The effect of cleft lip and palate, and the timing of lip repair on mother–infant interactions and infant development

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Background: Children with cleft lip and palate are at risk for psychological problems. Difficulties in mother–child interactions may be relevant, and could be affected by the timing of lip repair. Method: We assessed cognitive development, behaviour problems, and attachment in 94 infants with cleft lip (with and without cleft palate) and 96 non-affected control infants at 18 months; mother–infant interactions were assessed at two, six and 12 months. Index infants received either ‘early’, neonatal, lip repair, or ‘late’ repair (3–4 months). Results: Index infants did not differ from controls on measures of behaviour problems or attachment, regardless of timing of lip repair; however, infants having late lip repair performed worse on the Bayley Scales of Mental Development; the cognitive development of early repair infants was not impaired. Difficulties in early mother–infant interactions mediated the effects of late lip repair on infant cognitive outcome. Conclusions: Early interaction difficulties between mothers and infants having late repair of cleft lip are associated with poor cognitive functioning at 18 months. Interventions to facilitate mother–infant interactions prior to surgical lip repair should be explored. Keywords: Cleft lip and palate, mother–infant interactions, cognitive development, attachment, behaviour problems, infancy, intelligence, sensitive period. Abbreviations: MDI: Mental Development Index.

Psychological development of children with cleft lip and palate

Cleft lip and palate affects one in every 650 infants (Derijcke, Eerens, & Carels, 1996). Studies of affected children have often been subject to methodological problems (e.g., small, heterogeneous samples, lack of normal controls); nevertheless, findings concerning psychological effects of clefts have been broadly consistent. In the school years, affected children are at raised risk for both socio-emotional and cognitive difficulties (Millard & Richman, 2001). In infancy, socio-emotional problems, as measured by infant–mother attachment, are not so evident (Wasserman, Lennon, Allen, & Shilansky, 1987; Koomen & Hoeksma, 1993; Speltz, Endriga, Fisher, & Mason, 1997; Maris, Endriga, Speltz, Jones, & DeKlyen, 2000), although cognitive deficits do appear to be a particular risk (Starr, Chinsky, Canter, & Meier, 1977; Jocelyn, Penko, & Rode, 1996; Kapp-Simon & Krueckeberg, 2000; Speltz et al., 2000).

The role of parent–infant interactions

An important question is the extent to which the problems of infants with clefts may be explained by difficulties in parent–infant interactions, since these predict poor child functioning (Eshel, Daelmans, de Mello, & Martins, 2006). Giving birth to an infant with a cleft is often followed by shock and distress (Bradbury & Hewison, 1994), and this may affect parent–infant interactions. Previous studies of mother–infant interactions in cleft samples have generally been conducted before lip repair. While caution is required in considering previous findings, as raters were not blind to cleft status, impairments in maternal responsiveness have been reported (Field & Vega-Lahr, 1984; Wasserman et al., 1987; Speltz, Goodell, Endriga, & Clarren, 1994). Notably, both Wasserman and Allen (1985) and Speltz and colleagues (2000) found that observed maternal interaction difficulties predicted lower IQ in affected children.

The timing of lip repair

One factor that may affect parent–infant interactions in the context of infant cleft lip is the timing of surgical repair, since this effects dramatic cosmetic change (Slade, Emerson, & Freedlander, 1999). Although repair is generally performed around 3–4 months postpartum in the UK, in the past two decades some centres have conducted neonatal repair. Surgical outcome appears unaffected by this difference in timing (Goodacre, Hentges, Moss, Short, & Murray, 2004); however, the psychological consequences of early vs. later lip repair may differ.
On the one hand, late repair may be advantageous because parents have time to adjust to the infant's condition and plan for surgery (Munro, 1995), and parent–infant contacts immediately after the birth are not disrupted by the infant's removal for surgery and post-operative recovery. On the other hand, in favour of early repair is the possible benefit in terms of parent–infant interactions in the initial months (Bradbury & Hewison 1994; Munro, 1995), particularly since these involve face-to-face play, where parents of infants having early lip repair may find it easier to respond to infant social cues. In turn, any enhanced responsiveness in parents of early repair infants may have benefits for infant development. The only study examining this issue of which we are aware found no differences in the psychological impact of neonatal vs. late lip repair (Slade et al., 1999). However, the sample was small, and self-report, rather than observational measures, was used to assess maternal responses to the infant.

