Predictors of Resilience in Adolescents with a Cleft Lip and/or Palate

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Introductory Chapter: Thesis Overview

It is widely recognised that the presence of a physical health condition and its treatment can compromise the social and emotional wellbeing of those affected (Pinquart & Teubert, 2012). Alongside peer group affiliation, development of body image is reported to be one of the key tasks of childhood and adolescence (Erikson, 1959). Consequently, when a health condition or its treatment affects appearance, the challenges faced at these life stages may be augmented.

Despite these risks, many children and adolescents are reported to have successfully adapted or 'bounced back' from the challenges that have arisen from their health condition and its treatment; this concept is often referred to as resilience (Windle, 2011). In recent years, increased interest in resilience and its application to health policy and promotion has been highlighted (Cal, Sá, Glustak, & Santiago, 2015; Department of Health, 2013).

To the author's knowledge, however, there are currently are no comprehensive reviews examining resilience in children and adolescents with medical conditions associated with visible difference. Consequently, paper one of this thesis aims to address this gap in the evidence base by presenting a systematic review investigating the demographic, illness-related, and psychosocial factors associated with resilience in children and adolescents with medical conditions associated with acquired and congenital visible difference.

Research examining positive outcomes in individuals affected by a cleft lip and/or palate (CL/P), a congenital condition in which appearance may be altered, is sparse (Egan, Harcourt, Rumsey, Appearance Research Collaboration, & McBain, 2011). In 2013, Rumsey and Stock called for increased research efforts to be dedicated to the investigation of factors that facilitate resilience in children and young people with a CL/P. Consequently, the second paper of this thesis aims to explore factors associated with resilience in adolescents with this condition, to inform potential intervention and future service development.

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Resilience in Children and Adolescents with Medical Conditions Associated with Acquired and Congenital Visible Difference: A Systematic Review[1]

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[1]To be submitted to the Journal of Paediatric Psychology (30 page limit for review articles; Appendix A)

Abstract

Objective: To explore demographic, illness-related, and psychosocial factors associated with resilience in children and adolescents with medical conditions associated with acquired and congenital visible difference. *Data sources:* Searches of PsycINFO, Medline, and CINAHL Plus were conducted (inception to December 2014) for studies that contained key search terms. *Study eligibility criteria:* (1) full text written or available in English; (2) the mean age of participants was 19 years old or below; and (3) participants were required to have a medical condition associated with visible difference due to a congenital condition, illness, or acquired from an injury or as a result of medical intervention. *Study appraisal and synthesis methods:* The Quality Assessment Tool for Studies with Diverse Designs was used to appraise the 12 included studies. Narrative synthesis was used to summarise the main study findings. *Results:* No evidence for the relationship between resilience and demographic variables was documented. Inconsistent findings were highlighted regarding the relationship between resilience and illness-related variables and coping strategy use. The evidence for the relationship between resilience and family and peer support was more conclusive. Resilience was associated with increased quality of life and fewer internalising and externalising difficulties. Lack of methodological rigour and the heterogeneity of samples may obscure research findings and limit the interpretations that can be drawn. *Conclusion and implications:* Fostering positive familial and peer relationships may be useful steps for clinicians working with children and adolescents with medical conditions associated with acquired visible difference. Recommendations for future research are made.

Keywords: resilience, visible difference, chronic illness, systematic review, adolescents, children

With recent research trends shifting away from models of deficits and psychopathology, interest in 'positive psychology' including resilience has burgeoned (Hjemdal, Aune, Reinfjell, Stiles, & Friborg, 2007). A lack of consensus in defining resilience is acknowledged in the literature (Johnston et al., 2015). However, following a review of 271 peer reviewed journal articles by the Resilience and Healthy Ageing Network, resilience has been defined as a dynamic process of "effectively negotiating, adapting to, or managing significant sources of stress or trauma. Assets and resources within the individual, their life and environment facilitate this capacity for adaptation and 'bouncing back' in the face of adversity" (Windle, 2011, p. 152).

Initial resilience research focussed on children facing adversity in the form of maltreatment (Cicchetti & Rogosch, 1997), parental psychological distress (Garmezy, 1991a), and social disadvantage (Garmezy, 1991b). Currently, research in resilience in those with and affected by chronic medical conditions (i.e. families and health care professionals) is increasing, and its application to health policy and practice emerging (Friedli, 2009).

Children and adolescents with medical conditions are reported to be at risk of significant psychosocial difficulties due to the multiple challenges that their condition and its management may bring to physical, social, and academic functioning (Pinquart & Teubert, 2012). Whilst childhood and adolescence are considered to be periods of considerable growth and opportunity, it can also be a particularly challenging time, irrespective of the presence or absence of a chronic illness. Alongside many other lifecycle transitions, increased attention may be focused on appearance and body image (Rumsey & Harcourt, 2007; Wu, Sheen, Shu, Chang, & Hsiao, 2013). Consequently, when a medical condition or its treatment impacts upon physical appearance, the challenges confronted may be magnified. Visible difference can be classified as congenital (from birth) or acquired. Congenital conditions that may affect physical appearance include: craniofacial conditions, for example cleft lip and/or palate,

Treacher Collins, Apert, and Cruzon syndromes; birth marks; and neurofibromatosis. Visible difference may also be acquired as a result of a medical condition (e.g., vitiligo, eczema), acute illness (e.g., meningococcal septicaemia), injury (e.g. burns), or its treatment/management (Rumsey & Harcourt, 2007). For example, research highlights that all cancer treatments (including surgery, chemotherapy, and radiotherapy) are associated with both temporary and permanent changes in physical appearance (Wallace, Harcourt, Rumsey, & Foot, 2007). Treatment effects include scarring, amputation, alopecia, weight changes, and disrupted growth, development, and puberty (Eiser, 1998).

Systematic reviews examining factors associated with resilience in adults with chronic medical conditions have documented that this construct is related to a number of psychosocial factors including social support, coping strategy use, and quality of life (Cal, Sá, Glustak, & Santiago, 2015; Stewart & Yuen, 2011). However, as resilience involves the dynamic interplay between temporal and contextual factors, the variables associated with resilience in children and adolescents with visible difference may differ to those deemed important in adults with health conditions (Stewart & Yuen, 2011). Consequently, the aims of this review were to synthesise the existing evidence base regarding the demographic, illness-related, and psychosocial factors associated with resilience in children and young people with medical conditions associated with acquired and congenital visible difference, to subsequently inform clinical practice and to make recommendations for future research.

Method

Search Strategy

Searches of the electronic publication databases PsycINFO, Medline, and CINAHL Plus were conducted for peer reviewed journal articles and unpublished dissertations containing key search terms (Figure 1) in their title, abstract, or keywords. Databases were searched from their inception until 29th December 2014.

Resilienc* AND "congenital condition" OR "congenital anomaly" OR "congenital disorder" OR "visibl* differen*" OR "cleft lip" OR "cleft palate" OR "hare lip" OR harelip OR palatoschisis OR cheiloschisis OR "orofacial cleft" OR "facial cleft" OR "facial difference" OR "facial abnormality" OR "facial anomaly" OR "facial palsy" OR "oral cleft" OR craniofacial OR "cranial anomaly" OR burn* OR scar* OR appearance OR neurofibromatosis OR "skin cancer" OR "head and neck cancer" OR oncology OR amputation OR "skin condition" OR "skin disease" OR dermat* OR birthmark OR "port-wine stain" OR "infantile hemangiomas" OR "congenital melanocytic n*" OR vitiligo OR eczema OR psoriasis OR ichthyosis OR "physical appearance difference" OR "body image" OR injury OR accident OR illness OR trauma OR "long term condition" OR health OR disfigurement OR "Treacher Collins syndrome" OR micrognathia OR macrognathia OR chemotherapy OR radiotherapy OR surgery OR "medical intervention" OR alopecia AND p?diatric* OR child* OR infant OR "young people" OR adolescen* OR teen*

Figure 1. Key Search Terms

The search was supplemented by hand searching the reference lists of studies that met

the inclusion criteria. Alerts were set up on the aforementioned databases to ensure that any

new research published after the initial search was captured and included in the review.

Author searches were conducted to ensure saturation. A representative of the Centre for

Appearance Research (a centre specialising in psychological research in appearance, visible

difference, and body image) was contacted to enquire if they were aware of any relevant

studies that were due for publication within the next three months.

Selection Criteria

Eligible studies were required to meet the following criteria: (1) full text written or

available in English; (2) the mean age of participants was 19 years old (inclusive) or below in

accordance with the World Health Organisation's statement on adolescent development

(World Health Organisation, 2014); and (3) participants were required to have a medical

condition associated with visible difference due to a congenital condition, illness, or acquired

from injury or medical intervention. Studies were excluded from the review if authors

purported they were investigating resilience but had not implemented a resilience-specific

measure. Papers solely employing qualitative methodology were excluded on the basis that

the strength and nature of relationships between resilience and key variables could not be

investigated. Intervention studies and studies only examining the resilience of family members, carers, or professionals involved in the care of children and adolescents with medical conditions associated with visible difference were not included in the review.

An initial screen of all the study titles and abstracts generated from the electronic and hand searches was performed to determine whether they met the inclusion criteria. The full text version of the potentially eligible studies were obtained and scrutinised to ascertain if they fulfilled the study criteria. Two authors were contacted via email for additional information to clarify if their studies met the inclusion criteria; one author responded.

Quality Assessment

The quality of the studies that fulfilled the inclusion criteria of the review were appraised using the Quality Assessment Tool for Studies with Diverse Designs (QATSDD; Sirriyeh, Lawton, Gardner, & Armitage, 2012). The QATSDD was selected as it can be applied to studies employing a range of methodological designs. Sirriyeh et al. (2012) report the QATSDD possesses substantial inter-rater reliability (κ= 71.5%) and good test-retest reliability following a 6 week delay (scores ranging from κ= 69.8% to κ= 100%). Scores for each of the quality assessment criteria are rated from zero (not at all) to three (complete) for. Fourteen of the criteria apply to studies employing quantitative methodology, whilst 16 apply for mixed methods studies. Quality assessment criteria (Appendix B) included items such as consideration of: the sample size and whether participants were representative of the target group identified. Quality assessment was independently conducted by two reviewers to optimise reliability. Discrepancies in scoring were discussed and a consensus achieved.

Data Extraction

Purposely developed data extraction tables were used to record the main study and participant characteristics. Data extraction was independently conducted by two reviewers. Any discrepancies in the information collated were discussed and a consensus achieved.

Data Analysis

Due to the limited number of studies included in the review, the heterogeneity in the clinical samples, and the disparity in the outcome measures administered, it was not deemed appropriate to conduct a meta-analysis. A narrative synthesis of the characteristics of the included studies is therefore provided.

Results

Search Strategy Results

Search strategy results are illustrated in Figure 2 using the Preferred Reporting Items for Systematic Reviews (Moher, Liberati, Tetzlaff, Altman, and the PRISMA group, 2009).

Identification | Screening | Eligibility | Included

Records identified through database searching
(n = 958):
- PsycINFO (395)
- Medline (391)
- Cinahl Plus (172)

Additional records identified through other sources e.g. hand-searches and citation alerts
(n = 9)

Records after duplicates removed (n = 607)

Titles and abstracts screened (n = 607)

Records excluded
(n = 585)

Full-text articles assessed for eligibility (n = 22)

Full-text articles excluded, with reasons (n = 10):

-Did not use a resilience-specific measure (7)

- Inappropriate mean age (2)

- Unclear if parent or child resilience was measured (1)

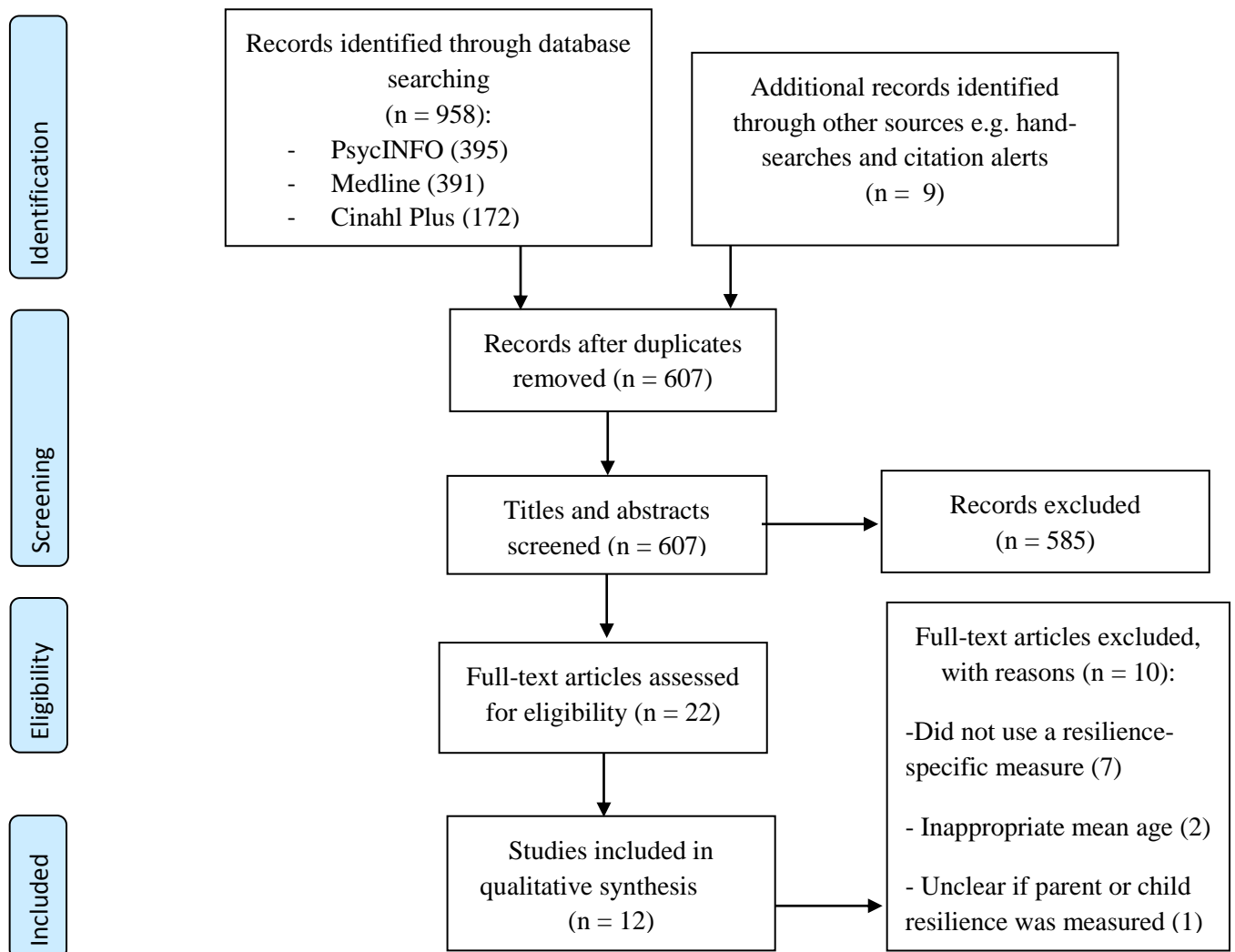Studies included in qualitative synthesis
(n = 12)

