

# ATTACHMENT, BONDING, AND PARENTAL STRESS IN CHARGE SYNDROME

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Parents of 25 children with CHARGE syndrome, ages 12 to 50 months, completed measures of child attachment, parental bonding, and family stress. Twelve children were classified as securely and 13 as insecurely attached. The time it took to appear attached and parents to feel bonded were related, as were length of time to appear attached and strength of parental bonding. Visual impairment was related to an insecure attachment as well as parenting stress. Twelve parents had scores indicating significant stress. Parenting stress was related to problems with bonding, and having a challenging child was related to insecure attachment. Being able to hold the child and a shorter stay in the hospital after birth were related to more secure attachment.

*Keywords:* attachment, CHARGE syndrome, intellectual disability, mental retardation, parenting, psychiatric, stress

CHARGE syndrome is a rare genetic condition, generally estimated at 1:10,000 births,<sup>15</sup> involving a number of structural birth defects as well as multi-sensory impairments including deaf-blindness, language processing and communication difficulties, airway difficulties along with impaired ability to swallow food, and balance problems due to damaged or missing semi-circular canals in the inner ear. The specific features found in CHARGE syndrome were originally identified independently by Hall<sup>12</sup> and Hittner, *et al.*<sup>21</sup> in 1979. The acronym CHARGE was developed in 1981 by Pagon, *et al.*<sup>29</sup> based on common features of the syndrome: C—coloboma of the eye (missing part of iris and/or retina); H—heart defects; A—atresia of the choanae (bony or membranous blocking of nasal passage); R—retardation of growth and/or development; G—genitourinary anomalies; E—ear anomalies and/or deafness.

There is a wide variation in both the occurrence and severity of the features in CHARGE. This makes diagnosis and management a challenging task. Various diagnostic schemes have been proposed.<sup>5,28,29,36,44</sup> These schemes are based on features that are considered to be “major” or “essential” to the diagnosis. A gene for CHARGE was identified in 2004,<sup>45</sup> but testing is not yet widespread, and it is not known whether or not there may be additional genes involved. Therefore, the diagnosis still relies on clinical features.

Bowlby's<sup>6</sup> theory of attachment describes a relational construct to which both the caregiver and the child contribute during the first years of

life. Attachment theory emphasizes the importance of the affectional bond between the primary caregiver and the child. According to Shaver and Tancredy,<sup>34</sup> “Attachment is not the same as closeness, liking, or relationship.” It reflects an innate behavioral system leading to a child becoming emotionally attached to the primary caregiver. As a relational construct, both parent and child make contributions, and thus researchers have looked at both parenting variables and child variables for their influence on attachment.<sup>26</sup>

Quality of parenting as an influence on attachment is well researched.<sup>2,3,9,23,25,35,38</sup> Caregiver sensitivity, responsiveness, emotional availability, acceptance toward child, and predictability are all associated with the quality of the attachment relationship.<sup>26</sup> In addition, the quality of attachment has been found to be related to maternal self-esteem,<sup>39</sup> mental illness,<sup>33,37,41</sup> and stress in the family.<sup>4,43</sup>

There is less research on how child characteristics influence the attachment relationship. One child characteristic that has received considerable consideration is temperament; however, there continues to be a lack of agreement about the nature of its relationship to attachment, if any.<sup>40,42</sup>

Specific types of disabilities may make a difference in attachment classification.<sup>20</sup> For instance children with disabilities that involve deformities may be more likely to be ignored by their mothers, affecting the quality of interactions and thereby attachment.<sup>8,46</sup>

Child disability has been only rarely examined for its possible influence on attachment. A meta-analysis by van Ijzendoorn, *et al.*<sup>41</sup> found that maternal characteristics were more important to attachment than child problems such as deafness. Other research on children who were deaf also suggests that any differences in attachment between children who are deaf and those who are not may be attributed to parental attitudes toward deafness,<sup>13</sup> and not to the child.

One study has looked at attachment in blind and visually impaired infants.<sup>24</sup> While there were few differences for visually impaired infants, those who were totally blind were more frequently classified as insecure than normative samples.