Aims of the current study

We aimed, first, to assess the effects of cleft lip on infant cognitive and socio-emotional development at 18 months. Second, we aimed to assess the role of social interactions in the development of infants with clefts, and in particular, to determine whether interaction difficulties might mediate any impact of the cleft on infant outcome at 18 months. We therefore assessed interactions at two, six and 12 months, and examined their relationship to infant outcome, as well as cleft status. Finally, we aimed to assess the effects of the timing of surgical lip repair; thus, two cleft groups were recruited: one where infants had neonatal lip surgery – the ‘early’ repair group; and a second where surgery was performed at 3–4 months postpartum – the ‘late’ repair group. We also recruited a normal control group.

Given that previous work has been conducted with infants where the cleft has been un repaired through the first few postpartum months, we expected results for our late repair group to be in line with those in the literature, i.e., as showing poor cognitive, but not poor socio-emotional functioning. Moreover, and consistent with research in both normal and cleft populations, we expected the poor cognitive outcome for late repair infants to be accounted for by early parent–infant interaction difficulties. For infants having early repair, by contrast, where there may be benefits in terms of early mother–infant interactions, we did not expect such adverse outcome. Our specific hypotheses were as follows:

1. Infants having late repair will not differ from controls in their socio-emotional functioning, but will have poorer cognitive outcome than control infants.
2. Cognitive deficits in late repair group infants will be mediated by difficulties in mother–infant interactions before surgical repair.
3. There will be no difference in cognitive or socio-emotional functioning between infants having early cleft repair and control infants.

In assessing the impact of cleft lip on mother–infant interactions and infant development, a number of other factors require consideration. Thus, infant sex, family social class and maternal depression are all potentially important (Murray & Cooper, 2003). Furthermore, when considering results for the two index groups, a number of cleft-specific variables need to be taken into account: these include the severity of the cleft, i.e., whether cleft palate is also present, and the degree of visible deformity (Langlois, Ritter, Casey, & Swain, 1995). For later infant outcome, feeding method may be important, as well as hearing impairment and the experience of additional surgical procedures. In comparing the study groups, we therefore took these variables into account.

Method

Sample

Index infants were recruited from four NHS regional centres for Cleft Lip and Palate Surgery. Two performed late lip repair (Radcliffe Infirmary, Oxford, and Alder Hey, Liverpool), and two, early repair (Stoke Mandeville, Aylesbury, and St George's, London). Infants were admitted to centres by place of birth: those eligible for the study had isolated clefts of the lip (with or without cleft palate). Controls were recruited from maternity hospitals in the same regions (Royal Berkshire Hospital, Reading, Women’s Hospital, Liverpool). Exclusion criteria for all infants concerned factors having independent effects on infant development, i.e., prematurity (<36 weeks gestation), low birthweight (<2.5 kg) or presence of other syndromes (e.g., oro-facial-digital syndromes, heart problems). In addition, if parents of index infants had elected to attend a centre other than their local one, they would not have been considered eligible for recruitment because of the self-selection involved: in fact, this did not arise. One hundred and forty-seven index infants were identified for recruitment by surgical staff when referred to the surgical team, prior to surgery. Parents were told that we were investigating the effects of cleft lip on parent–infant relationships and infant development: 110 (75%) consented, 28 (19%) refused, 4 (3%) could not be contacted and 5 (3%) were not approached. After each index infant was recruited, a potential control was selected by identifying an infant of the same sex whose time of birth was closest to that of the index infant, and, as far as possible, of the same birthweight and gestation. One hundred and twenty-five potential controls were identified for recruitment on postnatal wards by research staff, who explained to parents that we wished to study mother–infant interactions and infant development in a normal population, for comparison with infants with

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cleft lip: 100 (80%) consented, 22 (18%) refused, 2 (2%) could not be contacted, and 1 (1%) was ineligible. Subsequently, 5 further index infants were excluded as they were diagnosed with an additional disorder, and 2 dropped out, leaving 100 control and 103 index infants. Of the latter, 48 had early, neonatal, lip surgery, and 55 had late, 3–4 month, surgery. Ninety-six (96%) controls, 45 (93.8%) infants having early surgery and 49 (89.1%) having late surgery were assessed at 18 months. There were no differences between the study groups in rates of recruitment or follow-up. The study was approved by all local Ethics Committees. Parents gave written informed consent to participate.