Figure 2. Flow Diagram of Search and Review Process

Participant Characteristics

The 12 identified studies (including one unpublished dissertation) recruited a total of

n = 1173 participants whose ages ranged from three to 26 years. All studies included both

male and female participants; 55% of participants were male. Medical conditions of

participants included cancer (k = 9), atopic dermatitis (k = 2), and burns (k = 1). Mean time

since diagnosis of medical conditions/duration of illness ranged from 3.76 (SD 4.60) to 6.75

years (SD 3.93); five studies failed to report this information. Characteristics of the

participants in the studies included in the review are summarised in Table 1.

Table 1

Characteristics of Participants in Included Studies

No.	Author(s)	Medical condition	Age range (years)	Mean age (SD) in years	Male: female ratio	Mean time since diagnosis (SD) in years
1	Chen, Chen, & Wong (2014)	Cancer	13-18	15.29 (1.61)	96:84	Not reported
2	Chou & Hunter (2009)	Cancer	18-21	19.85 (1.10)	61:37	6.38 (1.21)
3	Haase, Kinter, Monahan, & Ross (2014)	Cancer	10-26	15.83 (2.70)	111:90*	3.76 (4.60)
4	Harper et al. (2014)	Cancer	3-12	6.60 (3.24)	62:41	Not reported
5	Harper, Penner, Peterson, Albrecht, & Taub (2012)	Cancer	3-12	6.38 (3.14)	22:19	Not reported
6	Im & Kim (2012)	Atopic dermatitis	7-15	9.51 (2.43)	48:54	6.75 (3.93)
7	Kim & Im (2014)	Atopic dermatitis	7-15	9.51 (2.43)	48:54	6.75 (3.93)
8	Kim & Yoo (2010a)	Cancer	10-15	13.11 (2.19)	37:37	4.22 (3.83)
9	Powers (2011)	Burns	9-18	13.25 (2.75)	13:7	5.75 (4.42)
10	Smorti (2012)	Cancer	11-20	15.56	47:33	Not reported
11	Wu, Sheen, Shu, Chang, & Hsiao (2013)	Cancer	11-19	14.70 (2.27)	73:58	5.05 (4.35)
12	Wu et al. (in press)	Cancer	13-20	16.35 (1.98)	25:15	Not reported

*NB: one value missing

Description of Included Studies

Characteristics of the 12 studies included in the qualitative synthesis are documented in Table 2. Nine studies were cross-sectional in nature (one study was an arm of a longitudinal study); two utilised case-control methodologies; while the remaining study employed a mixed-methods design. The studies were conducted in Asia (k = 6), North America (k=5), and Europe (k=1). In 11 of the 12 included studies, the clinical population were recruited directly via medical centres, hospital clinics or wards. In the remaining study (study nine), participants were initially recruited via a burns camp. Normative/control data for the two case-control studies (studies one and 10) were obtained from school samples. A range of psychometric tools were administered to measure resilience across studies; seven different resilience-specific measures were used in total. The most commonly used resilience measures were the Resilience Questionnaire (Kim & Yoo, 2010b) and the Haase Adolescent Resilience in Illness Scale (2004) which were each administered in three studies. Studies aims were disparate.

Table 2

Characteristics of Included Studies

	Author(s)	Country	N	Method	Resilience measure	Aim(s)	Main findings
1	Chen, Chen, & Wong (2014)	Taiwan	180	Quantitative; case-control	Haase Adolescent Resilience in Illness Scale (Haase, 2004)	To investigate the influence of health problems on the resilience of adolescent survivors of brain tumours compared to healthy controls.	Resilience scores in the clinical population did not differ at a statistically significant level to healthy controls. Survivors of brain tumours without emotional difficulties had higher mean resilience scores than survivors and controls with emotional difficulties (t=4.32, p<.01 and t=3.67, p<.01 respectively).
2	Chou & Hunter (2009)	Taiwan	98	Mixed methods; questionnaire & semi-structured interviews	Resiliency Attitudes and Skills Profile (Hunter & Hurtes, 2001)	To investigate the relationship between risk factors, protective factors, resilience, and quality of life (QoL) in survivors of leukaemia and brain tumours.	Resilience was positively correlated with QoL and protective factor scale scores (r=.52, p<.01 and r=.98, p<.01 respectively). Inverse relationships were reported between resilience and illness-related risks and individual risks (r=-.38, p<.01 and r=-.77, p<.01 respectively). In regression analyses, resilience was not a statistically significant predictor of QoL.
3	Haase, Kinter, Monahan, & Ross (2014)	USA & Canada	202	Quantitative; cross-sectional	Haase Adolescent Resilience in Illness Scale (Haase, 2004)	To evaluate the Resilience in Illness Model.	A negative and statistically significant relationship between illness-related uncertainty and resilience was reported (r=-.32, p<.01). Correlational analyses revealed statistically significant relationships between resilience and variables including: support from family (r=.42, p<.01), peers (r=.32, p<.01), and health providers (r=.33, p<.01); confrontive (r=.31, p<.01), optimistic (r=.43, p<.01), and supportant coping (r=.30, p<.01); and hope-derived meaning from cancer experience (r=.69, p<.01). In Structural Equation Modelling, courageous coping and hope-derived meaning were identified as 'paths' to resilience.
4	Harper et al. (2014)	USA	103	Quantitative; cross-sectional	Resiliency Q-sort (Eisenberg et al., 2003)	To examine the relationship between effortful control, resilience, and QoL in	Higher scores on the resilience scale were associated with higher scores on five QOL subscales including communication and treatment anxiety (r=.30, p<.01)

						children with cancer.	and $r=.33$, $p<.01$). The three remaining QoL subscales (worry, pain and hurt, and procedure anxiety) were not correlated with resilience at a statistically significant level. Authors indicated that resilience mediated the relationship between effortful control and QoL.
5	Harper , Penner, Peterson, Albrecht, & Taub (2012)	USA	41	Quantitative; cross-sectional	Resiliency Q-sort (Eisenberg et al., 2003)	To examine the relationship between positive dispositional attributes (e.g., resilience) and pain/distress in children with cancer undergoing a medical procedure. To investigate if this relationship is mediated by caregivers' affect.	Higher resilience scores were inversely associated with self-reported pain ($r=-.36$, $p<.05$) and caregivers' reports of their child's distress ($r=-.37$, $p<.05$). A positive and statistically significant correlation between resilience and parents' empathic concern was documented ($r=.42$, $p<.01$). Authors reported that parents' empathic responses mediated the relationship between their child's resilience and pain/distress.
6	Im & Kim (2012)	South Korea	102	Quantitative; cross-sectional	Resilience Questionnaire (Kim & Yoo, 2010b)	To examine the relationship between resilience and demographic factors, illness-related variables, parenting practices, and peer and teacher relationships in children with atopic dermatitis.	Higher resilience scores were correlated with shorter duration of symptoms ($r=-.31$, $p<.05$), less severe symptoms ($r=-.33$, $p<.05$), better quality relationships with peers ($r=.34$, $p<.01$) and teachers ($r=.35$, $p<.01$), and higher maternal and paternal warmth-acceptance scores ($r=.38$, $p<.01$ and $r=.36$, $p<.01$ respectively). These variables explained 39% of the resilience model. Only shorter duration of illness ($\beta=-.39$, $p<.01$) and relationships with peers ($\beta=.30$, $p<.01$) were identified as significant predictors.
7	Kim & Im (2014)	South Korea	102	Quantitative; cross-sectional	Resilience Questionnaire (Kim & Yoo, 2010b)	To examine protective factors for internalizing and externalizing problems in children with dermatitis.	Higher resilience scores were associated with lower internalizing ($r=-.26$, $p<.05$) and externalizing difficulties scores ($r=-.25$, $p<.05$).
8	Kim & Yoo (2010a)	South Korea	74	Quantitative; cross-sectional	Resilience Questionnaire (Kim & Yoo,	To examine the relationship between resilience and demographic factors, illness-	Resilience was positively and significantly correlated with relationships with peers ($r=.53$, $p<.01$ teachers ($r=.32$, $p<.01$) and family functioning ($r=.54$, $p<.01$).

				2010b)	related variables, and relationships with peers, parents, and teachers in children with cancer.		In regression analysis only family functioning and peer relationships were significantly associated with resilience (β=.26, p<.05 and β=-58, p<.01 respectively). These variables accounted for 34.8% of variance in the regression model.
9	Powers (2011)	USA	20	Quantitative; cross-sectional	Resiliency Scales for Children and Adolescents (Prince-Embury, 2007)	To identify the relationship between demographic variables, resilience, QoL, and post-traumatic stress symptom (PTSS) severity in children with a burn injury.	Lower emotional reactivity and higher sense of relatedness scores (resilience subscales) were indicative of lower levels of PTSS severity (r= .67, p<.01 and r=-.49, p<.05 respectively). Resilience was correlated with child and parent reports of QoL (r=.76, p<.01 and r=.86, p<.01 respectively).
10	Smorti (2012)	Italy	80	Quantitative; case-control	Ego-Resiliency Scale (Block & Kremen, 1996)	To investigate the relationship between optimistic expectations of the future, resilience, and coping strategy use in adolescents after bone cancer treatment. To compare these variables to healthy controls.	The clinical population had lower resilience scores, (F=18.48, p<.01), and used more avoidant forms of coping than controls (F=92.11, p<.01). Optimistic expectations for the future were inversely correlated with resilience (r=-.46, p<.01). No statistically significant relationships between coping strategy use and resilience were reported in either population.
11	Wu, Sheen, Shu, & Chang (2013)	Taiwan	131	Quantitative; cross-sectional	Haase Adolescent Resilience in Illness Scale (Haase, 2004)	To identify factors related to anxiety and resilience in young people undergoing chemotherapy.	Resilience was positive correlated with cognitive coping (r=.71, p<.01), problem-oriented coping (r=.58, p<.01) and defensive coping (r=.25, p<.01). Defensive and cognitive coping were significant predictors of resilience in regression analyses (β=.75, p<.01, and β=-.17, p<.01) accounting for 46.5% of the regression model.
12	Wu et al. (in press)	USA	40	Quantitative; cross-sectional	Resilience Scale (Wagnild & Young, 1993)	To explore if resilience mediated the relationship between cancer symptom distress and QoL.	Resilience was correlated with cancer symptom distress (r=-.44, p<.01) and QoL (r=.56, p<.01). Resilience was reported to mediate the relationship between cancer symptom distress and QoL.

Quality Assessment

Overall, the methodological quality of the included studies was variable. Results of the quality assessment using the QATSDD are presented in Table 3.

Eleven studies did not conduct or failed to report a priori power calculations; study two was the exception. Particular concerns were raised regarding studies five, nine, and twelve where the studies were clearly underpowered for the analyses used. The results of the included studies should therefore be interpreted with caution due to the risk of type II error.

All studies were deemed to be of moderate risk of selection bias due to the use of convenience sampling via specific clinics and hospital wards; the recruitment of children through a burns camp register; and the exclusion of eligible participants in study one (based on their stated inclusion criteria) as participants were not considered to be attending follow-up appointments on a routine enough basis. Furthermore, five studies (studies one, two, five, six and seven) failed to adequately report the study response rate. The remaining studies cited response rates ranging from 8% (study nine) to 90% (study three). Based on the aforementioned factors, questions were raised regarding the representativeness of samples in relation to the target population identified by the study authors, the potential differences between responders and non-responders, and consequently the generalizability of the results.

With regards to missing data, five studies reported incomplete outcome data (studies three to seven inclusive). In studies reporting missing data, missing data accounted for 1-6% of study samples. The methods used to manage missing data (mean substitution or deletion of missing cases) were considered appropriate based on the small percentage of data missing. In studies where authors failed to report the percentage of missing data, it is unclear if bias may have been introduced into the sample by limiting the representatives of findings, compromising the validity and strength of relationships between variables under investigation, and affecting the external validity of the results.