Another study looking at child disability factors considered children with cerebral palsy.<sup>26</sup> Neither the presence of cerebral palsy nor the severity of the impairment were related to the attachment classification. However, child impairment was related to parent stress and as the severity of the impairment increased so did levels of stress. Also, the security of attachment was associated with lower levels of parent stress. A meta-analysis of 16 studies on autism and attachment found that children with autism were less often securely attached, but that this difference disappeared with higher intellectual development. The authors concluded that attachment may be a problem when autism and intellectual disability are both present.<sup>31</sup>

Hartshorne, Murray, and Scott<sup>18</sup> reported on a pilot study of 17 parents of children with CHARGE syndrome regarding their experiences with bonding and attachment. The respondents tended to fall into two different groups: those that bonded and felt their child attached early in the relationship, and those who felt it took several weeks or months to start to experience that closeness. Therefore, it may be difficult to generalize the experience of attachment and bonding in CHARGE; some parents bond and their child attaches quickly and for others it takes more time.

Capuzzi<sup>7</sup> attributes problems with attachment for infants with disabilities to the child's difficulty sending clear cues to the mother and responding to the mother in ways the mother can understand. In addition, she believes the stress experienced by the parents having a child with disabilities may make them less sensitive and attentive to their child's cues.

The current study explored attachment, bonding, and parental stress in families with

children diagnosed with CHARGE syndrome. Five questions addressed the nature of the attachment and bonding experience of parents of a child with CHARGE. First, what is the attachment and bonding experience for parents of children with CHARGE? Is the severity of the child's condition and sensory impairments associated with attachment and bonding? Is the amount of stress parents experience associated with their experience with attachment and bonding? Are these stress levels related to the severity of the child's condition and sensory impairments? Do certain early experiences due to the child's health impact attachment, bonding, or family stress?

## METHOD

### Participants

This research was approved by the Institutional Review Board at the authors' university and by the Research Committee of the CHARGE Syndrome Foundation. Thirty parents of children ages 12 months through 54 months with CHARGE syndrome were contacted. Of the thirty, one was returned for an incorrect address, two of the children were over the 54 month age limit, one respondent scored low on the PSI-SF's defensive responding scale suggesting that the results were not valid, and one was not returned. Thus, analyses were based on the results of 25 children. The participants in this study were all from the United States and were contacted through the CHARGE Syndrome Foundation.

### MEASURES

The CHARGE History Questionnaire (based on Salem-Hartshorne and Jacob<sup>32</sup>) was used to gather demographic and basic information on the child's CHARGE characteristics.

The Revised Maternal Attachment Inventory (R-MAI),<sup>27</sup> a 26-item measure, was used to assess parental bonding between the parent and the child. Bonding describes the feelings of the parent toward the child. The R-MAI was revised from a previous version in order to increase reliability and validity. The author reports acceptable concurrent validity and internal consistency reliability.

The Parenting Stress Index Short Form (PSI-SF)<sup>1</sup> is a 36-item self-report measure that was used to identify parenting-related stressors. The items were developed from clinical experience and research on infant development, parent-child interaction, attachment, child abuse and neglect,

child psychopathology, child bearing practice, and stress. The PSI-SF includes three subscales. Parental Distress reflects parenting difficulties experienced by the parent due to personal factors. Parent-Child Dysfunctional Interaction indicates the parent's perception that their child and interactions with their child are problematic and do not meet expectations. Difficult Child measures behavioral characteristics of the child that make them challenging to parent.

Most investigations of child attachment utilize the Ainsworth Strange Situation,<sup>3</sup> a laboratory procedure in which parent and child interaction is observed in specific situations of separation and reunion in order to classify the child into one of three attachment categories, insecure-avoidant, insecure-ambivalent, and secure. Because of the survey nature of the present study, observation utilizing this technique was not possible, and no published substitute was found. In addition, concerns have been expressed regarding the applicability of the Strange Situation to disabled populations.<sup>11,22</sup> Therefore, in order to assess attachment styles, the Strange Simulation Questionnaire (SSQ) was developed by the authors based on the laboratory technique. (Appendix A) This four-item measure gathered parent assessment of the child's reaction to the caregiver's departure and return and reaction to new caregivers. The questions asked about the child's reaction to being taken to a new place, being left with an alternate caregiver, experiencing their primary caregiver returning to them, and being left with a new caregiver. Each item has three response choices, each purporting to represent one of three attachment classifications: secure, insecure-avoidant, insecure-ambivalent.

