not directly investigated.

This semi-structured interview study researching the phenomenology of skin picking in people with PWS and the interventions that people use has added to the research literature by further describing the phenomenology of skin picking, particularly frequency, duration and type of skin picked. Furthermore, this research has examined the influence of pain on skin picking and explored the types of management strategies that families or people with PWS use to try to manage skin picking. Future research may benefit from exploring potential emotion regulation difficulties in people with PWS but also interventions for skin picking in people with PWS. It may be helpful to draw on the more well developed psychological interventions used in the typically developing population to support this.

Volume One: Chapter 3 – Executive summary

3.1 SYSTEMATIC LITERATURE REVIEW

3.1.1 Introduction

Prader-Willi syndrome (PWS) is a neurodevelopmental disorder caused by the loss of genetic information from a specific area of Chromosome 15. PWS is characterised by distinctive facial features, a mild to moderate learning disability and an insatiable appetite. A large number of people with PWS also engage in skin picking (a self-injurious behaviour). Research has explored skin picking and tried to describe some of the key features. One aim of the review was to examine the research describing skin picking. Research has also examined possible strategies to try to reduce skin picking in people with PWS, these studies were also reviewed.

Research has also shown that some people without a learning disability can also engage in skin picking. There has been no research that has looked at the similarities and differences between skin picking in these two different groups. A further aim of the review was to explore the similarities and differences.

3.1.2 Method

A search of the research literature was conducted to identify all research that had explored skin picking in people with PWS with the exception of any studies that only documented how common skin picking was in this population. Each research study was reviewed according to the quality of the research.

To examine the similarities and differences between skin picking in people with PWS and skin picking in people who do not have a learning disability, the research findings were compared to already published reviews of skin picking in people without a learning disability.

3.1.3 Findings

Research describing skin picking in people with PWS varied in quality but all were either satisfactory or good quality. Research showed that skin picking seemed to start around the ages of six to seven and the areas most commonly picked were head, arms, hands and legs. People used their fingers or fingernails to pick both healthy skin and skin with an imperfection (e.g. a spot or dry skin). Few research studies examined the frequency, duration and damage caused to skin and when it was explored it was poorly defined. Frequency and duration were reported to vary and damage to the skin was reported, although the type of damage caused was not clear. One trigger to skin picking appeared to be times of boredom, so skin picking may provide the person with something to do or stimulation. Other triggers to skin picking identified were times of high emotion, e.g. anxiety or to gain attention from somebody.

Strategies evaluated for reducing skin picking in people with PWS included both medication and behavioural techniques such as trying to encourage different behaviours to skin picking. Studies exploring whether medication reduced skin picking varied in quality with some poor quality studies. A range of different medications were tried with most studies reporting a reduction in skin picking. Studies exploring behavioural techniques were of a satisfactory quality although both only evaluated the outcome of the technique with one person. Both found a reduction in skin picking.

Finally when comparing skin picking between people with PWS and people without a learning disability there were more similarities than differences. Similarities included type of skin picked, how skin was picked, how long skin was picked for, damage caused and triggers to skin picking. Differences were the age at which skin picking first began and areas of skin picked. Strategies to try to reduce skin picking in people without a learning disability included more strategies focusing upon thoughts. We may be able to adapt and develop some of the strategies used in this population for people with PWS, although this will need to be explored in the future.

3.2 EMPIRICAL CHAPTER

3.2.1 Introduction

As mentioned above some studies exploring skin picking in people with PWS have not clearly described some aspects such as frequency, duration and damage to the skin caused. The aim of this paper was to try to find out more about some of the aspects of skin picking in people with PWS and to ask parents or carers about the strategies that they have to try to reduce skin picking.

3.2.2 Method

A telephone interview was conducted with parents or carers of people with PWS who skin pick. Nineteen participants took part recruited via an advert at a conference and by contacting people from a research database. Nine participants were male and the participants were aged between seven and forty years old. The interview asked questions about skin picking that the parent or carer observed.

3.2.3 Findings

Most participants in this study began picking between the ages of three to eight. The findings of this study confirmed that of previous studies as face, arms, hands and legs were the body sites most commonly picked. The results showed that participants most commonly picked at least monthly and episodes of skin picking typically lasted between fifteen minutes and an hour. The majority of participants picked skin with an imperfection compared to healthy skin and were reported to be happy and content whilst picking. The majority of informants reported triggers to be anxiety and boredom. Finally most participants did not appear to feel pain whist picking, this may explain why people may pick for long periods as pain usually is a signal to stop. The most common strategies used by parents and carers was distraction by giving the person something to do, such as playing a computer game.