Qualitative descriptions of the characteristics of the resilience measures used were

limited. Example sample items were provided in less than half of the studies (studies four, six, seven, eight, and twelve). However, all but one study (study nine) reported the statistical assessment of the internal reliability of the resilience measures used. The administration of the resilience measures was complemented with the use of a number of additional psychometric tools in all studies. Seven of the studies (one to six and 8) reported the administration of adapted and/or shortened versions of psychometric tools to meet the needs of their studies. In some instances questions were raised regarding the validity of these adapted measures. In studies two, three, four, five, and eight insufficient detail regarding the adapted measures was provided to enable the replication of studies. The remaining studies appeared to employ appropriate and adequate measures. The ratings for the rationale of data collection tools administered in each of the studies were rated as 'moderate' to 'complete'; justification cited by the authors for their choice of measurement tools included their use in previous relevant research, their fit with the target population, and the psychometric properties of the measures. One study (study five) was considered to have failed to adequately explain the rationale behind the statistical analyses chosen. In the majority of studies it was often not stated if possible confounding variables were controlled for in the analysis, which could therefore affect the interpretation of the results.

Assessment of the reliability of findings of the qualitative component of study two was not reported, whilst there was a 'moderate' fit between the research question and the format and content of the data collection tool. Selective reporting of outcome data was not considered to be an issue in any of the included studies. None of the authors cited service user involvement in the study design. No studies were considered to be 'complete' with regard to their critical appraisal of the research undertaken.

Table 3

Quality Assessment Ratings

Criteria	Study											
	1	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**	**10**	**11**	**12**
1. Explicit theoretical framework	3	3	3	0	0	0	0	0	3	1	3	3
2. Statement of aims/objectives in main body of report	3	3	2	3	3	3	3	3	3	3	3	3
3. Clear description of research setting	3	2	3	2	3	3	3	3	3	3	3	3
4. Evidence of sample size considered in terms of analysis	0	3	0	0	0	0	0	0	0	0	0	0
5. Representative sample of target group of a reasonable size	2	2	3	2	2	2	3	2	1	3	2	1
6. Description of procedure for data collection	2	2	3	3	3	3	3	3	3	3	3	3
7. Rationale for choice of data collection tool	2	2	2	2	2	2	3	3	3	2	2	1
8. Detailed recruitment data	0	0	1	3	0	0	0	3	3	3	3	3
9. Statistical assessment of reliability & validity of tool(s) (Quantitative only)	2	2	2	2	2	2	2	2	0	2	2	2
10. Fit between stated research question & method of data collection (Quantitative)	3	2	3	3	3	3	3	3	3	3	3	3
11. Fit between research question & format & content of data collection tool (Qualitative)	n/a	2	n/a	n/a	n/a	n/a	n/a	n/a	n/a	n/a	n/a	n/a
12. Fit between research question & method of analysis	2	2	3	3	3	3	2	3	3	3	2	3
13. Good justification for analytical method selected	2	2	3	3	0	2	2	2	3	2	2	2
14. Assessment of reliability of analytical process (Qualitative only)	n/a	0	n/a	n/a	n/a	n/a	n/a	n/a	n/a	n/a	n/a	n/a
15. Evidence of user involvement in design	0	0	0	0	0	0	0	0	0	0	0	0
16. Strengths & limitations critically discussed	2	2	2	2	2	2	2	2	2	1	1	2
Total score	26	29*	30	28	23	25	26	29	30	29	29	29

Criteria: 0=not at all; 1= very slightly; 2=moderately; 3=complete.
*NB: Possible maximum score on study 2 is 48. Maximum total score on the remainder of studies is 42.

Narrative Synthesis

Demographic factors.

Correlational analyses revealed no statistically significant relationships between the resilience of children and adolescents and the following variables: age (studies six, eight, and 11); gender; their perceived level of academic achievement; family structure; mother's level of education (studies six and eight); mother's age; father's age; father's level of education (study six); or receipt of government financial assistance/economic status (studies two, four, and six).

Illness-related variables.

Inconsistent findings were reported with respect to the relationship between illness-related variables and resilience. Studies eight and 11 highlighted no evidence for a statistically significant relationship between resilience and time since diagnosis in adolescents with cancer. However, in young people with atopic dermatitis (study six) the authors reported a medium inverse relationship between these two variables (r=-.31, p<.05). Study six also reported a negative correlation (r=-.33, p<.05) between condition severity and resilience. In subsequent regression analyses in study six, only duration of illness (β=-.39, p<.01) and relationship with peers were statistically significant predictors of resilience (β =.30, p<.01).

The authors of study two found no difference in resilience levels based on cancer type (brain tumour or leukaemia), whilst study four concluded that length of treatment was not reliably associated with resilience. Furthermore, the authors of study one found that there was no statistically significant difference in the resilience of young people with a brain tumour and a normative sample matched by gender, level of education, and area of living (urban or rural). In contrast, the authors of study 10 reported lower resilience scores in adolescents who were one month post bone cancer treatment compared to controls (F=18.48, p<.01). Crucially, in study 10 controls were not matched on key demographic variables. The authors of study four did, however, report a negative and statistically significant correlation between illness-related uncertainty and resilience (r=-.32, p<.01) in a sample of adolescents with cancer.

Familial and parental relationships.

Four studies (studies four, five, six, and eight) examined the relationship between the resilience of children and adolescents and parenting/family variables. Study six documented higher resilience scores in participants who reported higher levels of warmth and acceptance as demonstrated by their mothers ($r=.38$, $p<0.01$) and fathers ($r=.36$, $p<0.01$). Study five reported a statistically significant correlation between caregiver's empathic concern and their child's resilience ($r=.42$, $p<.01$). The authors of this study postulated that caregivers' empathic concern mediated the relationship between their child's resilience and the pain/distress they experienced during a medical procedure. However, the small sample size (n=41) limits the validity of this finding. Furthermore, the authors of studies three and eight reported positive correlations between the resilience of children and adolescents and family support (study three: $r=.42$, $p<.01$), adaptability (study three: $r=.46$, $p<.01$; study eight: $r=.47$, $p<.01$), and cohesion (study three: $r=.40$, $p<.01$; study eight: $r=.51$, $p<.01$). In subsequent regression analyses, family functioning was significantly associated with resilience in study eight ($\beta=.26$, $p<.05$).

Relationships with peers.

Three studies examined the relationship between resilience and the quality of peer relationships (studies three, six, and eight). With respect to the quality of peer relationships, statistically significant relationships between this variable and resilience were found in children with atopic dermatitis (study six: $r=.34$, $p<.01$) and adolescents with cancer (study three: $r=.32$, $p<.01$; study eight: $r=.52$, $p<.01$). Peer relationships were significantly associated with resilience in regression analyses in studies six and eight (study six: $\beta=.30$, $p<.01$; study eight: $\beta=.58$, $p<.01$).

Relationships with teachers and health care providers.

In studies six and eight, higher resilience scores were documented in children and adolescents who reported better quality relationships with their teachers (study six: $r=.35$, $p<.01$; study eight: $r=.32$, $p<.01$). However, in subsequent regression analyses this variable was not

significantly associated with resilience. Only one study (study three) explored the relationship between resilience and the support of health care providers, where a positive and statistically significant relationship was revealed ($r=.33$, $p<.01$).

Satisfaction with appearance.

Three studies contained quality of life measures which included subscales pertaining to appearance concerns (studies two, four, and 12). Results of study two suggested that adolescents with leukaemia had more body image concerns than those with brain tumours. Furthermore, the authors of study 12 suggested that although a subset of participants reported body image concerns in relation to their cancer treatment, the majority did not. Only one study included in the review (study four) examined the relationship between parent-reported physical appearance and resilience; this subscale was positively correlated with resilience at a statistically significant level ($r=.20$, $p< .05$).

Coping.

Inconsistent findings were reported regarding the relationship between coping strategy use and resilience; all studies involved children and adolescents with cancer. Results of study 10 indicated no significant correlations between these variables. Conversely, the authors of study 11 found large and statistically significant correlations between resilience and problem-oriented coping and cognitive coping ($r=.58$, $p<.01$ and $r=.71$, $p<.01$ respectively). The results from study three were mixed; the authors reported no significant relationship with resilience and defensive coping, whilst 'courageous' coping (confrontive, supportive and optimistic coping) were correlated with resilience at a statistically significant level ($r=.31$, $p<.01$; $r=.43$, $p<.01$; $r=.30$, $p<.01$ respectively). The authors of this study also reported that the results of structural equation modelling suggested that 'courageous' coping was a path to resilience.

Quality of life and psychological well-being.

Positive and statistically significant correlations were reported between resilience and quality of life as reported by children who had sustained a burn injury and parental report of

their child's quality of life (*r*=.76, *p*<.01 and *r*=.86, *p*<.01 respectively; study nine). These findings were further corroborated in evidence by studies two and 12 where positive and statistically significant correlations were found between resilience and child self-reported quality of life (studies two: *r*=.52, *p*<.01 and study 12: *r*=.56, *p*<.01) and informant reports of their child's quality of life (study 4) in cancer populations. However, in subsequent regression analyses in study two, resilience was not a statistically significant predictor of quality of life. The authors of study 12 also proposed that resilience mediated the relationship between cancer symptom distress (e.g., pain and fatigue) and quality of life. However, the small sample size for this method of analysis (n=40) limits the validity of this finding.

Studies also demonstrated that higher resilience scores were indicative of lower levels of: anxiety (study 11); internalising difficulties more generally (studies one and seven); externalising difficulties (study seven); pain/distress during medical procedures as rated by multiple informants (study five); and post-traumatic stress symptom severity (study nine). Self-esteem and confidence were also reported to be positively correlated with resilience, whilst 'courageous' coping and hope-derived meaning was reported to be paths to resilience in structural equation modelling (study four).

Discussion

Review of the twelve studies highlighted no evidence for statistically significant relationships between resilience and a range of demographic variables. Inconsistent findings were reported with regard to the relationship between resilience and illness-related variables such as the duration of illness/time since diagnosis. The inconsistencies in reporting between studies may be accounted for by the heterogeneity in the medical conditions under investigation, in this instance atopic dermatitis and cancer. Whilst some similarities can be drawn between symptoms of these conditions e.g., fatigue (De Jager, De Jong, Evers, Van De Kerkhof, & Seyger, 2011), they may be considered heterogeneous with regard to prognosis, degree of permanence, alongside the treatment trajectory and its intensity (Rumsey, 2002).

Even within the studies investigating resilience in cancer populations, participants were relatively heterogeneous with regard to their age, cancer type, and their stage of the cancer trajectory. For example, participants in study 12 were undergoing chemotherapy, whilst the inclusion criteria for study 11 specified that participants were at least one month post successful treatment. These disparities may account for the discrepancies in the research findings to date. Consequently, and similar to findings in a systematic review by Stewart and Yuen (2011) in adults with health conditions, no firm conclusions can be drawn regarding the relationship between resilience and demographic or illness-related variables. Furthermore, the findings for the relationship between resilience and coping strategy use were inconsistent. This may be indicative of wider issues in the coping literature with regard to methodological limitations and measurement error (see Compas, Jaser, Dunn, & Rodriguez, 2012).

Rowland (1990) reported that changes in appearance are the greatest source of stress for adolescents with cancer. Despite this, few studies in the current review examined the relationship between appearance concerns and resilience. In study four it is of note that there was a small and statistically significant relationship between perceived physical appearance and resilience. However, this rating was based on parental report and there is evidence to suggest poor levels of agreement between parental and child report of appearance related concerns (e.g., Thomas, Turner, Rumsey, Dowell, & Sandy 1997). Consequently, the strength of the relationship between satisfaction with appearance and resilience requires further investigation. The authors of study 12 reported that a minority of participants included in their study (adolescents undergoing treatment) reported body image concerns. Rumsey and Harcourt (2004) have suggested that people "disfigured by treatment for a life threatening condition such as cancer may suppress their appearance-related concerns during efforts to conquer the disease" (p. 87). Appearance related concerns may therefore change along the cancer treatment journey. Furthermore, the findings in study 12 may also be attributed to socially desirable responding; it has been suggested that patients with cancer may be reticent to disclose

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appearance related concerns when facing this potentially life-threatening condition as they may fear that others will regard their concerns as frivolous (Wallace, 2004).

There was more consensus amongst studies regarding the protective factors of relationships with peers and family members. Relationships with others (peers, family, teachers, and health providers) were consistently shown to be correlated or associated with resilience in adolescents, irrespective of medical condition. Findings were most reliable for the relationship between resilience in adolescents and peer relationships. These findings may reflect developmental changes within adolescence where affiliation with peers is a key task.

The links between resilience and psychological wellbeing were consistent across medical conditions. Higher resilience scores were associated with more favourable outcomes in terms of quality of life, and fewer internalising and externalising difficulties. These results are consistent with wider research findings where resilience has been described as the achievement of favourable outcomes despite adversity (Rutter, 2012).

Quality assessment revealed a number of limitations to the current evidence base which may have implications for the overall robustness of the findings and the conclusions that can be drawn. Similar to a systematic review investigating resilience in adults with chronic illness, there was an over-reliance on cross-sectional methodology (Cal et al., 2015). This precludes the examination of the direction of effects which could be rectified by employing longitudinal methods to examine the dynamic process of resilience over time. Consistent with findings by Stewart and Yuen (2011), this review highlighted that few studies examined the relationship between factors associated with resilience via mediational or moderation analyses. Perhaps the small sample sizes of studies included within this review precluded the use of more complex statistical analyses; a shortcoming that should be addressed in future research.

