AN INVESTIGATION OF THE EFFECTS OF PRE-SURGICAL ORTHOPAEDICS ON FEEDING IN INFANTS WITH CLEFT LIP AND/OR PALATE

by

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Abstract

The establishment of a safe and efficient method of feeding in infants with cleft lip and palate is one of the major concerns for parents and the cleft lip and palate team. In spite of a poor knowledge base of the physiological basis of feeding difficulties amongst these infants, numerous management techniques are advocated. Few however are scientifically evaluated. The use of pre-surgical orthopaedics (PSO) is one such technique.

This thesis reports a randomised control trial investigating the effect of PSO on feeding in infants with unilateral cleft lip and palate (UCLP) or isolated cleft palate (ICP).

Infants with non-syndromic UCLP or ICP were eligible for inclusion in the trial. Following recruitment they were randomised to PSO or No PSO. All other aspects of management were standardised. Primary outcome measures were oral motor assessment and anthropometry at 12 months of age. Secondary outcome measures included parent report of feeding at 3, 6 and 12 months of age, physiological measures of feeding, oral motor assessment and anthropometry at 3 and 6 months of age.

Thirty-four infants with UCLP and sixteen infants with ICP were recruited. Equal numbers within each cleft type were randomised to PSO and No PSO.

Although this trial was underpowered the results suggest that PSO have no significant effect on feeding or general body growth post palate repair at 12 months of age. While occasional improvements were found to be associated with the use of PSO pre palate repair at 6 months of age, the differences were not considered clinically significant.
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# Table of Contents

**Chapter** | **Page**
--- | ---
1. Introduction |  
1.1 An overview of cleft lip and/or palate | 22  
1.1.1 Causes | 23  
1.1.2 Presentation | 24  
1.1.3 Classification | 27  
1.2 Management of cleft lip and/or palate | 30  
1.3 Feeding issues | 31  
1.4 Pre-surgical orthopaedics | 32  
1.5 Thesis outline | 36  
2. Literature Review 1: Feeding |  
2.1 Feeding in the cleft lip and palate population | 37  
2.2 Consequences of impaired feeding in the cleft lip and palate population | 42  
2.2.1 Growth | 43  
2.2.2 General development | 46  
2.3 Management of feeding in the cleft lip and palate population | 49  
2.3.1 Positioning | 49  
2.3.2 Assistive feeding devices | 50  
2.3.3 Breast feeding | 54  
2.3.4 PSO and feeding | 55  
2.4 Normal feeding | 60  
2.4.1 Anatomy | 61  
2.4.2 Stages of swallowing | 71  
2.5 Factors which affect the feeding process | 86  
2.5.1 Intrinsic factors of the bolus | 86
<table>
<thead>
<tr>
<th>Section</th>
<th>Page</th>
</tr>
</thead>
<tbody>
<tr>
<td>2.5.2 Feeding method/equipment</td>
<td>87</td>
</tr>
<tr>
<td>2.5.3 Mechanical factors</td>
<td>88</td>
</tr>
<tr>
<td>2.5.4 Demographic and social factors</td>
<td>89</td>
</tr>
<tr>
<td>2.5.5 Prenatal and Perinatal history</td>
<td>89</td>
</tr>
<tr>
<td>2.5.6 Psychological factors</td>
<td>90</td>
</tr>
<tr>
<td>2.5.7 Infant factors</td>
<td>90</td>
</tr>
<tr>
<td>2.5.8 Behavioural aspects</td>
<td>90</td>
</tr>
<tr>
<td>3.1 Background information</td>
<td>92</td>
</tr>
<tr>
<td>3.2 Clinical feeding evaluation</td>
<td>93</td>
</tr>
<tr>
<td>3.3 Paediatric feeding assessment scales</td>
<td>94</td>
</tr>
<tr>
<td>3.3.1 Leaf and Gisel Observational System</td>
<td>94</td>
</tr>
<tr>
<td>3.3.2 Neonatal Oral-Motor Assessment Scale</td>
<td>94</td>
</tr>
<tr>
<td>3.3.3 Schedule for Oral Motor Assessment</td>
<td>95</td>
</tr>
<tr>
<td>3.3.4 Oral Motor/Feeding Rating Scale</td>
<td>96</td>
</tr>
<tr>
<td>3.3.5 Paediatric Oral Skills Package</td>
<td>97</td>
</tr>
<tr>
<td>3.4 Diagnostic tests and procedures</td>
<td>97</td>
</tr>
<tr>
<td>3.4.1 Pulse oximetry</td>
<td>98</td>
</tr>
<tr>
<td>3.4.2 Cervical auscultation</td>
<td>101</td>
</tr>
<tr>
<td>3.4.3 Exeter Dysphagia Assessment Technique</td>
<td>103</td>
</tr>
<tr>
<td>3.4.4 Intra-oral visualisation during suckle feeding</td>
<td>104</td>
</tr>
<tr>
<td>3.4.5 Measurement of sucking using pressure transducers</td>
<td>104</td>
</tr>
<tr>
<td>3.4.6 Whitney strain gauge</td>
<td>106</td>
</tr>
<tr>
<td>3.4.7 Videofluoroscopic assessment of swallowing</td>
<td>106</td>
</tr>
<tr>
<td>3.4.8 Fibreoptic Endoscopic Evaluation of Swallowing Safety</td>
<td>109</td>
</tr>
<tr>
<td>4.1 Reasons for development of the GOSMIF</td>
<td>112</td>
</tr>
<tr>
<td>4.2 Components of the GOSMIF</td>
<td>114</td>
</tr>
<tr>
<td>4.2.1 Hardware</td>
<td>116</td>
</tr>
</tbody>
</table>
4.2.2 Software ................................................................. 118
4.3 Storage of data .............................................................. 137
4.4 Modifications to the GOSMIF during its development .... 137
4.5 Normative data .............................................................. 137
4.6 Reliability ....................................................................... 147