This research may help to start to think about strategies that may be helpful to reduce skin picking in people with PWS, for example distraction. It would also be interesting to conduct more research to see if strategies could try to stop skin picking from happening by trying to intervene early during childhood. However, this research was limited to exploring skin picking that was observed by parents or carers. Some skin picking may not be observed so future research could ask people with PWS who skin pick to comment on their own picking.

Volume One: Appendices

CONTENTS

APPENDIX A – Quality criteria item by item

Table A1: Item by item scores for phenomenological quantitative studies based on Sale & Brazil (2004)

Authors	Quality criterion																													
	Confounding variables identified	Confounding variables controlled	Statement about comparability of groups	Comparison group treated equally aside from intervention	Informed consent stated	Ethical review	Confidentiality protection stated	Statement of purpose	Objectives clearly stated	Outcome measures defined	Assessment of outcome blinded	Description of setting	Design stated	Recruitment described	Sample randomly selected	Inclusion and exclusion criteria stated	Study population defined	Sampling made clear	Source of controls stated	Selection of controls described	Control/comparison group	Statement about non responders/drop-outs	Missing data addressed	Power calculation	Statistical procedures stated	p values stated	Confidence intervals given	Data gathering procedures	Data collection instruments or source described	Significance acknowledged	Standardisation of observers described
Dykens (2014)	0	0	-	-	1	0	0	1	1	1	0	1	0	0	0	0	1	0	-	-	-	1	1	0	1	0	1	1	1	1	1
Hall et al. (2013)	1	1	-	-	1	1	0	1	1	1	0	1	1	0	0	0	0	0	-	-	-	-	1	0	1	1	1	1	1	1	1
Hustyi et al. (2013)	1	1	-	-	0	0	0	1	1	1	0	1	0	1	1	1	1	1	-	-	-	1	1	0	0	1	0	1	1	1	1
Arron et al. (2011)	1	0	1	1	1	0	0	1	1	1	0	1	0	1	1	0	1	0	1	1	1	1	1	0	1	1	1	1	1	1	1
Wigren & Heimann (2010)	1	0	-	-	0	1	0	1	1	1	0	0	0	1	1	1	1	1	-	-	-	0	1	0	1	-	1	1	0	-	0
Buono et al. (2010)	0	0	0	1	1	0	0	1	1	1	0	1	0	0	0	0	1	0	1	0	0	0	1	0	1	-	1	1	1	-	1
Morgan et al. (2010)	0	0	-	-	1	0	1	1	1	1	1	1	0	1	1	1	1	1	-	-	-	1	1	0	1	1	1	1	1	1	1
Didden, et al. (2007)	1	1	-	-	0	0	0	1	1	1	0	1	0	1	1	1	1	1	-	-	-	1	1	0	1	1	0	1	1	1	1
Hiraiwa et al. (2007)	0	0	1	0	0	0	0	1	1	1	0	1	0	1	1	1	1	1	1	1	1	1	1	0	1	1	1	1	1	1	0
Buono et al. (2005)	0	0	-	-	1	1	0	0	0	0	0	0	0	1	0	1	1	1	-	-	-	1	1	0	1	1	0	1	0	1	0
Hartley et al. (2005)	1	0	-	-	1	1	0	1	1	1	0	1	0	0	0	0	1	0	-	-	-	0	1	0	1	1	0	1	1	1	1