Studies rarely reported a priori power calculations and sample sizes were generally modest. Adequate sample sizes should be determined by a priori power calculations in future research to permit "the detection of realistic and clinically important effects" (Norman et al.,

2015, p. 308). Potential confounders should also be controlled for, for example, the presence of additional health conditions. Of note, half of studies included in the review were conducted in Asia; the influence of cultural factors on resilience (Ungar, 2012) may therefore limit the generalizability of these findings to other cultures.

Seven studies were excluded from the review as they did not employ the use of a specific resilience measure. This reflects wider issues in the field of resilience regarding a lack of consensus regarding the definition of this construct and a 'gold standard' for its measurement (Windle, Bennett, & Noyes, 2011). Furthermore, many studies used adapted or shortened measures and questions were raised regarding their validity and reliability. The use of valid and reliable tools in future research is recommended to ensure data quality. Obtaining reports from multiple informants may also increase the reliability of the data.

Studies that met the inclusion criteria for this review included children and adolescents with diagnoses of atopic dermatitis, cancer, and burns; all acquired conditions which may have resulted in temporary or permanent alterations in physical appearance. The review has highlighted that there is a paucity of studies examining resilience in children and young people with congenital visible difference, for example, craniofacial conditions which may present children and adolescents with a host of unique challenges. Furthermore, despite growing evidence highlighting the impact of medical conditions such as cancer, skin conditions, and burns and its treatment on appearance (e.g., Wallace, et al., 2007), only one study examined the relationship between appearance related concerns and resilience. Future research should therefore include this variable to further substantiate these findings.

Strengths and Limitations

This review consisted of a robust literature search across a number of relevant electronic databases. Data extraction and quality assessment was conducted independently by two reviewers to reduce error and increase reliability. Despite these strengths, a number of limitations to this review are acknowledged. Although efforts were made to limit publication

bias (e.g., by searching unpublished dissertations and contacting an expert in the field for unpublished research) it is possible that key studies were inadvertently excluded, for example, unpublished literature. For pragmatic reasons, the inclusion criteria for this review specified that all full text articles must be available in English. In doing so, the risk of publication bias is increased. Furthermore, the review is relatively small due to the few numbers of studies that met the inclusion criteria. However, in conducting the review, attention to the paucity of research in this area has been drawn and the need for further research in this area highlighted.

For practical reasons, chronological age was used to define the upper age limit for adolescence to inform the inclusion of studies in this review. However, age has been described as "a somewhat blunt instrument for defining developmental stage and cannot take into account early and late maturation" (Moss, Bailey, Griffiths, Lawson, & Williamson, in press, p. 17), factors which are of particular pertinence in appearance distress and research (Rogol, Clark, & Roemmich, 2000). As a dynamic process, resilience is reported to vary across the lifespan (Windle et al., 2011). The wide age range of participants included in this study (three to 26 years) may underpin the lack of uniformity in the synthesis of information regarding the relationships between resilience and demographic, illness-related, and psychological variables. A number of studies within the review included participants spanning a number of developmental stages, for example in study three participants were aged between 10-26 years, which precluded the ability to explore age cohort comparisons. This is therefore highlighted as an area of future research to determine if certain variables are associated with resilience at specific developmental stages.

A paucity of rigorous and validated quality appraisal tools for the assessment of non-randomised studies is acknowledged within the literature (Sanderson, Tatt, & Higgins, 2007). The QATSDD was elected as it can be used to assess the quality of studies employing a range of methodologies. However, in its attempts to be a holistic tool, some aspects have been compromised. For example, one factor deemed important in the quality assessment of case-

control studies, the appropriateness of the control sample and if they were matched for important characteristics, is not a requisite item on the QATSDD.

Clinical Implications

The results of the current review highlight clinical interventions focussed on developing positive familial and peer relationships may be useful first steps for clinicians working with children and adolescents with medical conditions associated with acquired visible difference. However, in a review by the Medical Research Council, the authors concluded that "more research has focussed on identifying protective factors that underlie the resilience process, but less on designing and testing interventions that might change negative outcomes" (Windle, Salisbury, & Ciesla, 2010, p. 2). Limitations to these resilience-enhancing interventions include a lack of or failure to explicitly state the underlying theoretical framework, and insufficient detail regarding the content of the intervention (Hammall, Heard, Inder, McGill, & Kay-Lambkin, 2014). The need for additional research in this area is therefore highlighted.

<div align="center">

Conclusion

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Results of the narrative synthesis have highlighted inconsistent findings regarding the relationship between illness-related variables and coping strategy use in children and adolescents with medical conditions associated with acquired visible difference. No evidence for the relationship between resilience and demographic variables was documented. Evidence for the protective role of social support, and the relationship between resilience and quality of life and psychological wellbeing was more conclusive. However, participants included in the review were heterogeneous with regard to age, medical condition, duration of illness, and treatment trajectory which may obscure findings. Lack of methodological rigour also limits the conclusions that can be drawn. The results presented are therefore tentative and recommendations for future research are made. Despite these limitations, fostering positive parental and peer relationships may be promising methods for promoting the resilience of young people with medical conditions associated with acquired visible difference.

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Predictors of Resilience in Adolescents with a Cleft Lip and/or Palate[1]

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Abstract

Objective: This cross-sectional study aimed to explore predictors of resilience in 12-16 years old with a cleft lip and/or palate. ***Methods***: Adolescents (*n*=126) were recruited via a regional cleft service and an online support group. The relationships between self-reported resilience (Connor-Davidson Resilience Scale) and peer relationships and experiences (Childhood Experience Questionnaire), family competence (Self-report Family Inventory-II), satisfaction with appearance (Satisfaction With Appearance questionnaire) and coping strategy use (KidCope) were explored. ***Results***: In multiple regression analysis, peer relationships and experiences (β =.33, *p*<.01), family competence (β =-.23, *p*<.01) and satisfaction with appearance (β =.20, *p*<.05) were significantly associated with resilience. Negative coping strategy use was negatively correlated with resilience; however, it was not included in the final regression model. ***Conclusions***: Clinical intervention should focus on developing peer and family relationships and addressing appearance-related cognitions. The need for additional research and evaluation of resilience-enhancing interventions generally, and within this population, is highlighted.

Keywords: cleft, resilience, peer relationships, satisfaction with appearance, family functioning, coping

Cleft lip and/or palate (CL/P) is one of the most common types of congenital craniofacial conditions, affecting approximately one in every 700 live births (Vieira, 2008). Caused by the incomplete fusion of facial structures during the first trimester of pregnancy, individuals affected by this condition may confront appearance and/or functional differences including speech, hearing, and feeding difficulties. The presence of a CL/P and its treatment may bring with it a number of additional life stressors, which have the potential to compromise the psychological well-being of those affected and the family unit as a whole (Rumsey & Stock, 2013).

Poor self-image (Lockhart, 2003), higher levels of social inhibition (Richman & Millard, 1997), and elevated risk of anxiety and depression (Ramstad, Otten & Shaw, 1995) have been documented in this population. However, research findings to date have failed to consistently demonstrate that young people affected by CL/P experience psychosocial adjustment difficulties (Hunt, Burden, Hepper, & Johnston, 2005). Furthermore, parents and adults affected by craniofacial conditions report that they have derived benefits from their experiences including: greater inner strength; a sense of purpose in life; adaptability/ flexibility (Eiserman, 2001); and resilience (O'Hanlon, Camic, & Shearer, 2012).

Alongside developments in surgical techniques, the routine provision of multidisciplinary cleft care, and social changes, a number of methodological reasons have been cited for the inconsistencies in cleft psychosocial literature to date (see Rumsey & Stock, 2013). As recent research trends have shifted away from deficit models of illness and psychopathology attention is drawn towards "a more positive psychology of visible difference" (Feragen, Borge, & Rumsey, 2009, p. 65), including resilience.

Multiple definitions for resilience have been proffered; however, based on the synthesis of over 270 peer reviewed journal articles, the Resilience and Healthy Ageing Network defined resilience as "the process of effectively negotiating, adapting to, or

managing significant sources of stress or trauma" (Windle, 2011, p. 152). Resources within the individual and their environment are essential in facilitating this dynamic process (Windle, 2011). Within paediatric populations, good relationships with friends have been identified as a strong protective factor in the resilience of school children with cancer and dermatitis (Im & Kim, 2012; Kim & Yoo, 2010). Positive and statistically significant correlations have also been found between the resilience of children with these medical conditions and parental warmth and acceptance (Im & Kim, 2012); parents' empathic responses (Harper, Penner, Peterson, Albrecht, & Taub, 2012); and family adaptability and cohesion (Kim & Yoo, 2010). However, inconsistent findings have been reported regarding the relationship between coping strategy use and resilience in young people with cancer (Smorti, 2012; Wu, Sheen, Shu, Chang, & Hsiao, 2013).

Although limited, resilience in parents and children has previously been investigated within the cleft population. For example, Feragen et al. (2009) reported that 'psychosocial resilience' was associated with higher satisfaction with appearance and lower levels of anxiety, depression, and self-reported teasing in ten year olds with a CL/P. Furthermore, in a mixed methods study, O'Hanlon et al. (2012) compared the self-reported resilience of parents with a CL/P who also had a child with a cleft to that of controls (i.e. parents of children with a cleft, but without a cleft themselves). Higher resilience scores were reported by parents with a diagnosis of cleft in comparison to controls, however this difference was small and not at a statistically significant level. Qualitative analysis of questionnaire data highlighted that all parents reported greater resilience in both themselves and their child as a result of their child's cleft.

Resilience in adolescents with CL/P, however, has not been systematically explored. Characterised by the developmental life stage of 'identity versus role confusion' (Erikson, 1959), adolescence presents with a host of unique challenges and transitions. According to

Erikson's (1959) theory of psychosocial development, successful resolution of this life stage is based on the presence or absence of a number of intrinsic and extrinsic factors, for example, individual characteristics, family characteristics, and social networks. Adolescence is a critical period of body image development (Wu et al., 2013). In accordance with a cognitive behavioural model of body image development (Cash, 2011), interpersonal experiences are postulated to influence appearance-related thoughts, feelings, and behaviours. As self-evaluations of appearance are considered to be particularly pertinent in adolescence, and in visible difference, the predictive value of satisfaction with appearance on resilience, in addition to peer and family relationships, was examined. Alongside social support, coping strategy use is regarded as one of the most important determinants of positive outcomes in those affected by a cleft (Baker, Owens, Stern, & Willmot, 2009). According to Lazarus and Folkman's (1984) transactional model of stress and coping, cognitive and/or behavioural coping strategies are adopted to "reduce psychological disruptions of homeostasis and psychological negative affect" (Suedfield, Krell, Wiebe, & Steel, 1997, p. 156) caused by challenge(s) appraised as stressful. As resilience requires individuals to successfully negotiate adversity to achieve positive outcomes, it is suggested that an adaptive coping style can result in resilience (Wu et al., 2013), therefore the predictive value of this construct was explored.

Due to a paucity of research, this study aimed to discern which intrinsic and extrinsic variables were most associated with resilience in adolescents with a CL/P, while building upon the shortcomings identified in the research by Feragen and colleagues (2009), including the absence of a validated measure of resilience and heavy reliance on parental report despite numerous studies highlighting poor agreement between parent and child report in the cleft population (e.g., Thomas, Turner, Rumsey, Dowell, & Sandy, 1997). As adolescence is considered a critical period of development, early intervention and/or preventative efforts have the potential to confer benefits throughout the lifespan, reducing the need for future

mental health service use. It was hoped that further investigation would develop our understanding of protective factors to provide a framework for possible psychological interventions for young people experiencing difficulties as a result of their cleft; support those who thrive to do so at higher levels (Eiserman, 2001); and inform service development. It was hypothesised that higher satisfaction with appearance, positive peer experiences and relationships, family functioning, and positive coping strategy use, would each account for significant unique variance in adolescent resilience scores when controlling for the possible effects of gender, age, visibility of cleft, and the presence of additional conditions.

Method

Participants

A sample of 126 young people aged 12-16 years (inclusive) with a diagnosis of a CL/P were recruited into the study via opportunistic and self-selected sampling between October 2014 and March 2015. As the research aimed to accurately represent the cleft population as whole, adolescents with all cleft types, associated syndromes, and additional health needs were eligible to participate. Inability to read/communicate in English and age restrictions were the only exclusion criteria. The sample comprised of 58 males (46%) and 68 females (54%), with a mean age of 14.24 (*SD* 1.40) years.

Materials

Participants completed a demographic information sheet (Appendix C); details on their age, gender, cleft type, and the presence and type of any additional conditions were collated. Participants completed the following self-report questionnaires, the order of which was randomised (Appendix C):

Resilience: The 25 item Connor-Davidson Resilience Scale (CD-RISC; Connor & Davidson, 2003) measures self-reported resilience. Responses to statements such as 'I tend to bounce back after illness, injury, or other hardships' were rated on a 5-point Likert scale. The

authors report good psychometric properties including divergent and convergent validity, high test-retest reliability (.87), and good internal consistency (Cronbach's alpha = .89; Connor & Davidson, 2003). In the current study, Cronbach's α was .92.