Procedure

Research psychologists video-recorded mother–infant interactions in the home, in quiet conditions, at two, six and 12 months. For interactions at two and six months, the infant was placed in a supportive seat when alert and contented; the mother was asked to sit opposite her infant, and play with him/her for five minutes. A mirror placed adjacent to the infant showed the mother's full-face reflection, and the infant's face opposite her infant, and play with him/her for five minutes. A mirror placed adjacent to the infant showed the mother's full-face reflection, and the infant's face and torso, in the same camera frame. At 12 months, mothers were videotaped when playing with their infants on the floor, using three age-appropriate toys, each presented for 2 minutes, following the procedure described by Stein and colleagues (Stein, Woolley, Cooper, & Fairburn, 1994).

At 18 months, trained psychologists, blind to the timing of lip repair, assessed infant cognitive development. Infant attachment to the mother was assessed, and mothers completed a questionnaire about infant behaviour problems. These assessments were conducted in University research rooms (Reading and Liverpool).

At six months, index group mothers completed questionnaires evaluating the support received from the surgical teams to determine comparability between the centres. In addition, mothers were interviewed at two and six months, by psychologists trained to criterion, to establish whether they had been depressed since the birth. Medical details for index infants were obtained from the surgical teams and hospital records.

Rating procedures and measures

Mother–infant interactions. Interactions were rated from videotapes by trained psychologists. For face-to-face interactions, a masking disc was digitally superimposed on the mouth and nose of all infants, to ensure that ratings were done blind to infant group. Different researchers scored maternal and infant behaviours.

For the 12-month interactions, a further pair of researchers, who had no knowledge of the earlier interactions or maternal group, scored the videotapes, following the procedures of Stein and colleagues (1994) and Wolke and colleagues (1990).

Measures were based on the literature on the effects of cleft lip/palate on mother–infant interactions, and that on predictors of infant cognitive and socio-emotional development. Three core measures (following Wolke, Skuse, & Mathison, 1990; and Murray, Fiori-Cowley, Hooper, & Cooper, 1996), each rated on five-point scales, were used at all ages, namely, maternal sensitivity (empathic, accepting, responsive and appropriate in relation to infant behaviour¹, maternal positive involvement (emotionally expressive, positive, and actively engaged with the infant), and infant distress (fretful vocalisations, crying, frowning, grimacing, back-arching and squirming). Two further measures were used: for face-to-face play, each partner's gaze to the other's face was timed, and expressed as a percentage of the interaction time, using custom-made software to record gaze onsets and offsets in 20 millisecond intervals. For 12-month play, we rated maternal verbal elaboration on the toys (5-point scale). In line with our previous work (Murray et al., 1996), ratings were made for the whole five minutes of face-to-face play; for 12-month interactions, ratings were made for play with each toy separately, and mean scores calculated.

A random 10% sample of videotapes from each assessment was rated by two researchers to establish reliability. Intraclass correlations showed good agreement (range .71 to .94, mean .84.).

Eighteen-month infant outcomes. Infant cognitive development was assessed using the Bayley Scales of Infant Development (1993) to obtain the Mental Development Index (MDI) score. This is a standardised, robust measure of infant cognitive functioning, widely used in research and clinical settings. It is reliable and, when used at this age, predicts later child intelligence quotient (Slater, 1995).

Infant attachment was assessed using the Strange Situation Procedure (Ainsworth, Blehar, Waters, & Wall, 1978). Infants were filmed through a one-way mirror in an unfamiliar play-room for 21 minutes, both in the presence of their mother and of a female stranger. Infant responses to separation from, and reunion with, the mother were used to classify them as either secure (showing distress at the mother's departure, but seeking proximity and being comforted by her upon her return), or else insecurely attached (i.e., either showing avoidant, resistant or disorganised responses), following the criteria of Ainsworth and colleagues (1978) and Main and Solomon (1990). The videotapes were scored by two researchers, trained to reliability, who were blind to the nature of the study.