5 Trial Methodology

5.1 Hypothesis ...................................................................... 158
5.2 Aims and research questions ......................................... 158
5.3 Outcome measures .......................................................... 160
5.4 Context of trial ............................................................... 161
5.5 Procedure ....................................................................... 161
  5.5.1 Patient selection ..................................................... 161
  5.5.2 Prenatal contact ..................................................... 162
  5.5.3 Early counselling .................................................. 163
  5.5.4 Feeding advice ...................................................... 163
  5.5.5 Intra-oral impressions ........................................... 164
  5.5.6 Surgery .................................................................... 166
  5.5.7 Cleft Lip and Palate Clinic appointments (CLPC) ........ 166
  5.5.8 Multidisciplinary team ........................................... 169
  5.5.9 Orthodontic appointments ..................................... 169
5.6 Assessment tools ........................................................... 170
  5.6.1 Anthropometry ..................................................... 170
  5.6.2 Oral motor skills .................................................... 172
  5.6.3 Physiological measures of bottle feeding ................. 173
  5.6.4 Feeding questionnaire ........................................... 173
  5.6.5 Videofluoroscopy .................................................. 174
5.7 Potential confounders/additional information .................. 175
  5.7.1 Structured interviewer led questionnaires ............... 175
  5.7.2 Denver II ............................................................. 175
5.8 Data collection routine ..................................................... 176
5.8.1 Neonatal assessment (Data collection point 1) ..... 178
5.8.2 3 month assessment (Data collection point 2) ..... 178
5.8.3 6 month assessment (Data collection point 3) ..... 179
5.8.4 9 month assessment (Data collection point 4) ..... 180
5.8.5 12 month assessment (Data collection point 5) ..... 180
5.9 Blinding .................................................................................................. 181
5.10 Statistical analysis ................................................................................ 182
5.11 Sample size .......................................................................................... 183
5.12 Availability of patients ......................................................................... 185

6 Results
6.1 Recruitment ......................................................................................... 187
6.2 Numbers of patients assessed and analysed at different data collection points .................................................................................. 189
6.3 Adherence to protocol specified assessment times ......................... 189
6.4 Baseline data ....................................................................................... 194
6.5 Baseline risk factors for developmental delay and feeding difficulties ................................................................................................. 196
   6.5.1 Prenatal factors ............................................................................. 197
   6.5.2 Perinatal factors ........................................................................... 198
   6.5.3 Later postnatal factors ................................................................. 200
6.6 Subsequent risk factors for developmental delay and feeding difficulties ................................................................................................. 201
   6.6.1 Family stress ................................................................................. 201
   6.6.2 Infants’ medical problems ............................................................ 202
   6.6.3 General development .................................................................... 203
6.7 Feeding advice ..................................................................................... 204
6.8 Orthodontics ....................................................................................... 204
6.9 Compliance with PSO .......................................................................... 205
6.10 Intention to treat analyses .................................................................. 206
   6.10.1 Feeding methods ......................................................................... 206
6.10.2 Parent report of feeding characteristics ................. 220
6.10.3 Oral motor skills ....................................................... 231
6.10.4 GOSMIF .................................................................... 248
6.10.5 Videofluoroscopy findings ..................................... 271
6.10.6 Anthropometry.......................................................... 272

6.11 Per protocol analyses ....................................................................... 286
6.11.1 Oral motor skills ........................................................ 286
6.11.2 Anthropometry.......................................................... 286