Satisfaction with appearance: The 20 item Satisfaction With Appearance questionnaire (SWA), developed by the Cleft Psychology Special Interest Group (2007), was used to measure respondents' satisfaction with aspects of their appearance (e.g., lips, nose, profile) on a 10-point Likert scale. Two items (satisfaction with braces and hearing aid) were not relevant for the majority of respondents. If this was the case, respondents were asked to omit these questions. A mean score was calculated for each respondent based on the number of items they completed. Emerson and colleagues (2004) reports that the SWA demonstrates excellent internal reliability (Cronbach's α=.90). In the current study, the Cronbach alpha coefficient was .89 for the 18 item version.

Positive peer relationships and experiences: The 20 item self-reported Childhood Experience Questionnaire (CEQ; Centre for Appearance Research, adapted from Pertschuk and Whitaker, 1982) was administered to measure the presence of positive peer experiences and relationships. To reduce response bias, ten statements are positively worded e.g., 'I go to parties' and 10 negatively worded e.g., 'I am teased.' Items are rated on a 5-point Likert scale. Negatively worded items are reverse scored so that higher scores on the CEQ reflect more positive peer relationships and experiences. The CEQ is reported to possess good internal consistency (Cronbach's α=.82; Emerson, Spencer-Bowdag, & Bates, 2004). In the current study, the Cronbach alpha coefficient was .83.

Family functioning: The 36 item Self-report Family Inventory-II (SFI-II; Beavers & Hampson, 1990) measures self-reported perceptions of family competence, cohesion, conflict, directive leadership, and emotional expressiveness. Example item include 'we argue a lot' (family conflict) and 'our family is good at solving problems together' (family

competence). The five subscales map onto two higher-order dimensions; family competence (i.e. the structure and adaptive flexibility of the family unit) and family style. Beavers and Hampson (2000) report Cronbach's α between .84 and .93. In the current study, the Cronbach alpha coefficient was .90. Preliminary analysis revealed a large and statistically significant inter-correlation between the family competence and family style subscales ($r=.82$, $p<.01$), which violated the assumption of singularity. Consequently, as recommended by Tabachnick and Fidell (2013), the family style subscale was removed as all items on this subscale were included within the more comprehensive family competence subscale.

Coping: Based on their age, participants completed either the child (7-12 year olds) or adolescent (13 years and older) version of the KidCope checklist (Spirito, Stark & Williams, 1988); a self-reported measure of a range of cognitive and behavioural coping strategies in response to a stressor. Each strategy can be categorised as either 'negative coping' including social withdrawal (e.g., 'I stayed by myself') and self-criticism (e.g., 'I blamed myself for causing the problem') or 'positive coping' such as problem-solving (e.g., 'I tried to fix the problem by thinking of answers') and cognitive restructuring (e.g., 'I tried to see the good side of things'). Spirito et al. (1988) report the measure possesses moderate test-retest reliability (values ranging from .41 to .83) and concurrent validity. In the current study, the Cronbach's α were .87 and .81 for the child and adolescent versions respectively.

Procedure

Ethical approval for the study was sought and obtained from the National Research Ethics Service via the Integrated Research Application System (IRAS) application (Appendix D). As demonstrated in Figure 3, participants were recruited via multiple methods.

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┌─────────────────┐      ┌─────────────────┐      ┌─────────────────┐
│ 698 packs sent  │      │ 35 viewed online│      │ 54 approached in│
│ via post by a   │      │ site via support│      │ out-patient     │
│ regional cleft  │      │ group           │      │ clinic waiting  │
│ service         │      │ advertisement   │      │ area            │
└─────────────────┘      └─────────────────┘      └─────────────────┘
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┌───────────────────────┐                         ┌───────────────────────┐
│ 5 x packs returned as │                         │ 1 x had already       │
│ not known at address  │                         │ participated online   │
│ 2 x parents declined  │                         │ 1 x parent declined   │
│ due to child's        │                         │ due to child's        │
│ additional health     │                         │ additional health     │
│ needs                 │                         │ needs                 │
│ 4 x parents did not   │                         │ 2 x requested packs to│
│ think the research was│                         │ take home & failed to │
│ relevant to their     │                         │ return by post        │
│ child                 │                         │                       │
└───────────────────────┘                         └───────────────────────┘
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┌─────────────────┐      ┌─────────────────┐      ┌─────────────────┐
│ 74 assented/    │      │ 22 assented/    │      │ 50 assented/    │
│ consented to    │      │ consented to    │      │ consented to    │
│ participate     │      │ participate     │      │ participate     │
└─────────────────┘      └─────────────────┘      └─────────────────┘
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┌─────────────────┐      ┌─────────────────┐      ┌─────────────────┐
│ 61 completed    │      │ 15 completed    │      │ 50 completed    │
│ questionnaires  │      │ questionnaires  │      │ questionnaires  │
│ online          │      │ online          │      │ in clinic       │
│ (9% response    │      │ (43% response   │      │ (93% response   │
│ rate)           │      │ rate)           │      │ rate)           │
└─────────────────┘      └─────────────────┘      └─────────────────┘
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                         ┌─────────────────┐
                         │ Total of 126    │
                         │ questionnaires  │
                         │ completed (16%) │
                         └─────────────────┘
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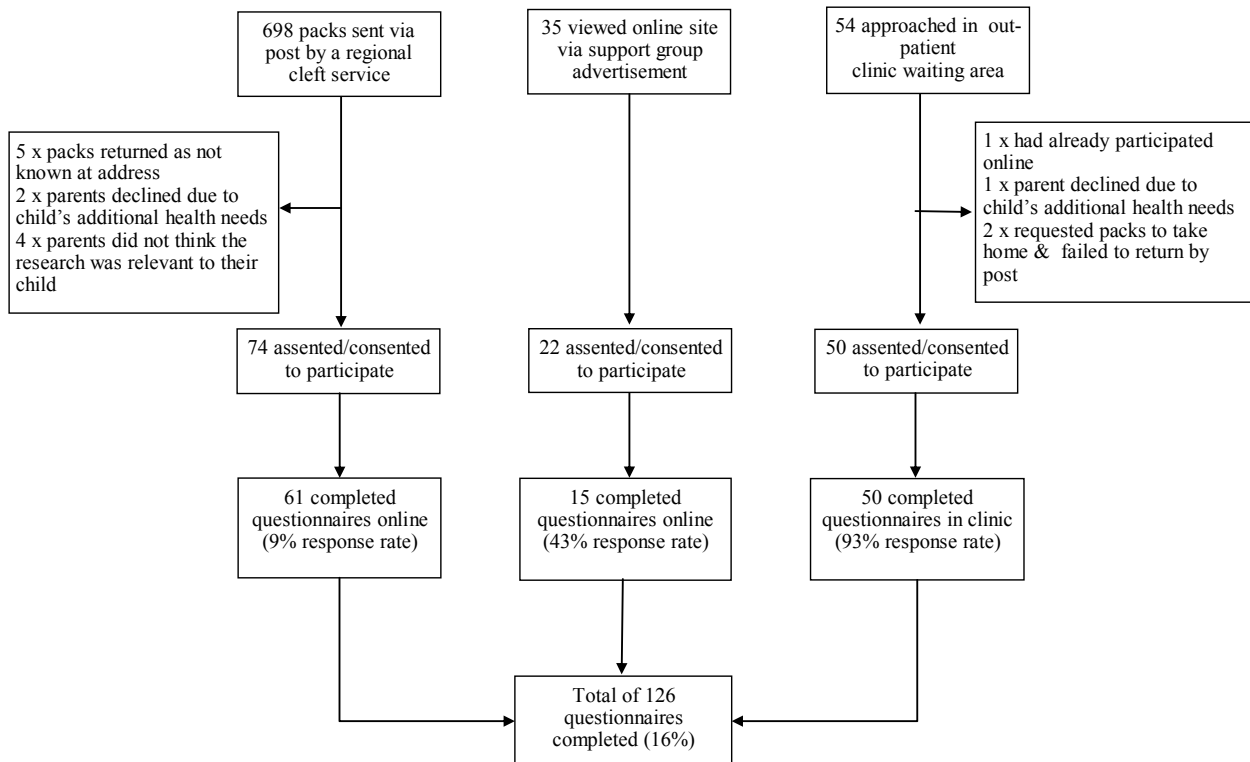
Figure 3. Flow of respondents through the study and response rates

Advertisements (Appendix E) were listed on the website, e-newsletter and social media sites of the Cleft Lip and Palate Association (CLAPA; a CL/P support group), whilst letters (Appendix E) were sent by post to 16 year olds and the parent/guardian of 12-15 years olds (n=698) under the care of a regional National Health Service (NHS) CL/P service in the North West of England. The advertisement and letters contained information regarding the study (including details on confidentiality, consent/assent, and the right to withdraw) and a link which participants could either click on or type into their web browser to enable them to access the study questionnaires on a secure online survey website. Adolescents aged 12-15 years old were required to indicate that they had sought parental consent to participate in the study before proceeding with the completion of the online assent form and questionnaires, whilst 16 year olds were required to complete a consent form (Appendix F). For postal recruitment methods, a reminder letter (Appendix G) was sent to potential participants or their parent/guardian six to eight weeks following the initial letter of invitation to the study.

Patients attending an outpatient appointment at the aforementioned cleft service were approached to participate in the study and provided with age appropriate study information leaflets (n=54) in the clinic waiting area. Potential participants were invited to complete the questionnaire pack in the clinic waiting area, in a private room with the assistance of the main researcher, or at home and return the questionnaires in a pre-paid envelope. Consent (16 year olds) or assent (12-15 year olds) was inferred on the completion of questionnaires. It took participants approximately 20 minutes to complete the questionnaires. Participants were provided with the opportunity to opt into a prize draw to thank them for their time and effort. The overall study response rate was 16%; response rates ranged from 9% for postal recruitment to 93% for clinic recruitment.

Data Analyses

On the basis of a priori power calculation using G*Power (version 3.1; Faul, Erdfelder, Lang, & Buchner, 2007), an approximate sample size of 114 participants was required to detect a medium sized effect (f^2=.15) with power of 80% when employing the α = .05 criterion of statistical significance for nine predictor variables in multiple regression analyses. The effect size estimation was derived from examination of previous paediatric resilience research where small to medium effects have typically been observed. SPSS version 22 was used to analyse the data. The binary variables of gender (male or female), visibility of cleft (non-visible [i.e. cleft of the palate only] or visible [i.e. cleft involving the lip]), and additional conditions (absent or present) were coded as either zero or one for the purpose of data analysis. Pearson's correlation coefficient or Spearman's rank correlation coefficient were used to examine the relationship between resilience and the independent variables. Hierarchical multiple regression was used to determine the unique contribution of the independent variables (peer relationships and experiences, satisfaction with appearance, family competence, positive and negative coping strategy use) on resilience when controlling

for age, gender, visibility of cleft, and the presence of additional conditions. Mann-Whitney U was used to determine if adolescents with and without additional health conditions differed in their reports of negative coping strategy use and peer relationships and experiences.

Results

Data Screening

Visual inspection of histograms, calculation of the skewness and kurtosis scores, and the use of the Kolmogorov-Smirnov Test (Appendix H) assisted in determining the distribution of the data. Exploration of the data revealed that resilience scores were normally distributed and met the assumptions of parametric tests. As the remaining variables were positively skewed, Spearman's rank coefficient was used for correlational analyses.

Preliminarily analyses confirmed that the assumptions of normality, linearity, and homoscedasticity were satisfied (Appendix H). No cases had associated Cook's distances over 1 or Mahalanobis distance scores exceeding the critical chi square value of 27.88 and were therefore not judged as having an undue influence upon the model (Tabachnick & Fiddell, 2013). Overall, less than 4% of the data for the outcome and predictor variables was missing. Results of Little's MCAR test indicated that the data were missing completely at random (chi-square $= 28.84$, $df = 96$, $p = .34$). To address the missing data, person-mean imputation was used ($n=34$ instances) to calculate questionnaire totals and subscale totals in cases where three items or less from the same measure were missing. In instances where respondents had omitted four or more responses from one questionnaire they were excluded from the analysis by the pairwise deletion method, as recommended by Pallant (2013).

Sample Characteristics

Seventy three participants (59%) indicated that they had a visible cleft (a cleft involving the lip) with the remainder having a non-visible cleft (cleft palate only). The most

commonly occurring additional medical condition was asthma (6%). The main demographic

and clinical characteristics of respondents are presented in Table 4.

Table 4

Demographic and Clinical Characteristics of the Sample (n=126)

Variable	Categories	*N* (%)
Gender	Male	58 (46%)
	Female	68 (54%)
Age	12 years old	18 (15%)
	13 years old	24 (20%)
	14 years old	22 (18%)
	15 years old	29 (24%)
	16 years old	30 (24%)
Cleft type (missing n=2)	Visible Cleft	73 (59%)
	Non-visible cleft	51 (41%)
Additional conditions	Present	39 (31%)
	One condition	27 (21%)
	Two conditions	10 (8%)
	Three conditions	2 (2%)
	Absent	87 (69%)

Note: Totals of percentages are not 100 for every characteristic because of rounding

Correlations

As indicated in Table 5, no statistically significant correlations between resilience and

demographic or clinical variables were found. Correlational analyses suggested that resilience

was positively associated with positive peer relationships and experiences (r_s =.52, *p*<.01)

and satisfaction with appearance (r_s =.45, *p*<0.01), and inversely associated with negative

coping strategy use (r_s =-.33, *p*<.01) and family competence (r_s =-.40, *p*<.01). As lower

scores on the SFI-II indicate greater family competence, the results suggest that increased

family competence is associated with increased resilience.