¹When rating maternal sensitivity, infant behaviour needs to be considered; e.g., maternal smiling or laughing may contribute to a rating of high sensitivity if occurring in response to excited, positive infant behaviour. However, if this maternal behaviour occurred in response to infant distress, it would be rated as low in sensitivity, since a sensitive response would be to show concern and empathy.
Infant behaviour problems were assessed by maternal report. Mothers completed an age-modified version (Murray, 1992) of the Behaviour Screening Questionnaire (BSQ) (Richman & Graham, 1971). This is a widely used, reliable and valid measure that covers the frequency and severity of common childhood behaviour problems (e.g., sleeping and feeding difficulties, separation problems). Total scores were used to characterise infant behavioural difficulties.

Maternal interviews/questionnaires. Maternal depression was assessed by interview, using the Structured Clinical Interview for DSM diagnoses. This is a widely used, standardised instrument, with good reliability and validity (First, Spitzer, Williams, & Gibbon 1994).

Professional support was assessed by index group mothers’ completing a questionnaire designed for the study, covering the personal (trust, confiding fears), and practical (adequate information, and practical help) support provided by the surgical teams, using eight-point scales; Cronbach’s alphas for the two measures were .89 and .66, respectively (see Hill, Murray, Woodall, Parmar, & Hentges, 2004 for further details).

Cleft-specific factors. The following were recorded from medical records using a standard protocol, and were agreed upon by two of the study investigators: cleft palate, antenatal diagnosis, hearing problems (e.g., otitis media), surgical operations in addition to those for initial lip (and any palate) repair. In addition, whether or not the infant’s disfigurement was severe was rated from standardised medical photographs taken before surgery (see appendix).

Data analytic strategy

The three study groups (early repair, late repair and controls) were compared on infant 18-month outcomes, and on mother-infant interactions at two, six and 12 months. At two months, only the early index group had had lip repair, whereas by six months, both index groups had had surgery. One-way ANOVAs, ANCOVAs, Kruskall–Wallis or Chi Square tests were used. Wherever these showed significant effects, we conducted post-hoc tests of the pair-wise comparisons, making appropriate Bonferroni adjustments. Univariate associations were examined between the outcomes and the other relevant factors (SES, infant sex, etc.). If significant, the relevant variable was included as a covariate in group comparisons.

Where significant differences emerged between the early and late surgery groups, we also examined the role of cleft-specific factors. In each case, we tested for main effects, and for whether cleft-specific variables moderated the effects of early and late lip repair. Furthermore, since the rate of antenatal diagnosis differed between the two index groups, we investigated its effects for all outcomes, regardless of whether or not early and late repair groups differed. For normally distributed variables, two-way ANOVAs were conducted, with surgery group and cleft-specific variables as between-subject factors; for non-normally distributed variables, we conducted separate Mann–Whitney U-tests for early and late repair groups.

Finally, where group differences in mother-infant interactions were found that paralleled differences in 18 month outcomes, and where these interaction variables were also associated with the infant outcome in question, we conducted mediational analyses (following Baron & Kenny, 1986), using hierarchical regressions to investigate whether the effects of group on infant outcome still obtained after taking account of mother-infant interactions, or were reduced to non-significance.

Results

The sample characteristics are shown in Table 1. There were no differences between the two index (early and later repair) and the control groups on any of the demographic variables, apart from maternal depression and feeding method at two months. Mothers in both index groups were more likely to be depressed than control mothers, and were more likely to bottle feed; differences between the early vs. late surgery group were not significant. As to cleft-specific variables, only the rate of antenatal diagnosis distinguished the two surgery groups; this was higher in the early repair group.

Variable characteristics

Eighteen-month MDI scores were normally distributed, but BSQ scores were skewed. Mother-infant interaction variables were normally distributed, apart from maternal positive involvement at 12 months, and gaze; the former was normalised by square root transformation; maternal gaze was normalised by log transformations, but infant gaze could not be normalised.