The CHARGE Attachment Questionnaire (CAQ) is a six-item measure, revised from a pilot study,<sup>18</sup> used to gather information about the experience of attachment and bonding of children with CHARGE, including how it compared to that of children without CHARGE within the same family.

#### ANALYSIS

All data was analyzed using SPSS for windows version 14. Analyses that involved attachment classification from the SSQ were conducted using t-tests between secure and insecure groups. This was the same for looking at differences based on sensory impairment (blind, deaf, deaf-blind), except where these were analyzed with the SSQ when the phi coefficient was calculated. Phi was also used to look at various early childhood

experiences with dichotomous variables. Most other analyses utilized Pearson correlation coefficients to look at relationships with bonding on the R-MAI, parenting stress on the PSI-SF, and time on the CAQ.

## RESULTS

The measures were completed by twenty-three mothers and one father (one did not indicate). The children ranged in age from twelve months to fifty months. The children were 46% male and 54% female. The CHARGE characteristics of this sample are given in Table 1. The percentages are similar to those found in other studies.<sup>16,17</sup>

### The Attachment and Bonding Experience

#### STRANGE SIMULATION QUESTIONNAIRE (SSQ)

There were four questions on this measure designed to simulate the traditional strange situation method of identifying attachment styles. The questions related to alternate and new caregivers proved to be difficult for some parents to answer as, given their child's medical problems, in many cases they did not use alternate or new caregivers. The child's reactions were scored as secure, insecure-avoidant, and insecure-ambivalent. Only one child was scored on a single item as insecure-avoidant, and therefore responses were coded as secure or insecure. The instrument was scored by selecting the style that was most frequently given for the four questions. In cases of an equal number they were scored as insecure, on the assumption that there might be a positive bias. Twelve children were classified as securely attached and 13 as insecure.

#### THE REVISED MATERNAL ATTACHMENT INVENTORY (R-MAI)

The R-MAI measures feelings of security and attachment felt by the mother or primary caregiver toward their child. A reliability test of the measure conducted on this data obtained a Cronbach's alpha of .89 which is similar to that of the original 31-item Maternal Attachment Inventory (alpha = .85).<sup>27</sup> The mean score was 98.32 out of a possible 104 points with a range of 78 to 104, and a standard deviation of 6.71. Higher scores reflect a greater sense of bonding. These results can be compared with a study by Damato<sup>10</sup> who administered the R-MAI to parents of two groups of postnatal twins, and obtained means of 98.5 and 98.9, and standard deviations of 6.7 and 6.8 respectively. Thus the parents in this study

<b>TABLE 1. MEDICAL PROBLEMS IN THE SAMPLE OF 25 CHILDREN</b>	
<b>CONDITION</b>	<b>PERCENTAGE</b>
Delayed Motor Milestones	96
Vestibular Problems	96
Coloboma	88
Swallowing Problems	88
Heart Defect	88
Sensorineural Hearing Loss	84
Characteristic CHARGE External Ear	84
Growth Deficiency	60
Choanal Atresia or Stenosis	60
Frequent Middle Ear Infections	52
Genital Hypoplasia	48
Renal Problems	48
Tactile Defensiveness	40
Facial Palsy	36
Tracheoesophageal Fistula	28
Anosmia	20
Spine Anomalies	16
Cleft Lip or Palate	12
Hand Anomalies	12
Abdominal Defects	8

had bonding scores very similar to those in the Damato study, suggesting that the experience of bonding with a child with CHARGE is not very different from the experience of bonding that parents have with other children.

#### CHARGE ATTACHMENT QUESTIONNAIRE (CAQ)

There were six questions on the CAQ. Question one asked when the parent felt bonded with the child. As seen in Table 2, seven choices were provided ranging from after conception to still not feeling bonded. The modal response was after conception but there was considerable variability in response.

The second question asked whether the bonding experience was different with the child with CHARGE as compared with other children the parent may have. Fourteen parents answered this question, with seven saying it was the same,

and seven saying it was different, again quite variable. Differences identified included: the experience of diagnosis which involved anger, frustration, and worry; the medical complications; the lack of physical contact; and the longer time it took to bond.

The third question asked if the parent experienced something that made them realize they felt bonded. Three parents did not respond, but 10 indicated that something occurred while 12 did not. These occurrences included the child crying when the parents left, when the parent learned of her pregnancy, when the parent was able to touch the child, the child's smile and excitement, the birth experience, calming the child when upset, and the fear of the child dying.