- Not applicable

Authors	**Quality criterion**																													
	Confounding variables identified	Confounding variables controlled	Statement about comparability groups	Comparison group treated equally aside from intervention	Informed consent stated	Ethical review	Confidentiality protection stated	Statement of purpose	Objectives clearly stated	Outcome measures defined	Assessment of outcome blinded	Description of setting	Design stated	Recruitment described	Sample randomly selected	Inclusion and exclusion criteria stated	Study population defined	Sampling made clear	Source of controls stated	Selection of controls described	Control/comparison group	Statement about non responders/drop-outs	Missing data addressed	Power calculation	Statistical procedures stated	p values stated	Confidence intervals given	Data gathering procedures	Data collection instruments or source described	Significance acknowledged	Standardisation of observers described
Wigren & Hansen (2005)	0	0	-	-	0	0	0	1	1	1	0	1	0	1	1	0	1	0	-	-	-	1	1	0	1	1	1	1	1	1	1
Dykens (2004)	1	0	-	-	0	0	0	1	1	1	0	1	0	1	0	0	1	0	-	-	-	0	1	0	1	0	1	1	1	1	1
Holland et al. (2003)	0	0	1	1	0	1	0	1	1	1	0	0	0	0	0	0	1	0	0	0	1	0	1	0	1	0	1	0	1	1	1
Dimitropoulos et al. (2001)	1	0	0	1	0	1	0	1	1	1	0	1	0	1	1	1	1	1	1	1	1	1	1	0	1	0	1	1	1	1	1
Akefeldt & Gillberg (1999)	0	0	1	1	0	1	0	0	1	1	0	0	0	0	0	0	1	0	1	1	1	1	1	0	0	1	1	0	1	1	1
Boer & Clarke (1999)	1	0	-	-	0	0	0	0	1	1	0	1	0	1	1	1	1	1	-	-	-	1	0	0	1	1	1	1	1	1	1
Dykens et al. (1999)	0	0	-	-	0	0	0	1	1	1	0	1	0	1	0	1	1	0	-	-	-	0	1	0	1	1	1	1	1	1	1
Symons et al. (1999)	1	1	0	0	0	0	0	1	1	1	0	1	0	1	1	1	1	0	1	0	1	0	1	0	1	1	0	1	1	1	0
Dykens & Smith (1998)	1	1	1	0	0	0	0	0	1	1	0	0	0	1	0	0	0	0	1	1	1	0	1	0	1	1	1	0	1	1	1
Feurer et al. (1998)	1	0	-	-	0	1	0	1	1	1	0	0	0	0	0	0	1	0	-	-	-	0	1	0	1	1	1	1	1	1	1
Cassidy et al. (1997)	1	0	-	-	0	0	0	1	1	1	0	1	0	1	1	1	1	1	-	-	-	1	1	0	0	1	1	1	1	1	0

- Not applicable

Authors	**Quality criterion**																														
	Confounding variables	Confounding variables controlled	Statement about comparability	Comparison group treated equally aside from intervention	Informed consent stated	Ethical review	Confidentiality protection stated	Statement of purpose	Objectives clearly stated	Outcome measures defined	Assessment of outcome blinded	Description of setting	Design stated	Recruitment described	Sample randomly selected	Inclusion and exclusion criteria stated	Study population defined	Sampling made clear	Source of controls stated	Selection of controls described	Control/comparison group	Statement about non responders/drop-outs	Missing data addressed	Power calculation	Statistical procedures stated	p values stated	Confidence intervals given	Data gathering procedures	Data collection instruments or source described	Significance acknowledged	Standardisation of observers described
Clarke et al. (1996)	1	0	1	1	0	0	0	1	1	1	0	0	0	1	0	0	1	0	1	0	0	0	1	0	1	1	0	0	1	1	1
Dykens et al. (1992)	0	0	-	-	0	0	0	1	1	1	0	0	0	0	0	0	1	0	-	-	-	0	1	0	1	1	0	0	1	1	0

- Not applicable

Table A2: Item by item scores for phenomenological qualitative studies based on Sale & Brazil (2004)

Authors	**Quality criterion**																																	
	Triangulation of sources	Triangulation of methods	Triangulation of investigators	Triangulation of theory/perspective	Peer debriefing	Negative case analysis or searching for disconfirming evidence	Member checks	Use of quotations	Informed consent stated	Ethical review	Confidentiality stated	Consent procedures described	Statement of purpose	Statement of research question	Rationale for qualitative method	Rationale for the tradition within qualitative	Study context described	How setting selected stated	Sampling described	Rationale for sampling	Description of participants	Data gathering procedures described	Audio recording described	Transcription described	Field note procedures described	Data analysis described	Coding described	Data collection to saturation	Statement that reflexive journals/logbooks kept	Raw data described	External audit of process	External audit of data	Bracketing	Researchers assumptions stated
Didden et al. (2008)	0	0	1	0	1	1	0	1	1	0	0	1	1	1	1	1	0	0	0	0	1	1	1	1	0	1	1	1	0	1	1	1	1	1