Infant outcomes

Outcomes on the 18-month assessments are shown, by group, in Table 2. None of the potential covariates was associated with the socio-emotional outcomes, and therefore group comparisons were conducted unadjusted. There were no differences between the three groups in either behaviour problems (Kruskall–Wallis, H (2) = 2.072, ns), or quality of attachment, with all groups showing over two-thirds of infants to be securely attached (χ² (2) = .302, ns). A contrasting picture emerged for infant cognitive functioning, where, after controlling for the effect of infant sex, a significant difference between the groups was found on the Bayley MDI (F(2, 187) = 5.573, p < .01); post-hoc tests showed that infants having late lip repair performed significantly more poorly than control

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4 Developmental changes in social interactions across the first year make longitudinal data difficult to interpret, and repeated measures analysis was not considered appropriate.
group infants (difference = 7.53, MDI points, around half a standard deviation, p < .01), whereas those having early repair were comparable to controls (difference = 1.63 MDI points). Furthermore, when the two index groups were compared, there was a trend for the late repair group to have lower MDI scores (difference = 5.9 MDI points, p = .07). Antenatal diagnosis was not associated with infant outcome.

Mother–infant interactions

Means and standard deviations for each group on the different interaction variables at two, six and 12 months are shown in Table 3, together with specification of covariates included in the analyses. Two-month interactions

Having adjusted for relevant covariates, the three groups were found to differ on all dimensions of mother–infant interactions. Post-hoc comparisons showed this was principally due to difficulties in the late repair group: compared to controls, mothers of late repair infants were less positively involved and sensitive, and they looked less at their infants. In turn, infants in this group were more distressed, and they similarly looked less at their mothers. Early repair group mother–infant interactions were, by contrast, comparable to those of controls on all but the measure of maternal sensitivity, where they were rated more poorly.

With regard to early vs. late group comparisons, mothers of infants having late repair were less positively involved with their infants, and their infants also spent less time looking at their mothers. Examination of cleft-specific factors showed infant disfigurement to be important, with mothers of more disfigured infants being less positively involved (F[1, 56] = 5.58, p < .05); this effect was similarly pronounced in both surgery groups, the interaction between the two factors being non-significant, (F[1, 56] = .13, ns). In addition, although there was neither a main effect of severity of disfigurement on infant gaze at two months (U = 501, ns), nor an effect within the early surgery group (U = 138, ns), for infants having late lip repair, the effect was significant, with severely disfigured infants looking at their mothers less than those who were not severely disfigured (U = 44, p < .05).

Table 1 Sample characteristics

<table>
<thead>
<tr>
<th></th>
<th>Control (N = 100)</th>
<th>Early (N = 48)</th>
<th>Late (N = 55)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>N</td>
<td>%</td>
<td>N</td>
</tr>
<tr>
<td>Male</td>
<td>65</td>
<td>65</td>
<td>32</td>
</tr>
<tr>
<td>Middle/upper SES</td>
<td>55</td>
<td>55</td>
<td>24</td>
</tr>
<tr>
<td>1st born</td>
<td>44</td>
<td>44</td>
<td>18</td>
</tr>
<tr>
<td>Mothers depressed at 2 months</td>
<td>2</td>
<td>2</td>
<td>11***</td>
</tr>
<tr>
<td>at 6 months</td>
<td>10</td>
<td>10.4</td>
<td>3</td>
</tr>
<tr>
<td>Breast feeding 2 months</td>
<td>42</td>
<td>49.4</td>
<td>6***</td>
</tr>
<tr>
<td>Mean</td>
<td>28.93</td>
<td>5.36</td>
<td>28.83</td>
</tr>
<tr>
<td>Birthweight (grams)</td>
<td>3528.97</td>
<td>462.15</td>
<td>3406.24</td>
</tr>
<tr>
<td>Gestation (weeks)</td>
<td>39.88</td>
<td>1.34</td>
<td>39.49</td>
</tr>
<tr>
<td>Antenatal diagnosis</td>
<td>23</td>
<td>47.9</td>
<td>13**</td>
</tr>
<tr>
<td>Cleft lip only</td>
<td>21</td>
<td>42.9</td>
<td>20</td>
</tr>
<tr>
<td>Severely disfigured before lip surgery</td>
<td>24</td>
<td>57.1</td>
<td>11</td>
</tr>
<tr>
<td>Hearing problems</td>
<td>12</td>
<td>25.5</td>
<td>13</td>
</tr>
<tr>
<td>Extra operations</td>
<td>4</td>
<td>8.3</td>
<td>9</td>
</tr>
<tr>
<td>Mean</td>
<td>4.83</td>
<td>3.62</td>
<td>109.34</td>
</tr>
<tr>
<td>Age at lip surgery (days)</td>
<td>5</td>
<td>4</td>
<td>6</td>
</tr>
<tr>
<td>Personal support by surgical team 6 months</td>
<td>5</td>
<td>4</td>
<td>6</td>
</tr>
<tr>
<td>Practical support by surgical team 6 months</td>
<td>6</td>
<td>4</td>
<td>6</td>
</tr>
</tbody>
</table>

*a different from control group, b different from early surgery group.