The fourth question asked when their child began to demonstrate that they felt attached to the parent. Four choices were provided ranging

**TABLE 2. BONDING AND ATTACHMENT OF PARENTS**

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<i>When did parent feel bonded to child?</i>	
<b>Time</b>	<b>Percent of 25 Parents</b>
Conception	6 (24%)
At Birth	4 (16%)
First Time Held Child	5 (20%)
When Child Came Home From Hospital	4 (16%)
Several Weeks After Birth	1 (4%)
Several Months After Birth	4 (16%)
Still Feel Bond is Not Well Established	1 (4%)
<i>When did child demonstrate attachment toward parent?</i>	
<b>Time</b>	<b>Percent of 23 Parents</b>
Shortly After Birth	8 (34.7%)
Several Weeks After Birth	5 (21.7%)
Several Months After Birth	7 (30.4%)
Attachment is Not Well Established	3 (13.6%)

from shortly after birth to still not well established. The modal score was shortly after birth, but again, there was considerable variability. (Table 2)

Question five asked if this experience of attachment was different from their other children. Twelve parents responded, with nine saying it was different, and three that it was not. Differences cited included the experience of diagnosis including fear, worry, and stress; the child being tactilely defensive which included crying when held or hugged; the long length of hospitalization after birth; and the number of people who were in contact with the child. Two parents mentioned that their child with CHARGE was more affectionate than their other children.

The final question asked whether the parent experienced anything that made them realize their child felt attached. Sixteen parents responded, stating they experienced something, including the excitement the child showed exclusively to them, the reaction they received when they left and returned, their ability to calm their child, the change in the child's heart rate when held, the

smiles and excitement the child gave them, and the child sleeping calmly in their arms.

In order to explore the relationship between child attachment style and the parents' bonding, differences between the two groups of children as identified by the SSQ on the R-MAI were analyzed. The parents' R-MAI scores did not significantly differ for the two groups of children,  $t(23)=-1.589$ ,  $p=.126$ . Thus parental bonding and the child's attachment classification were independent of each other.

From the CAQ, the length of time it took for the child to appear to be attached was correlated with the R-MAI scores. The result was significant ( $r=-.567$ ,  $p=.005$ ). This suggests that how long it takes for the child to demonstrate his or her attachment with the parent may be related to the feelings of bonding experienced by the parent. So while attachment classification was not found to be related to degree of bonding, the length of time it took to attach was.

Correlation coefficients were computed among the responses from questions one and four on the

**TABLE 3. PARENTING STRESS INDEX-SHORT F: MEAN RAW SCORE AND SD; RANGE; MEAN PERCENTILE SCORE AND SD; AND NUMBER OF PARENTS SCORING ABOVE CRITICAL NUMBER OF 85 FOR 25 PARENTS**

Subscale	Mean Raw (SD)	Range	Mean % (SD)	Score >85
Overall PSI-SF	90.26 (20.65)	60-147	79.86 (20.27)	12
Parental Distress	33.66 (8.93)	17-48	85.5 (26.22)	12
Parent-Child Dysfunctional Interaction	25.62 (8.16)	12-46	71.52 (26.65)	10
Difficult Child	30.98 (7.87)	20-56	68.62 (22.8)	10

CAQ and the responses on the R-MAI. The correlation between the time in which the mother felt bonded (CAQ question 1) and the scores on the R-MAI was not significant ( $r = -.34$ ,  $p = .10$ ). This suggests that the time that the parent initially felt bonded is not associated with the present feelings the parent has of bonding with his/her child. However, the length of time it took the child to attach and the parent to bond, based on the CAQ questions 1 and 2, were significantly correlated ( $r = .539$ ,  $p = .008$ ). So while length of time to bonding was not associated with the quality of the bond, it was associated with how long it took for the child to seem attached to the parent.

Looking at the secure and insecure attachment groups on length of time to bond and to attach, no significant differences were found on either how long it took to feel bonded,  $t(23) = 1.54$ ,  $p = .136$  or how long it took for the child to appear attached,  $t(21) = -.147$ ,  $p = .88$ .