Hierarchical Multiple Regression

As illustrated in Table 6, the demographic and clinical variables entered at step 1 of

the model explained 3.6% of the variance in resilience scores. After the entry of peer

relationships, family competence, satisfaction with appearance, and positive and negative

coping strategy use at step 2, an additional 39.3% of the variance in resilience scores was

accounted for ΔR^2 =.39, ΔF (5, 107)=14.77, *p*<.01.

Table 5

Correlation Matrix of Study Variables, Means, Standard Deviations (SD), and Ranges within the Sample

Variable	1	2	3	4	5	6	7	8	9	10	Mean (SD)	Range in sample
1. Resilience	—										65.87 (16.69)	19-99
2. Visibility of cleft	.02	—									-	-
3. Gender	-.08	-.06	—								-	-
4. Age	-.07	.01	.05	—							14.24 (1.40)	12-16
5. Presence of additional conditions	-.16	-.25**	-.07	.07	—						-	0-3
6. Peer relationships & experiences	.52**	.036	-.07	-.06	-.26**	—					52.33 (10.85)	18-73
7. Satisfaction with appearance	.45**	-.11	-.19*	-.14	-.15	.44**	—				7.09 (1.62)	2.40-10.00
8. Family competence	-.40**	.09	.09	.21*	-.03	-.29**	-.35**	—			37.66 (11.41)	20-69
9. Positive coping	.14	-.10	-.03	.08	.13	-.05	.00	-.11	—		3.09 (1.08)	0-4
10. Negative coping	-.33**	-.05	.10	.15	.19*	-.42**	-.34**	.26**	.26**	—	4.19 (1.90)	0-7

NB: Lower scores on the family competence subscale of the SFI-II indicate greater family competence. $*p<0.05$, $** p<0.01$

Table 6

Multiple Hierarchical Regression Beta Values and Confidence Intervals (CI), Standardised Beta Values (β)and Standard Errors (SE.B), R^2, and Semi-partial Correlations (sr)

	B	95% CI	*SE.B*	β	R^2	sr
Step 1						
Visibility of cleft	.05	-6.96, 5.90	3.25	-.02	-	-.02
Gender	-3.06	-9.23, 3.12	3.12	-.09	-	-.09
Age	-.60	-2.80, 1.61	1.11	-.05	-	-.05
Additional conditions	-5.94	-12.80, .93	3.47	-.17	-	-.16
					.04	
Step 2						
Visibility of cleft	1.48	-3.65, 6.60	2.59	.04	-	.04
Gender	1.33	-3.68, 6.35	2.53	.04	-	.04
Age	.38	1.43, 2.18	.91	.03	-	.03
Additional conditions	-.83	-6.57, 4.91	2.90	-.02	-	-.02
Peer relationships & experiences	.51**	.22, .79	.14	.33	-	.26
Satisfaction with appearance	2.09*	.23, 3.95	.94	.20	-	.16
Family competence	.33**	-.57, .09	.12	-.23	-	-.20
Positive coping	1.85	-.64, 4.35	1.26	.12	-	.11
Negative coping	-1.00	-2.57, .56	.79	-.11	-	-.09
					.43	

*p<0.05, ** p<0.01

While the frequency of self-reported negative coping strategy use demonstrated a statistically significant relationship with resilience in correlation analyses, it was not a statistically significant predictor of resilience in the regression model. In the final model, only family competence, peer relationships and experiences, and satisfaction with appearance were statistically significant, with the peer relationships and experiences score reporting a higher beta value (β =.33, *p*<.01) than family competence (β =-.23, *p*<.01) and satisfaction with appearance (β =.20, *p*<.05). Statistically significant results are summarised in table 7.

Table 7: Summary Table Showing Standardised Beta Values (β) for Variables Identified as Statistically Significant Predictors of Resilience

Variable	β
Peer relationships & experiences	.33**
Satisfaction with appearance	.20*
Family competence	-.23**

*p<0.05, ** p<0.01

Semi-partial correlations indicated that peer relationships and experiences (sr=.26, p<.01), satisfaction with appearance (sr=.16, p<.05), and family functioning (sr= -.20, p<.05) each made a significant unique contribution in predicting resilience when controlling for the effects of the other variables included in the model; the effect sizes were small.

Additional Analyses

A Mann-Whitney U test revealed that adolescents with additional conditions were significantly more likely to report the use of more of negative coping strategies (U=1173.00, Z= -2.09, p, <.05, r=.19) and report less positive peer relationships and experiences (U=1118.50, Z= -2.83, p<.01, r=.25).

Discussion

Consistent with the study hypotheses, variables significantly associated with resilience in the regression model were family competence, peer relationships and experiences, and satisfaction with appearance. The hypothesis that positive coping was associated with resilience was not supported.

Demographic and Clinical Variables

Similar to research in children with medical conditions such as dermatitis, cancer, and CL/P, no statistically significant correlations between resilience and age or gender were found (Feragen et al., 2009; Im & Kim, 2012; Kim & Yoo, 2010). Furthermore, adolescents with a visible cleft did not differ in levels or resilience in comparison to their counterparts with a non-visible cleft. These findings are similar to findings by Feragen et al. (2009) and wider evidence suggesting that the objective severity and visibility of a cleft are not consistently related to psychological outcome (Appearance Research Collaboration, 2009). Resilience scores were slightly lower in adolescents with additional conditions, however this difference was not statistically significant. Notably, less positive peer relationships and experiences and the use of more negative forms of coping were reported by adolescents with

co-occurring conditions. This subgroup is reported to be more likely to experience negative social experiences and emotional distress in comparison to children with a cleft alone (Feragen & Stock, 2014). Perhaps the lower resilience scores in this subgroup is due to the diminished availability of these internal and external resources to draw upon in times of adversity. For example, social withdrawal, a form of negative coping, is proposed to maintain isolation and the likelihood of adverse social experiences via a 'negative social interactional cycle' (Robinson, Rumsey, & Partridge, 1996). Results of the current study are consistent with those of Feragen et al. (2009) who reported that the presence of co-occurring conditions was "not consistently associated with psychosocial non-resilience" (p. 71).

Family Functioning

Family competence (i.e. family responsiveness and adaptability) was identified as a statistically significant predictor of resilience. Young people who reported higher resilience scores also reported that their families were more able to negotiate, function, and deal effectively with the stressful situations that may arise from the presence of a cleft and/or its treatment. These findings were in accordance with previous research within paediatric populations where the importance of the relationship between resilience and parenting/family variables such as warmth, acceptance (Im & Kim, 2012), affection (Kim & Yoo, 2007), adaptability, and cohesion (Kim & Yoo, 2010) have been highlighted.

Peer Relationships and Experiences

Peer group affiliation is a key task in the developmental stage of 'identity versus role confusion' (Erikson, 1959). In the present study, the role of positive peer relationships and experiences, both in and outside of the school environment, in facilitating resilience was indicated. These findings are further corroborated in research by Im and Kim (2012) and Kim and Yoo (2010) who revealed statistically significant relationships between resilience and the quality and strength of peer relationships in adolescents with atopic dermatitis and cancer.

Satisfaction with Appearance

Appearance is considered to be particularly salient during adolescence. Consistent with findings of Feragen et al. (2009) and Harper et al. (2014), results from the current study indicate a positive and statistically significant relationship between satisfaction with appearance and resilience, with this variable being included in the final regression model. A strong and positive association between social experiences and satisfaction with appearance was also revealed; young people who reported more positive peer relationships and experiences reported increased satisfaction with aspects of their physical appearance. Such findings are supportive of Cash's (2011) cognitive behavioural model of body image development where interpersonal experiences are postulated to influence appearance-related thoughts, feelings, and behaviour. Equally, those more satisfied with their appearance may interpret their social experiences in a more positive light. However, due to the cross-sectional nature of the study, causality or the direction of this relationship cannot be confirmed.

Coping Strategy Use

As both peer and family relationships were significantly associated with resilience, it was anticipated that adolescents would mobilise the support systems around them via positive coping strategy use to assist them in 'bouncing back' from any potential challenges arising from their cleft. However, neither positive nor negative coping use significantly contributed to the final regression model. Within the existing resilience and coping literature in paediatric populations, inconsistent findings are apparent in investigating the relationship between these two variables. Smorti (2012) indicated no significant correlations between coping and resilience, whilst Wu et al. (2013) reported positive relationships between resilience and both problem-oriented and cognitive coping. The aforementioned inconsistencies in this study and previous research may reflect methodological limitations and measurement error acknowledged within the existing coping literature (see Compas, Jaser, Dunn, & Rodriguez,

2012). Due to its brevity, the KidCope is a popular screen of coping strategy use in paediatric populations; however, its conciseness yields less detailed information and its single item scales compromise its psychometric properties (Blount et al., 2008). The controllability/ perceived controllability of a stressor is deemed influential in determining the effectiveness of coping strategy use (Compas et al., 2012). However, this factor was not included in the KidCope and is considered a limitation.

Strengths

Children and young people with additional syndromes are often excluded from CL/P research or their difficulties are often not adequately recorded or accounted for in statistical analyses (Feragen, Stock, & Rumsey, 2014). The adoption of an inclusive approach, and controlling for and recording of additional conditions, are considered strengths of this study. When considering the impact of familial variables, most research within the CL/P population has focussed on mothers of children with a cleft (Rumsey & Stock, 2013). This study therefore addresses a gap in the evidence base by investigating the role of wider family functioning. The use of self-report, and the employment of a validated measure of resilience (instead of a proxy measure, a limitation of many resilience studies [Hjemdal, Friborg, Stiles, Martinussen, & Rosenvinge, 2006]) are also considered strengths.

Limitations

The cross-sectional nature of the current study is seen as a limitation as it obviates the possibility of exploring the direction of effects and causal inferences cannot be drawn. Although the sample size was adequate for the methods of analyses used, it was modest and precluded the use of more complex statistical analyses to gain a better understanding of the interplay between the variables under investigation. Despite the presence of procedures to maximise participation (e.g., reminder letters), the study response rate was poor and notably lower than previous cleft postal research, where rates of 34% have been achieved in parent-

adolescent dyads (Berger & Dalton, 2011) and 45% in parents alone (O'Hanlon et al., 2012). Possible reasons for the low response rate in the current study include time constraints, reticence by parents or young people to share personal information online, and because the research was solely conducted with adolescents. It is also possible that non-responders were, for example, less resilient than responders which may introduce a potential risk of bias. Furthermore, approximately half of participants were recruited via cleft treatment clinics and an online support group (populations actively seeking treatment or support). These factors raise questions about the representativeness of the sample and generalizability of the results.

All measures in the study were based on self-report, which is vulnerable to socially desirable responding. In addition to the aforementioned limitations of the KidCope, questions have been raised regarding the factor structure of the SFI-II (Goodrich, Selig, & Trahan, 2012). Consequently, further research is required. Although the study controlled for possible confounding variables such as the presence of additional conditions, other potentially influential variables including parental well-being and cultural factors were not measured or accounted for in the analysis. Future research would build on these shortcomings by employing a larger sample size and utilising longitudinal methods to enable more complex multivariate analysis. The inclusion of parental factors would be beneficial, in addition to gaining reports from multiple informants to increase the reliability of the data.

Clinical Implications

In 2013, the Department of Health (DoH) recognised adolescence as a period of "immense potential for preventive interventions and building resilience" (DoH, 2013, p. 158). Previous research aimed at enhancing resilience in paediatric populations has highlighted the need to adapt psychosocial interventions to meet the challenges and demands arising from individual chronic conditions (Mullins et al., 2015). Consistent with its theoretical basis, findings of the current study confirm that resilience operates across multiple levels; factors

within the individual (satisfaction with appearance) and their environment (family and peer relationships) were revealed as significant correlates in the regression model, indicating areas of potential clinical intervention. A review by Windle, Salisbury, and Ciesla (2010) highlighted that research focused on the development and evaluation of resilience-enhancing interventions is sparse. To the author's knowledge, no published literature evaluates the effectiveness of resilience-enhancing interventions in the cleft or visible difference populations. Furthermore, a systematic review by Norman et al. (2015) identified a paucity of methodologically robust studies evaluating the effectiveness of psychosocial interventions for those affected by CL/P, which precluded the authors' ability to draw firm conclusions about the optimal format, timing, and intensity of interventions. Nevertheless, Norman et al. (2015) tentatively suggested that social skills training and CBT may be beneficial in improving social support and positive peer interactions, and reducing appearance related-concerns. As the influence of positive peer relationships and experiences appears to be related to satisfaction with appearance and resilience, this may be an appropriate area for clinical intervention in order to enhance resilience in this population; however, more research is required.

Support groups may also provide a useful forum for young people with visible difference to bolster self-esteem and confidence, share experiences, learn new ways of managing questions and comments in relation to their appearance, and build friendships (Changing Faces, 2015). A number of UK based cleft teams and charities (e.g. CLAPA and Changing Faces) offer group based activity days and workshops for children and young people affected by visible difference, however there is no published evaluation documenting the effectiveness of these interventions. Results of the current study also highlight the possible protective role of family competence in contributing to the resilience of adolescents with a cleft; therefore another potential area for clinical intervention is indicated. Once again,

given that currently no published research evaluating the impact of interventions to improve family competence (e.g., family therapy) in this population is available, further research is required .