Table 2 Infant outcomes at 18 months

<table>
<thead>
<tr>
<th></th>
<th>Early (n = 45)</th>
<th>Late (n = 49)</th>
<th>Control (n = 96)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bayley MDI</td>
<td>Mean 93.04 SE 11.97</td>
<td>Mean 87.37 <strong>a</strong></td>
<td>Mean 94.88 11.5</td>
</tr>
<tr>
<td>BSQ</td>
<td>Mean 3.89 SE 3.37</td>
<td>Mean 3.24</td>
<td>Mean 3.67 2.97</td>
</tr>
<tr>
<td>Insecure attachment</td>
<td>N %</td>
<td>N %</td>
<td>N %</td>
</tr>
<tr>
<td></td>
<td>13</td>
<td>31</td>
<td>10</td>
</tr>
</tbody>
</table>

*a different from control group.

Effect of cleft lip on infant development

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Infants with both cleft lip and palate looked less at their mothers in both index groups; this effect was significant for the early repair group ($U = 132, p < .05$), and was observable as a trend for the late repair group ($U = 156.5, p < .1$).

Antenatal diagnosis was not related to any two-month interaction measure.

Six-month and 12-month interactions

There were no differences between the groups at either age on any of the interaction measures, and nor was there an effect of antenatal diagnosis.

Mediational analyses

Given that cognitive scores were significantly lower in infants having late lip repair compared to control infants, and that all dimensions of two-month interactions were poorer in the late repair group, we next examined the relationship between these interaction variables and infant MDI scores, restricting analyses to the late repair and control groups. Each two-month mother–infant interaction measure was associated with 18-month infant cognitive functioning: mothers who were more sensitive, who looked at their infants more, and who showed high rates of positive involvement had infants with higher MDI scores ($r = .39, p < .001$; $r = .20, p < .05$; and $r = .26, p < .01$, respectively); similarly, infants who were less distressed, and who looked more at their mother during the two-month interaction, performed better ($r = .18, p < .05$, and $r = .25, p < .01$, respectively).

To determine whether the nature of two-month mother–infant interactions accounted for the difference in cognitive performance between late repair and control group infants, we conducted a series of multiple hierarchical regression analyses, in which each interaction variable was entered first, before considering the effect of group. In each case, apart from that of maternal sensitivity, the effect of late repair group remained significant when the mother–infant interaction variable was taken into account.

When surgery group was added to the model after having accounted for maternal sensitivity, its effect was no longer significant (see Table 4). Thus, early maternal sensitivity mediated the effect of late lip repair on infant cognitive outcome. These results held when adjusting for the effect of infant sex. As a further check on the role of early interactions, we

<table>
<thead>
<tr>
<th>Predictor</th>
<th>B</th>
<th>SE B</th>
<th>$\beta$</th>
<th>$\Delta^2$</th>
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<tbody>
<tr>
<td>Step 1</td>
<td></td>
<td></td>
<td></td>
<td>.15**</td>
</tr>
<tr>
<td>Constant</td>
<td>76.26</td>
<td>3.99</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Maternal sensitivity</td>
<td>4.83</td>
<td>1.07</td>
<td>.387***</td>
<td></td>
</tr>
<tr>
<td>Step 2</td>
<td></td>
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<td>.017</td>
</tr>
<tr>
<td>Constant</td>
<td>80.79</td>
<td>4.93</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Maternal sensitivity</td>
<td>3.97</td>
<td>1.20</td>
<td>.318**</td>
<td></td>
</tr>
<tr>
<td>Group (Late vs. Control)</td>
<td>-2.16</td>
<td>1.40</td>
<td>-1.48</td>
<td></td>
</tr>
</tbody>
</table>

**$p < .01$; ***$p < .001$. 

Table 4 Hierarchical regression analysis: 2-month maternal sensitivity mediates effect of group (late repair vs. control) on infant MDI
investigated whether two-month maternal sensitivity still predicted MDI scores, even when later sensitivity was taken into account. This proved to be the case: thus, although six and 12 month maternal sensitivity each predicted infant MDI ($r = .33, p < .001$, and $r = .26, p < .01$, respectively), the correlation between two-month sensitivity and MDI scores that controlled for both earlier measures was significant ($r = .20, p < .05$).