Table A3: Item by item scores for behavioural intervention studies based on Downs & Black (1998)

Authors	**Quality Criterion**																							
	Aim	Outcomes to be measured	Participants	Interventions	Principal confounders	Main findings	Estimates of the random variability	Adverse events	Participants lost to follow-up	Actual probability values	Representative sample asked	Representative sample participated	Representative of typical treatment	Participants blind	Researchers blind	Any unplanned analysis was made clear	Adjusted analysis for different length of follow-ups	Appropriate statistical tests	Reliable compliance	Valid and reliable outcome measures	Double blind	Adjustment for confounding variables in analysis	Losses of participants to follow-up taken into account	Sufficient power to detect an effect
Wilson et al. (2012)	1	1	0	1	0	1	1	0	1	1	0	0	0	0	0	1	1	1	1	1	0	1	1	0
Radstaa-ke et al. (2011)	1	1	1	1	1	0	0	1	1	0	0	0	1	0	1	1	1	1	1	1	0	1	1	0

Table A4: Item by item scores for pharmacological intervention studies based on Downs & Black (1998)

Authors	**Quality Criterion**																										
	Aim	Outcomes to be measured	Participants	Interventions	Principal confounders	Main findings	Estimates of the random variability	Adverse events	Participants lost to follow-up	Actual probability values	Representative sample asked	Representative sample participated	Representative of typical treatment	Participants blind	Researchers blind	Any unplanned analysis was made clear	Adjusted analysis for different length of follow-ups	Appropriate statistical tests	Reliable compliance	Valid and reliable outcome measures	Comparison group from same population	Comparison group recruited at same time	Participants randomised to groups	Double blind	Adjustment for confounding variables in analysis	Losses of participants to follow-up taken into account	Sufficient power to detect an effect
Miller & Angulo (2013)	1	0	0	0	0	0	1	1	1	1	0	0	0	0	0	1	1	1	1	1	-	-	-	0	0	1	-
Ye et al. (2013)	0	0	0	1	0	0	0	0	1	0	0	0	0	0	0	1	1	0	1	0	-	-	-	0	0	1	-
Banga & Connor (2012)	1	0	0	1	0	0	0	1	1	0	0	0	0	0	0	1	1	0	0	0	-	-	-	0	0	1	-
Shapira et al. (2004)	1	0	0	1	1	1	1	1	1	0	1	0	0	0	0	1	1	1	1	0	-	-	-	0	0	1	-
Shapira et al. (2002)	0	0	1	1	0	1	1	1	1	0	0	0	0	0	0	1	1	1	1	0	-	-	-	0	0	1	-
Yaryura-Tobias et al. (1998)	0	0	0	1	1	0	0	1	1	0	0	0	1	0	0	1	1	1	1	0	-	-	-	0	0	1	-
Hellings & Warnock (1994)	0	0	0	1	0	0	0	1	1	0	0	0	1	0	0	1	1	1	1	0	-	-	-	0	0	1	-
Benjamin & Buot-Smith (1993)	0	0	0	1	0	0	0	1	1	0	0	0	0	0	0	1	1	1	1	0	-	-	-	0	0	1	-
Seikowitz et al. (1990)	0	1	0	1	1	1	1	0	1	1	0	0	1	1	1	1	1	1	1	1	1	1	1	1	0	0	1

- Not applicable

APPENDIX B – Semi-structured interview schedule based on Tunnicliffe et al. (2014)

Skin Picking Interview

The aim of this interview is for us to get a better understanding of X's behaviour. In particular, we are interested in skin picking and how often it happens. We are also interested in how long it can last for and when it is more likely to happen. I'm going to ask you a series of questions that should take no longer than 30 minutes. Do you have any questions before we begin?