#### **Severity of the CHARGE and Attachment and Bonding**

Because CHARGE has a highly variable phenotype, it is difficult to identify any particular set of anomalies as being most characteristic. Some researchers have utilized an impact measure which is the sum of those characteristics listed in Table 1 as a rough measure of how impacted the child is by the syndrome.<sup>16,32</sup> Total scores on this measure were compared between the two attachment styles as measured by the SSQ. There was not a significant difference,  $t(23) = 1.664$ ,  $p = .11$ . This impact measure was also correlated with scores on the R-MAI. The resulting correlation was also not significant,  $r = .11$ .

Sensory impairments may be hypothesized to interfere with normal attachment, and so those children who were deaf, visually impaired, or deaf-

blind were examined. Deaf was defined as having no better than moderate hearing impairment in the best ear. Twelve children were classified as deaf. No significant association was found between being deaf and classification on the SSQ,  $\phi = .08$ . Nor was there a difference in R-MAI scores for those children who were or were not deaf,  $t(22) = .966$ ,  $p = .34$ .

Visual impairment was defined as having no better than moderate visual impairment in the best eye. Seven children were classified as visually impaired. Of the 11 children who were securely attached, only one was visually impaired, while for the 13 who were not securely attached, six were visually impaired. This association was significant,  $\phi = .41$ ,  $p = .047$ . However, those who were visually impaired did not have significantly different R-MAI scores,  $t(22) = 1.61$ ,  $p = .12$ .

CHARGE is a major cause of deaf-blindness. However, by defining deaf-blind as both deaf and visually impaired according to the definitions adopted in this study, only three children were classified as deaf-blind. However, all three of these were classified as having insecure attachment. The average score on the R-MAI for these three children was not significantly different from the non-deaf-blind children,  $t(22) = .427$ ,  $p = .67$ .

#### **The Impact of Parenting Stress**

##### PARENTING STRESS INDEX

The PSI-SF means and standard deviations are shown in Table 3. The mean raw total score was 90.26 out of a possible 180 points, with a standard deviation of 20.65, a range of 60 to 147, and a percentile score of 79.86%. Twelve parents' scores were higher than the 85th percentile which is significant stress according to the manual.

**TABLE 4. CORRELATIONS BETWEEN R-MAI AND PSI-SF**

	<b>R-MAI</b>
Overall PSI-SF	-.532**
Parental Distress	-.344
Parent-Child Dysfunctional Interaction	-.498*
Difficult Child	-.490*
** $p < .01$ * $p < .05$	

Cronbach's coefficient alpha on this sample was .92, similar to the .90 found by Roggman, *et al.*<sup>30</sup>

On the Parental Distress subscale the mean was 33.66 out of a possible 60 points, with a standard deviation of 8.93, a range of 17 to 48, and a percentile score of 85.5%. Twelve parents responded higher than the 85th percentile on the parental distress domain. The mean on the Parent-Child Dysfunctional Interaction subscale was 25.62 out of a possible 60 points, with a standard deviation of 8.159, a range of 12 to 46, and a percentile score of 83%. Ten parents had scores higher than the 85th percentile. On the Difficult Child subscale the mean score was 30.98 out of a possible 60 points, with a standard deviation of 7.82, a range of 20 to 56, and a percentile score of 80%. Ten parents scored above the 85th percentile.

To assess whether the attachment style of the child (SSQ) was associated with greater or lesser parental stress, t-tests were conducted utilizing scores on the PSI-SF. The t-test was not significant for total stress,  $t(23) = 1.232$ ,  $p = .230$ . When looking at the subscale scores on the PSI-SF, statistically significant results were found for the Difficult Child subscale of the PSI-SF,  $t(23) = 2.44$ ,  $p = .023$ . This suggests that child attachment classification may be related to the stress the parent is feeling from the behavioral characteristics of their child. Scores for the other two domains were not significant with attachment classification.

To look at family stress and parental bonding, scores on the PSI-SF were correlated with the R-MAI. (Table 4) Total PSI-SF scores were highly related, as were scores on two of the subscales, Difficult Child and Parent-Child Dysfunctional Interaction. The correlations suggest that the

higher the stress, the less the experience of bonding.

#### **Stress and the Severity of the Child's CHARGE**

The total CHARGE impact measure was not significantly related to parenting stress, nor was being deaf. However, being blind was related to total stress,  $t(22) = 2.29$ ,  $p = .032$  (equal variances not assumed); however, there were no significant differences for the sub-domains.