In addition to multi-centre trials to ensure adequately powered studies, rigorous randomized controlled trials are required to enable more concrete conclusions to be draw regarding the most effective way of intervening in this clinical population (Norman et al., 2015). Furthermore, the recent formation of The Cleft Collective, a UK based cohort study reported to be the world's largest cleft research programme (The Cleft Collective, 2015), may provide the necessary platform to enable the collation of much needed longitudinal data to examine psychosocial adjustment to cleft longitudinally (Norman et al., 2015). With their varied skill set and position within the cleft multidisciplinary team, clinical psychologists are well positioned to be involved in the design, delivery, and evaluation of resilience-enhancing interventions in adolescents with a cleft, and to disseminate outcomes to inform service provision. It is hoped that increased research efforts in this area can subsequently inform evidence based clinical interventions to lead to improved outcomes in this clinical population.

Conclusion

The current study supported the relationship between resilience and satisfaction with appearance, peer relationships and experiences, and family functioning in adolescents with a CL/P. Such findings can assist in considering interventions to enhance resilience in this population. However, further research and evaluation of interventions is required.

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Appendices

Author Guidelines for the Journal of Paediatric Psychology

Instructions to Authors

The *Journal of Pediatric Psychology* is an official publication of the Society of Pediatric Psychology, Division 54 of the American Psychological Association. JPP publishes articles related to theory, research, and professional practice in pediatric psychology.

Types of Manuscripts:
• Original research, including case studies
• Review articles
• Commentaries

Organization of manuscripts

Manuscript Central will guide authors through the submission steps, including: Abstract, Keyword selection, and the Manuscript. The manuscript must contain an Introduction, Methods, Results, Discussion, Acknowledgements and Reference List.

Length of manuscript: Original research articles should not exceed 25 pages, in total, including references, figures, tables, etc. In the case of papers that report on multiple studies or those with methodologies that necessitate detailed explanation, the authors should justify longer manuscript length to the Editor in the cover letter. Case reports should not exceed 20 pages. Review articles should not exceed 30 pages. Commentaries should not exceed 4 pages. The Journal of Pediatric Psychology no longer accepts brief reports but will accept manuscripts that are shorter in length than the 25 page manuscripts.

Manuscripts (text, references, tables, figures, etc.) should be prepared in detailed accord with the Publication Manual of the American Psychological Association (6th ed.). There are two exceptions:

(a) The academic degrees of authors should be placed on the title page following their names, and

(b) a structured abstract of not more than 150 words should be included. The abstract should include the following parts:

(1) Objective (brief statement of the purpose of the study);
(2) Methods (summary of the participants, design, measures, procedure);
(3) Results (the primary findings of this work); and
(4) Conclusions (statement of implications of these data).

Key words should be included, consistent with APA style. Submissions should be double-spaced throughout, with margins of at least 1 inch and font size of 12 points (or 26 lines per page, 12-15 characters per inch). Authors should remove all identifying information from the body of the manuscript so that peer reviewers will be unable to recognize the authors and their affiliations. E-mail addresses, whenever possible, should be included in the author note.

Informed consent and ethical treatment of study participants. Authors should indicate in the Method section of relevant manuscripts how informed consent was obtained and report the approval of the study by the appropriate Institutional Review Board(s). Authors will also be asked to sign a statement, provided by the Editor that they have complied with the American Psychological Association Ethical Principles with regard to the treatment of their sample.

Clinical relevance of the research should be incorporated into the manuscripts. There is no special section on clinical implications, but authors should integrate implications for practice, as appropriate, into papers.

Terminology should be sensitive to the individual who has a disease or disability. The Editors endorse the concept of "people first, not their disability." Terminology should reflect the "person with a disability" (e.g., children with diabetes, persons with HIV infection, families of children with cancer) rather than the condition as an adjective (e.g., diabetic children, HIV patients, cancer families). Nonsexist language should be used.

Appendix B

Quality Assessment Tool for Studies of Diverse Design Scoring Guidance Notes

This text box is where the unedited thesis included the following third party copyrighted material:

Sirriyeh, R., Lawton, R., Gardner, P., & Armitage, G. (2012). Reviewing studies with diverse designs: the development and evaluation of a new tool. *Journal of Evaluation in Clinical Practice*, *18*, 746-752. doi: 10.1111/j.1365-2753.2011.01662.x

This text box is where the unedited thesis included the following third party copyrighted material:

Sirriyeh, R., Lawton, R., Gardner, P., & Armitage, G. (2012). Reviewing studies with diverse designs: the development and evaluation of a new tool. *Journal of Evaluation in Clinical Practice*, *18*, 746-752. doi: 10.1111/j.1365-2753.2011.01662.x

Appendix C

Demographic Information Sheet and Questionnaires

Information about you

1. **How old are you?**

2. **What is your gender? (please tick)**

   Male ☐   Female ☐

3. **What type of cleft do you have (please tick)?**
   Unilateral cleft lip (cleft on one side of the lip but not palate ☐

   Bilateral cleft lip (cleft on both sides of the lip but not palate) ☐

   Cleft of the soft palate (muscles at the back of the palate) ☐

   Cleft of the hard palate ☐

   Unilateral cleft lip and palate (one side of the lip and some/all of palate) ☐

   Bilateral cleft lip and palate (both sides of the lip and some/all of palate) ☐

   Submucous cleft palate ☐

   Non-cleft related speech difficulties ☐

4. **Do you have any other medical conditions? If yes, please write them below:**

This text box is where the unedited thesis included the following third

party copyrighted material:

Connor, K.M., & Davidson J.R.T. (2003). Development of a new

resilience scale: The Connor-Davidson Resilience Scale (CD-RISC).

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Pertschuk, M.J., & Whitaker, L.A. (1982). Social and psychological effects of craniofacial deformity and surgical reconstruction. *Clinics in Plastic Surgery, 9*, 297–306

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Beavers, R., & Hampson, R.B. (1990). *Successful families: Assessment and intervention*. New York: Norton.

This text box is where the unedited thesis included the following third party copyrighted material:

Beavers, R., & Hampson, R.B. (1990). *Successful families: Assessment and intervention*. New York: Norton.

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Cleft Lip and Palate Association (CLAPA) Advertisement

Study into resilience in young people (aged 12-16 years old) with a cleft lip and/or palate

Jenna Cuddy (Trainee Clinical Psychologist at the University of Liverpool) is conducting a research study into what helps young people (aged 12-16 years old) overcome or 'bounce back' from any challenges that growing up with a cleft may bring. It is hoped that increasing knowledge in this area may help strengthen the support provided to young people with a cleft in the future.

Jenna is looking for 12-16 year olds to complete some questionnaires on a secure website. The questionnaires will take up to 20 minutes to complete. In appreciation for their time, all young people will be given the opportunity to enter into a prize draw to win one of three high street vouchers (first prize - £30, second prize - £15, and third prize - £10).

Jenna's study has been reviewed and given a favourable opinion by a Research Ethics Committee. It has also been approved by the University of Liverpool Research Review Committee. All young people under the age of 16 will be required to indicate that their parent/guardian has consented to their participation in the study.

If you would like any further information you can go to the study website: Alternatively you can contact Jenna on jcuddy@liverpool.ac.uk.

Explanatory Note

Participant information leaflets were similar for all methods of recruitment (postal, clinic, and online via the Cleft Lip and Palate Association; CLAPA). Please note that due to word restrictions, only the participant information leaflets for postal recruitment are presented. However, for clarity, the main differences between the various participant information leaflets are listed below:

- For the recruitment of participants via CLAPA, participants were advised to make contact with CLAPA if the completion of questionnaires highlighted any distress. CLAPA would then be able to signpost families to their local cleft psychology service.

- For pragmatic reasons the main researcher did not offer the option of home visits for participants recruited via CLAPA.

- Participants who were recruited via CLAPA were not allocated a unique participant code. Participants were therefore not offered the opportunity to withdraw their data from the study within one month of the completion of study questionnaires as it would not be possible to identify their data.

- For clinic recruitment, potential participants were offered the opportunity to complete the questionnaires in: the clinic waiting area; in a private side room with the main researcher; at home and return hard copies of the questionnaires by post in a pre-paid envelope; at home and complete the questionnaires online; at home with the main researcher; or at the participant's GP practice.

ASSENT FORM FOR ONLINE COMPLETION OF QUESTIONNAIRES

Has somebody (e.g. a parent or guardian) explained this project to you?   Yes/No

Do you understand what this project is about?   Yes/No

Have you asked all the questions you want?   Yes/No

Have you had your questions answered in a way you understand?   Yes/No

Do you understand it is OK to stop taking part at any time?   Yes/No

Are you happy to take part?   Yes/No

If participants selected **NO** for any of the above statements the following message was displayed on the secure survey website:

Please discuss the study with your parent/guardian. If you need any more information, please ask your parent or guardian to contact me. Contact details are available in the information leaflet posted to your parent/guardian.

CONSENT FORM FOR ONLINE COMPLETION OF QUESTIONNAIRES

Have you read and understand the information sheet for the above study?   Yes/No

Have you asked all the questions you want?   Yes/No

Have you had your questions answered in a way you understand?   Yes/No

Do you understand it is OK to stop taking part at any time?   Yes/No

Are you happy to take part?   Yes/No

If participants selected **NO** for any of the above statements the following message was displayed on the secure survey website:

If you have any questions or would like any additional information, please ask the main researcher (Jenna Cuddy, Trainee Clinical Psychologist). Contact details are available in the information leaflet posted to you.

Protocol reference number: UoL001052
16 year old consent form for online completion of questionnaires
Version 1: 01 08

Appendix H

Data Screening to Test the Assumptions of Multiple Hierarchical Regression

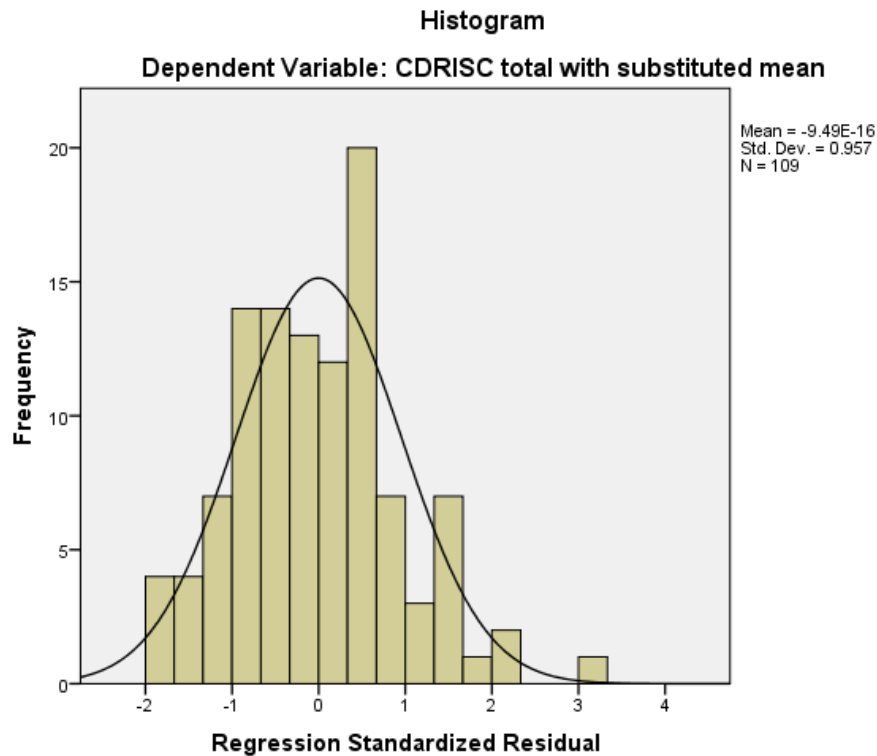*Figure 1.* Distribution of Scores on the CDRISC
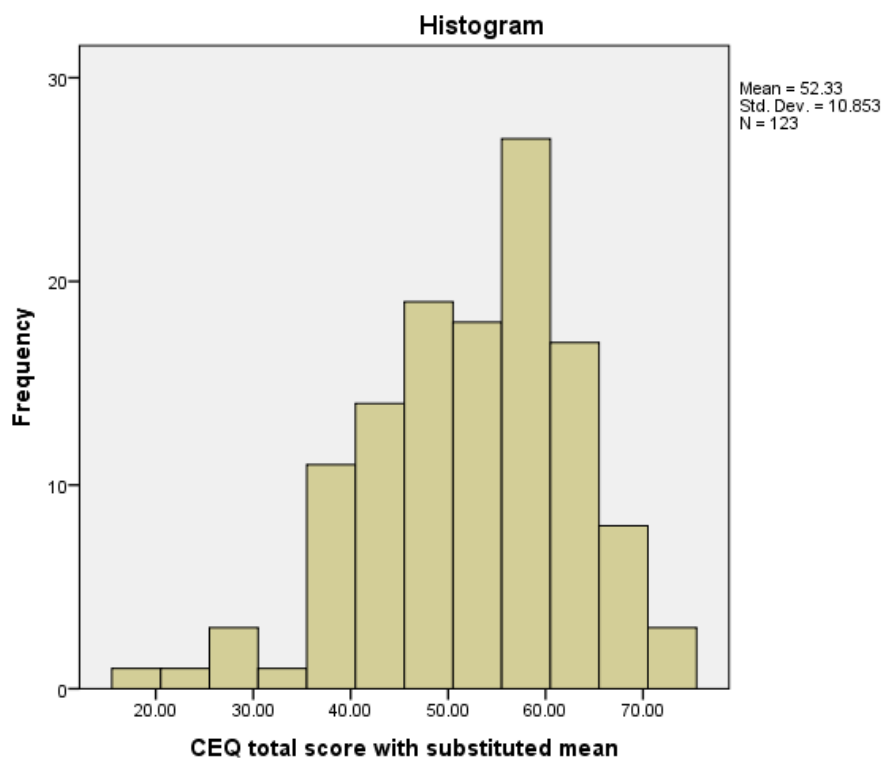