Name: ____________________ Gender: Male ☐ Female ☐

Age: _____ Genetic subtype: _____________________

Name of respondent: ___________ Date of interview: ___________

1) **How old was X when you first noticed skin picking?**
……………………………………………………………………………………………
……………………………………………………………………………………………

2) **Where on their body is X most likely to skin pick?**
……………………………………………………………………………………………
……………………………………………………………………………………………
……………………………………………………………………………………………
……………………………………………………………………………………………
……………………………………………………………………………………………
……………………………………………………………………………………………
Are there any other areas where X has skin picked?
……………………………………………………………………………………………
……………………………………………………………………………………………

3) **What type of skin does X skin pick? (i.e. healthy, spot, scab, rash)**
……………………………………………………………………………………………
……………………………………………………………………………………………
……………………………………………………………………………………………
What type of skin was it before the picking began?
……………………………………………………………………………………………
……………………………………………………………………………………………
……………………………………………………………………………………………
What type of skin does X now pick?
……………………………………………………………………………………………
……………………………………………………………………………………………
……………………………………………………………………………………………

4) What is the method that X uses to skin pick? (i.e with fingernails, using an object)

....

....

....

5) In the last year, has X damaged their skin when skin picking?

....

....

If yes....what damage? i.e. sore red skin, bleeding, deep wounds, scarring, infection, disfigurement

....

....

....

....

....

6) Think about how often skin picking has happened in the last month. If there was no change and you watched this person now, then would you *<u>definitely</u>* see more skin picking :

In the next 15 minutes	In the next hour	By this time tomorrow	By this time next week	By this time next month

7) In the last month, for how long did the *<u>longest</u>* period of skin picking last for?

Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

8) In the last month, how long have the periods of skin picking *<u>typically</u>* lasted on average?

Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

9) Thinking about the longest period of skin picking in the last month that continued for over an hour- how long did it last?

....

....

10) What keeps skin picking going for long periods of time (i.e. for more than one hour)?

....

....

....
....

11) **Over a period of around 6 months, are there times when X seems to skin pick a lot followed by times when X does not skin pick at all?**
....
....
....
....
Prompt = Are there any patterns to X’s skin-picking that you have noticed?
....
....
....

12) **Are there any indicators before X starts to skin pick?**
Prompt = any indicators in X’s <u>behaviour</u>?
....
....
....
Do these always happen?
....
....
....

13) **When you see these behaviours is there anything that you could do to prevent skin picking from occurring?**
....
....
....
....
How likely is it to be successful? (out of 10)
....
....

14) **During a period of skin picking, how would you describe X’s emotion?**
Prompt= how do you think that X feels when X is skin picking?
....
....
....
....

15) **What does X do after skin picking?**
Prompt= Do they do anything? Say anything?

..
..
..
..

16) **How would you describe X's emotion after skin picking?**
Prompt= how do you think X is feeling?
..
..
..
..

17) **Are there any strategies that you have that can help stop X skin picking?**
..
..
...

18) **Roughly how many times would you respond in this way?**

Always ☐ More often than not ☐ Sometimes ☐

Occasionally ☐ Rarely ☐

19) **In what other ways might you respond? When would you respond in these ways?**
Prompt: e.g. are there any differences at home compared to in public?

1...
2...
3..
4..
5..

20) **What are the reasons that you might respond in different ways?**
..
..
...

21) **Are there any strategies that X has to help them to stop skin picking?**
..
..
...

22) **Roughly how many times would do this?**

Always ☐ More often than not ☐ Sometimes ☐

Occasionally ☐ Rarely ☐

23) If not already mentioned......
Have you tried covering the area of skin that they are picking?
...
...
...
...
Was it successful?
..
...
...
When the area of skin was covered, did X start to pick anywhere else?
...
...
...

24) If you ask X to stop do they stop?
...
...
Do you think that they are able to stop?
...
...

25) Thinking about the last period of skin picking that X showed, what seemed to trigger it?
...
...
..
..

26) Would you say that this is the most common trigger? If not, what is?
...
...
..

27) Out of 10, what proportion of all skin picking that X shows seem to be caused by the trigger that you have identified?
...
...
..

28) Does the trigger that you mentioned always result in skin picking?

Yes ☐ **No** ☐

***If no,* out of 10, how often does the trigger that you mentioned result in a period of skin picking?**

...

What happens on the occasions that it does not trigger skin picking? What is different about these times?
Leave free response initially

..
..
..
..
..

Prompts:
-Is the trigger definitely the same?
-Are there different people present?
-Is X's mood different in someway?
-Does X do something that means that they are not thinking about the trigger or do not notice that it occurs?
-Do you do anything to prevent the skin picking from occurring?

29) We have established that …..is a common trigger to X's skin picking. Are there other triggers?

Yes ☐ **No** ☐

If yes, list below in order of most frequent to trigger skin picking.

1)…………………………………
2)…………………………………
3)…………………………………
4)..