#### **Early Experiences and Attachment, Bonding, and Family Stress**

Parents were asked whether they were able to breastfeed their child, how long it was before they could hold their child, how long the child was hospitalized after birth, and how many hospitalizations and surgeries the child had experienced. (Table 5)

Table 5 includes the average scores on the R-MAI and PSI-SF for each category of these five variables. A look at the data does not show any trends, except for breastfeeding, where there is a trend for parenting stress to decrease with successful breastfeeding. However, none of these breakdowns was statistically significant.

Contingency table analysis was used to examine whether these variables are related to attachment classification. For these analyses, breastfeeding was collapsed to two cells by combining the two "yes" categories, holding was collapsed to two cells by combining the second two categories, length of stay after birth and number of surgeries were collapsed to three cells maintaining the middle category, and number of hospitalizations was collapsed to two categories of equal size.

**TABLE 5. EARLY EXPERIENCES REPORTED BY 25 PARENTS AND SCORES, MEAN AND SD FOR R-MAI AND FSI-SF**

	N (%)	R-MAI Mean (SD)	FSI-SF Mean (SD)
<i>Breastfeeding</i>			
Yes - Successfully	1 (4%)	102	79
Yes - Stopped Early	3 (12%)	100.33 (6.35)	88.33 (13.58)
No - Child Could Not Breastfeed	21 (84%)	97.86 (6.97)	91.07 (22.04)
<i>Age parents could hold the child</i>			
At Birth	11 (44%)	96.64 (5.92)	90.09 (18.34)
Within Week	6 (24%)	98.83 (4.45)	88.25 (13.88)
More than a Week	8 (32%)	100.25 (9.02)	92.00 (28.93)
<i>Length of hospital stay after birth</i>			
Came Home with Mother	4 (16.6%)	96.25 (6.18)	95.00 (25.07)
Less than 2 Weeks	2 (8.3%)	100.5 (4.95)	71.5 (4.95)
2-6 Weeks	8 (33.3%)	99.25 (4.46)	86.38 (13.45)
7-12 Weeks	5 (20.8%)	97.80 (7.60)	84.60 (17.64)
>12 Weeks	5 (20.8%)	97.00 (11.14)	99.3 (28.54)
<i>Number of surgeries</i>			
None	3 (12.5%)	101.00 (3.61)	86.67 (31.21)
1-4	6 (25%)	100.33 (3.72)	84.00 (11.68)
5-8	9 (37.5%)	93.33 (8.14)	94.11 (25.91)
9-12	5 (20.8%)	101 (4.64)	89.50 (11.39)
>13	1 (4.2%)	104	75
<i>Number of hospitalizations</i>			
None	2 (8.3%)	97.5 (.71)	100.00 (29.70)
1-4	10 (41.6%)	95.80 (9.14)	91.60 (25.12)
5-8	10 (41.6%)	101.10 (3.73)	83.95 (14.40)
9-12	2 (8.3%)	95.00 (2.83)	89.00 (11.31)

Two of these analyses were significant. The age parents could hold their child and attachment classification was found to be significantly related, Pearson chi square (1, n=25) = 4.81,  $p = .028$ ,  $\Phi = .439$ . Of those who could hold early, 73% were securely attached, while for those who could only hold later, 29% were securely attached.

The length of time the child remained in the hospital after birth and attachment classification was found to be significantly related, Pearson chi square (2, n=24) = 9.50,  $p = .009$ ,  $\Phi = .629$ . All six of those children who went home in less than two weeks were securely attached, while 25% of those between two and six weeks, and 30% of those who came home later than six weeks were securely attached.

## DISCUSSION

As with many characteristics of CHARGE syndrome, the experience of child attachment and parental bonding was found to be quite variable. It is interesting that the bonding scores were nearly identical to the only normative data published in the literature. Thus, the parents of children with CHARGE did not demonstrate bonding differently from the parents of twins.<sup>10</sup> There was considerable variability for when parents felt bonded and when they believed their child felt attached. While the experience of bonding did not vary by attachment classification, the longer it took for the child to demonstrate attachment the lower the bonding score.

Furthermore, there was an association between the length of time it took the child to attach and how long it took the parent to bond. These findings suggest that it is important to facilitate the attachment and bonding experiences as early as possible.