Figure 2. Distribution of Scores on the CEQ

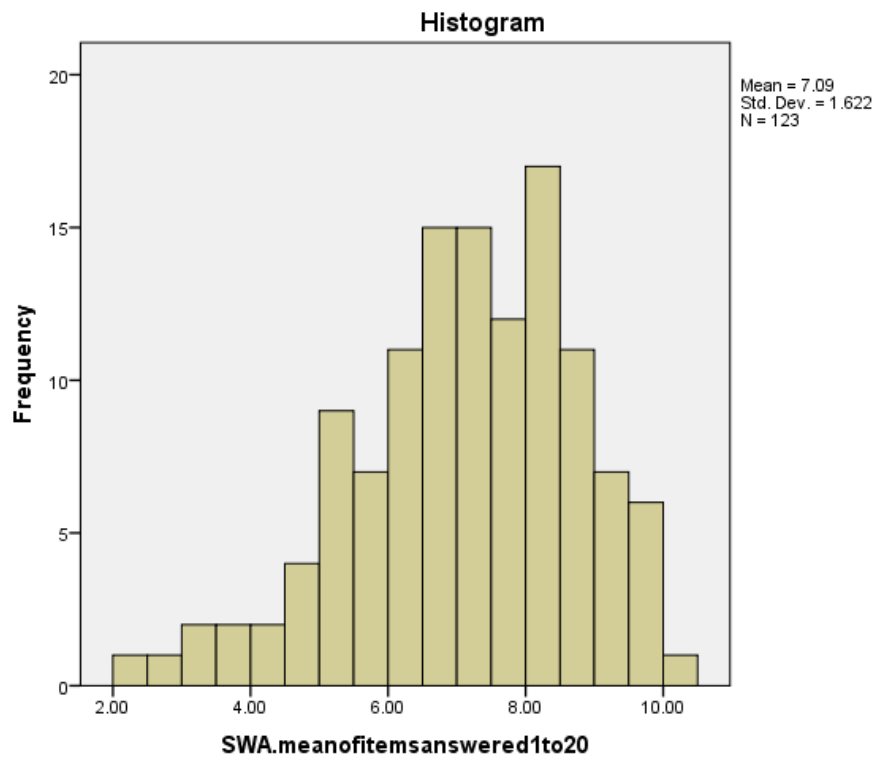*Figure 3*. Distribution of Scores on the SWA

Figure 4. Distribution of Scores on the Family Competence Subscale of the SFI-II
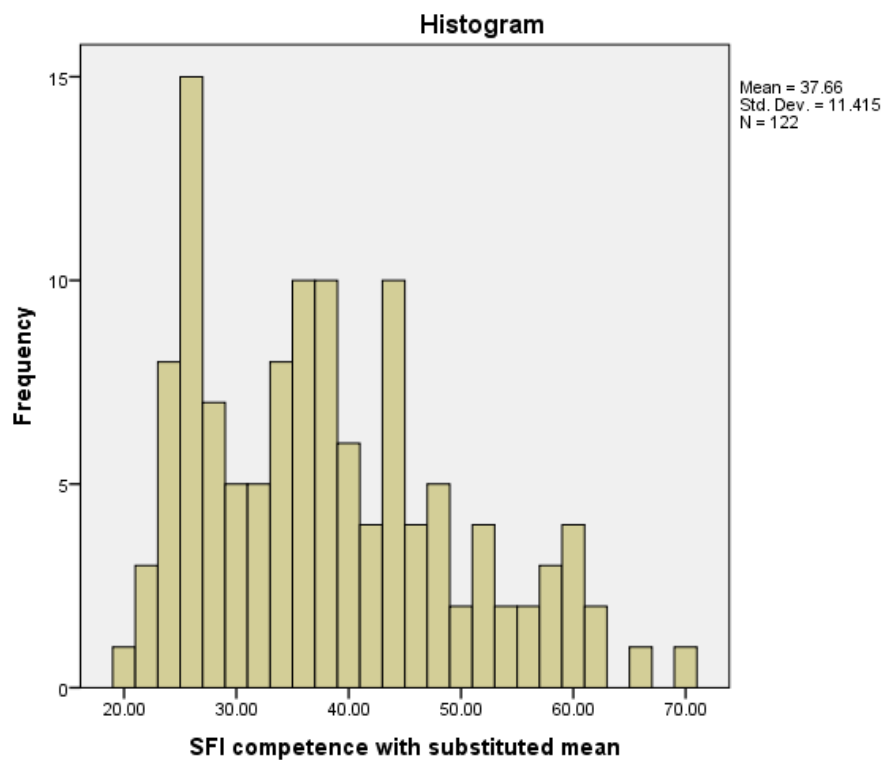
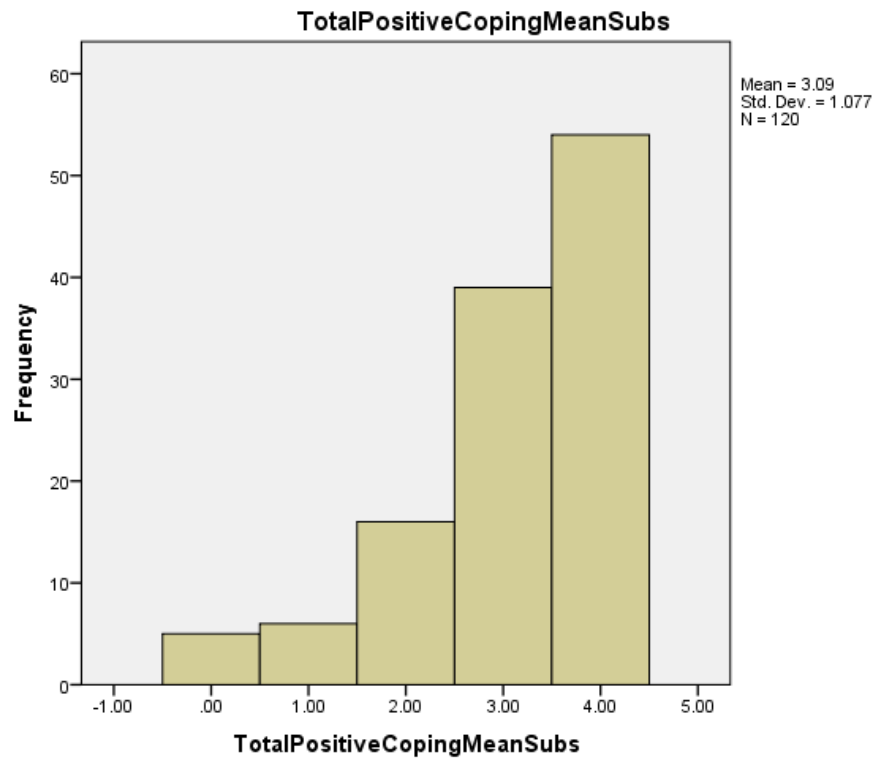*Figure 5.* Distribution of Positive Coping Strategy Use Scores on the KidCope

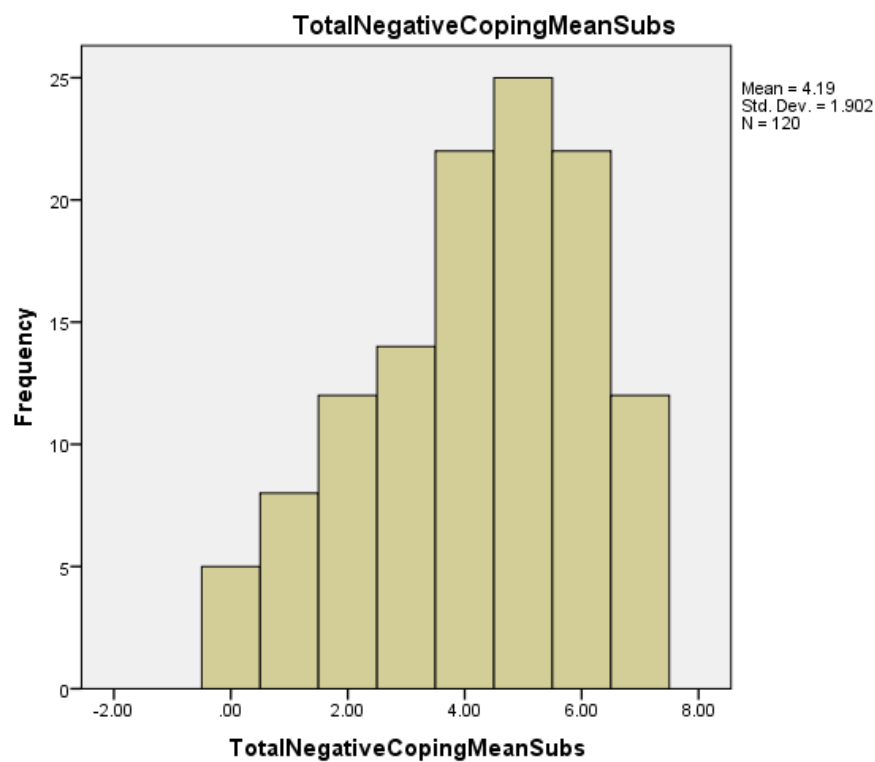*Figure 6.* Distribution of Negative Coping Strategy Use Scores on the KidCope

Table 1

Skewness and Kurtosis Z Scores

Variable	Skewness and kurtosis statistics	
	Skewness z score	Kurtosis z score
Resilience (CD-RISC)	0.82	0.82
Positive peer relationships & experiences (CEQ)	2.54	0.68
Satisfaction with appearance (SWA)	2.77	0.33
Family competence (SFI-II)	2.67	0.89
Positive coping (KidCope)	5.67	2.45
Negative coping (KidCope)	2.08	1.37

NB: Skewness and kurtosis scores with the accepted ranges of ±1.96 indicate normal distribution (Field, 2013).

Table 2

Results of the Kolmogorov-Smirnov Test of Normality

Variable	Kolmogorov-Smirnov		
	Statistic	df	Significance level
Resilience (CD-RISC)	.05	125	.20
Positive peer relationships & experiences (CEQ)	.09	123	.30
Satisfaction with appearance (SWA)	.06	124	.20
Family competence (SFI-II)	.10	122	.01
Positive coping (KidCope)	.25	120	.00
Negative coping (KidCope)	.16	120	.00

A non-significant result (p>.05) indicates normal distribution.

Table 3

Collinearity Statistics

	Collinearity Statistics	
Variable	Tolerance value	Variance inflation factor (VIF)
Visibility of Cleft	.87	1.15
Gender	.90	1.12
Age	.89	1.13
Presence of Additional Conditions	.81	1.24
Peer relationships and experiences (CEQ)	.58	1.72
Mean Satisfaction with Appearance (SWA)	.58	1.72
Family Competence (SFI-II)	.78	1.28
Positive Coping (KidCope)	.80	1.25
Negative Coping (KidCope)	.66	1.51

Tolerance scores below .10 and average VIF scores substantially greater than 1 indicate that multicollinearity may be present (Pallant, 2013).

Figure 7. Normal Probability Plot of Standardised Residuals for the CD-RISC
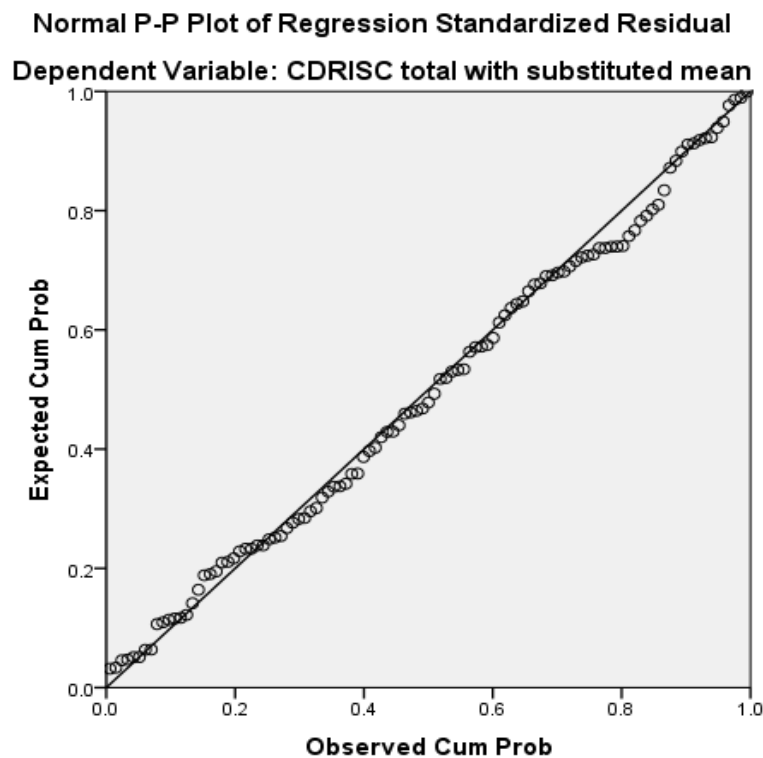
Figure 8. Scatterplot of Standardised Residuals for the CD-RISC

Scatterplot
Dependent Variable: CDRISC total with substituted mean