**Discussion**

In this study of infants with cleft lip (with and without cleft palate) and normal control infants, we found lowered cognitive scores in index infants having late lip repair. For infants having early, neonatal repair, by contrast, cognitive functioning was comparable to that of control infants. Index infants did not differ from controls in terms of attachment or behaviour problems, regardless of the timing of surgery.

Mother–infant interactions at two months, but not six or 12 months, mirrored the pattern of group differences in infant cognitive outcome, with the late repair group showing more interaction difficulties than controls on all dimensions. By contrast, the early repair group was broadly comparable to the controls. Further analyses showed that reduced sensitivity to the infant in late repair group mothers at two months mediated the adverse effect of late repair on infant cognitive outcome.

Our study had a number of strengths: the sample size was relatively large, representative of the target population, and carefully recruited and monitored to ensure the influence of additional syndromes or infant developmental problems did not confound the interpretation of the findings. We also took account of other variables that might affect infant development. The fact that we found a pattern of development in those having late lip repair that was consistent with the results of previous studies of infants with unrepaired clefts, lends weight to previous findings. Nevertheless, our finding that cognitive delay did not occur in infants having neonatal repair suggests that the overall conclusions regarding outcome for infants with clefts may require revision. Before drawing firm conclusions, however, two considerations are warranted. First, it is important to note that this is, to our knowledge, the only work to investigate whether two-month maternal sensitivity to the infant in late repair group mothers still predicted MDI scores, even when later sensitivity was taken into account. This proved to be the case: thus, although six and 12 month maternal sensitivity each predicted infant MDI ($r = .33, p < .001$, and $r = .26, p < .01$, respectively), the correlation between two-month sensitivity and MDI scores that controlled for both earlier measures was significant ($r = .20, p < .05$).

Second, although the Bayley MDI is quite robust, and when administered at this age predicts later child intelligence moderately well, it is important to establish the full clinical significance of our findings by determining whether group differences persist.

Our finding that less sensitive maternal interactions accounted for the group differences in infant cognitive outcome is consistent with the general literature on the role of maternal behaviour in influencing infant cognitive development, as well the findings of Wasserman and Allen (1985) and Speltz and colleagues (2000) in relation to infants with clefts. Further, the fact that two-month interactions had an effect on infant MDI scores even when accounting for subsequent maternal behaviour is consistent with the view that there may be a sensitive period during which particular kinds of social experience may be important for normal cognitive development. Identifying such effects can be difficult: in normal samples, this is partly because the environment is often stable (Cicchetti, 2003; O’Connor, 2003); and in high risk populations, although the early environmental disruption may be time-limited, it cannot always be assumed to occur independently of other factors (Rutter, 2000; O’Connor, 2003). These difficulties did not apply in the current study.

Our findings of interaction difficulties in the late repair group raise the question of why these effects obtain. It could be argued that, prior to surgery, having a disfigured infant may adversely affect maternal mental health, which in turn may affect interactions with the child. We assessed maternal depression in our study, but found no evidence for this explanation. Field and Vega-Lahr (1984) suggested that the disfigurement to infants caused by unrepaired clefts not only makes these children less appealing to look at, but also makes it difficult for parents to interpret infant expressions. It was a limitation of our study that we could not examine interactions before two months, and so the way in which difficulties developed, including the direction of effects, is unknown. Nevertheless, our findings regarding the effects of initial infant disfigurement within the late repair group do suggest that the appearance of the infant, which remained the same over the first few weeks in this group, may have been important. Given that lip repair is generally conducted at 3–4 months, it is important that future research examine the natural history of mother–infant relationships in the context of infant cleft lip, from birth through the early postpartum months, and explore the possible benefits of interventions to facilitate social interactions.

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**Supplementary material**

The following supplementary material is available for this article:

Appendix. Ratings of severity of disfigurement.

This material is available as part of the online article from:


(This link will take you to the article abstract).

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