30) What percentage of the time does X look at what they are picking?
..
..
..
..

31) When X is skin-picking do they show any signs of pain?
..
..
..
..

32) **Are there times when X skin picks even when you think that they don't want to do it?**

..
..
..
..

33) **What does X say about their skin picking?**

..
..
..
..

34) **Does X try to hide their skin-picking?**

...
..
..

35) **Is there anything else about skin picking that you would like to mention that has not been asked about?**

....
....
....
....

APPENDIX C – Ethical approval

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APPENDIX D – Coding template

Number	Item details	Coding instructions
1	Age of onset	Report age stated or “don’t know”
2	Body sites most common	Report body sites identified ensuring most common identified. If unable to identify state “don’t know”
3	Any other body sites	Report all body sites mentioned
4	Type of skin picked	Report all types identified or state “don’t know”
5	Type of skin before	If different to item 5
6	Type of skin now	Report type or state “don’t know”
7	Method	Report they method that participants use or state “don’t know”
8	Damage in last year	Report yes, no or don’t know
9	Type of damage	Record type of damage caused
10	Frequency in last month	Taken from CBI (Oliver et al., 2003), fixed choice
11	Longest duration in last month	Taken from CBI (Oliver et al., 2003), fixed choice
12	Typical duration in last month	Taken from CBI (Oliver et al., 2003), fixed choice
13	If longest duration over an hour, how long	Report duration if applicable or state “don’t know” if unable to identify
14	If more than an hour what keeps it going	Report reason given if applicable or state “don’t know” if unable to identify
15	Any pattern over 6 months	State yes, no or don’t know. If yes documents reason for pattern
16	Precursors	State yes, no or don’t know
17	Do precursors always happen	State yes, no or don’t know. If no and frequency given state it or if none given report it
18	Prevention strategies	State yes or no. If yes document intervention
19	How successful are prevention strategies	Document success out of 10 or state if question not applicable
20	Emotion whilst picking	Report all emotions identified or report “don’t know”
21	What happens after	Report any behaviours, speech, emotions reported
22	Emotion after picking	Report all emotions identified or report “don’t know”
23	Main intervention to stop picking	Report principal intervention or more than one if informant states so
24	How often use strategy	Fixed choice answer as on interview schedule
25	Other interventions	Report all other interventions used or state if there are no additional interventions
26	Reasons use different interventions	Report reasons for using different interventions or state “don’t know” of informant unable to identify
27	Participant own strategies	
28	How often use own strategies	Fixed choice answer as on interview schedule
29	Tried covering area	State yes or no

Number	Item details	Coding instructions
30	Was covering area successful	State yes or no and any reasons given if applicable
31	When covered did person pick elsewhere	State yes or no if applicable
32	If ask to stop do they	State yes, no or don't know
33	Are they able to stop	State yes, no or don't know
34	Trigger to last episode	Listen to the description of the last episode and document the antecedent or state "don't know"
35	Most common trigger	Document the most common trigger
36	Proportion of all skin picking triggered by most common trigger	Document number out of 10 or state unable to answer
37	Trigger always cause skin picking (if no how often)	State yes, no or don't know. If no state frequency with number out of 10
38	What happens when trigger doesn't result in skin picking	Identify any reasons given or state if informant is unable to provide any
39	Other triggers	Document all other antecedents or state if all have already been reported in above items
40	Percentage of time look at picking	Report percentage given or state "don't know"
41	Any sign of pain	State yes, no or don't know
42	Does person pick when they don't want to	State yes, no or don't know
43	Say anything	State yes or no If yes state what participant says or topic spoken about
44	Try to hide it	State yes or no
45	Anything else	Add additional information that is not an answer to any previous item

APPENDIX E – All body sites reported by informants

Table A5: Frequency of body sites reported by informants

Site	Most common site	Other sites
Legs	6	3
Hand	6	4
Arms	4	2
Wrist	5	1
Face	4	3
Around fingernails	3	0
Lip	1	0
Breasts	1	0
Hips	1	0
Stomach	1	2
Site of any skin imperfection	1	7
Feet	0	4
Shoulders	0	1
Ears	0	1
Neck	0	1

APPENDIX F – Author guidelines from *Clinical Psychology Review*

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APPENDIX G – Author guidelines from *Journal of Intellectual Disability Research*

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