Because there are so many anomalies associated with CHARGE syndrome, it would be statistically challenging with a small sample size to examine all of them in relation to attachment, bonding, and stress. However, a sum of the number of anomalies was not found to be related to any of the measures.

Having CHARGE and being deaf-blind has been found to be related to more challenging behaviors,<sup>16</sup> autistic-like behaviors,<sup>17</sup> and executive dysfunction.<sup>19</sup> Thus it was of interest to see whether sensory impairments are related to attachment, bonding, and stress. Because only three of the children were deaf-blind, this study focused more on being deaf or blind. Being deaf was not found to be associated; however, being blind was related to attachment classification, with more blind being insecurely attached, consistent with the findings of Macrae.<sup>24</sup> Being blind was also related to increased parenting stress. The possible directions for these relationships would be of interest in future research.

It is likely that having a child with CHARGE is stressful for parents, and indeed nearly half of these parents had total scores in the "significant stress" range. This was true for the total stress score as well as the three sub-domains. Parenting stress is highly likely to affect attachment and bonding, and while total stress was not related to attachment classification, scores on the difficult child domain were. This subscale measures things like difficult temperament, demanding behavior, and for very young children problems in self-regulatory processes. Self-regulation is often related to executive dysfunction which has been found to be problematic in CHARGE.<sup>19</sup> Stress was also found to be related to the experience of bonding on the part of the parent. Thus high

parenting stress, which seems to be common in CHARGE families, is related to problems with secure attachment and parent bonding.

The five early experiences, breastfeeding, holding the child, length of time in hospital after birth, number of surgeries and number of hospitalizations overall, did not seem highly related to attachment, bonding, and parenting stress. Breastfeeding showed a clear trend toward being associated with parenting stress, but did not achieve significance, in part perhaps because so few parents were able to breastfeed. However, being able to hold the child early on, and a shorter length of stay in the hospital after birth were associated with a more secure attachment relationship. More research to identify how certain early experiences are related to attachment style is warranted.

It is evident from this study that in many cases children with CHARGE become securely attached and parents develop a positive bond with their child. While there is considerable parenting stress associated with having a child with CHARGE, some parents do quite well, and the literature suggests that most families make a reasonable adjustment.<sup>14</sup> However, the difficulties found in this study suggest that more research is needed to better understand the early variables that may impact on attachment, bonding, and stress.

This study made use of a novel strategy for measuring attachment. Because children with CHARGE are not found in concentrated areas, it is not possible to bring many of them into a clinic in order to observe attachment style. However, the SSQ relies on parent report, which could easily be influenced by parent stress and bonding among other variables. While this measure did produce some interesting results in the present study, it would be helpful to validate the measure in a clinic setting.

Having a child with CHARGE syndrome is likely to be a challenging experience for parents. Early interventionists should be alert to parenting stress and problems with attachment and bonding as they provide support to these families.

**APPENDIX A****Strange Simulation Questionnaire**

To be filled out by the primary caregiver in reference to your child with CHARGE syndrome. Please select the **one** answer that best describes your son/daughter.

Q1. When I take my child to a new place my child \_\_\_\_\_.

- A. Acts “clingy” towards me at first but then freely explores the environment, coming back to me occasionally for reunions.
- B. Acts “clingy” towards me, is uncomfortable, and will not leave my side.
- C. Doesn’t seem to mind being in a new environment and freely explores without returning to me.

Q2. When I leave my child with an alternate caregiver my child \_\_\_\_\_.

- A. Is calm and does not seem to mind being left with others, my child acts indifferent.
- B. Is upset at first and caregivers have told me that my child calms down after I leave.
- C. Is upset when I leave and caregivers have mentioned that he/she remained upset throughout my absence and/or would not interact with others.

Q3. When I return from being away my child \_\_\_\_\_.

- A. Acts as if he/she is angry with me.
- B. Acts as if he/she is excited to see me.
- C. Acts as if he/she is indifferent.

Q4. When I leave my child with a new caregiver my child \_\_\_\_\_.

- A. Acts “clingy” and/or upset with me before I leave but the caregiver has told me that they warmed up easily after I left.
- B. Acts “clingy” and/or upset towards me while I leave and the caregiver has told me that they remained upset while I was gone and wouldn’t warm up to them.
- C. Doesn’t seem to mind being left with the new caregiver.

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