6.12 Summary ........................................................................................... 296

7 Discussion and conclusions
7.1 Results................................................................................................. 301
7.2 Factors which may have affected the results of the trial .......... 324
7.2.1 Study design ................................................................................. 325
7.2.2 Subjects ............................................................................... 326
7.2.3 Numbers of infants recruited ................................... 329
7.2.4 Variation in aspects of standardised care .......... 330
7.2.5 Orthodontics ............................................................................ 331
7.2.6 Adherence to protocol specified assessment times ......................................................... 331
7.2.7 Assessment tools ........................................................................ 332
7.2.8 Compliance with PSO............................................................ 339
7.2.9 Medical problems over time............................................ 340
7.2.10 Analysis .............................................................................. 341

7.3 Generalising these findings to other cleft types and protocols for management of infants with CL and/or P .......... 342

7.4 Further research.................................................................................. 343
7.5 Conclusions ....................................................................................... 344

Appendices ........................................................................................................................ 346

References .......................................................................................................................... 427
### List of figures

<table>
<thead>
<tr>
<th>Number</th>
<th>Description</th>
<th>Page</th>
</tr>
</thead>
<tbody>
<tr>
<td>1.</td>
<td>Diagrammatic representation of cleft lip (a), cleft palate (b), unilateral</td>
<td>26</td>
</tr>
<tr>
<td></td>
<td>cleft lip and palate (c) and bilateral cleft lip and palate (d)</td>
<td></td>
</tr>
<tr>
<td>2.</td>
<td>Photographs and CARE classification of an infant with complete</td>
<td>28</td>
</tr>
<tr>
<td></td>
<td>unilateral cleft of the lip and palate</td>
<td></td>
</tr>
<tr>
<td>3.</td>
<td>Photographs and CARE classification of an infant with an incomplete</td>
<td>29</td>
</tr>
<tr>
<td></td>
<td>cleft of the hard palate and complete cleft of the soft palate</td>
<td></td>
</tr>
<tr>
<td>4.</td>
<td>“Active” baby plate (DiBiase and Hunter, 1983)</td>
<td>33</td>
</tr>
<tr>
<td>5.</td>
<td>“Passive” baby plate (DiBiase and Hunter, 1983)</td>
<td>34</td>
</tr>
<tr>
<td>6.</td>
<td>Diagrammatic representation of differences between infant, older</td>
<td>62</td>
</tr>
<tr>
<td></td>
<td>child and adult anatomy (Arvedson and Brodsky, 2002)</td>
<td></td>
</tr>
<tr>
<td>7.</td>
<td>Simplified diagram demonstrating the principles of the pharynx acting</td>
<td>63</td>
</tr>
<tr>
<td></td>
<td>as a “combined channel” (Wolf and Glass, 1992)</td>
<td></td>
</tr>
<tr>
<td>8.</td>
<td>Diagram showing the distortion of the alar cartilages and nasal</td>
<td>65</td>
</tr>
<tr>
<td></td>
<td>septum in cleft lip (Sommerlad, 2000)</td>
<td></td>
</tr>
<tr>
<td>9.</td>
<td>A schematic drawing of the lip musculature in unilateral cleft lip as</td>
<td>66</td>
</tr>
<tr>
<td></td>
<td>compared to normal (Sommerlad, 2000)</td>
<td></td>
</tr>
<tr>
<td>10.</td>
<td>Diagram of the cleft muscular anatomy (Sommerlad, 2000)</td>
<td>67</td>
</tr>
<tr>
<td>11.</td>
<td>Diagrammatic representation of the levels of airway protection</td>
<td>70</td>
</tr>
<tr>
<td></td>
<td>(Arvedson and Brodsky, 2002)</td>
<td></td>
</tr>
<tr>
<td>12.</td>
<td>Diagrammatic representation of larynx in relation to the tongue and</td>
<td>70</td>
</tr>
<tr>
<td></td>
<td>soft palate</td>
<td></td>
</tr>
<tr>
<td>13.</td>
<td>Diagrammatic representation of interdependence of the oral,</td>
<td>72</td>
</tr>
<tr>
<td></td>
<td>pharyngeal and oesophageal stages of swallow (Groher, 1984)</td>
<td></td>
</tr>
<tr>
<td>14.</td>
<td>Diagrammatic representation of the oral and pharyngeal stage</td>
<td>73</td>
</tr>
<tr>
<td></td>
<td>sequence (Groher, 1984)</td>
<td></td>
</tr>
</tbody>
</table>
15. Diagrammatic representation of positive and negative pressure generation during sucking (Wolf and Glass, 1992) .................................................. 78
16. Diagrammatic representation of the differences between non-nutritive and nutritive sucking (Wolf and Glass, 1992) .................................................. 80
17. Block diagram of GOSMIF .................................................................................. 115
18. Photograph of GOSMIF .................................................................................. 116
19. Photographs of NUK vented (a) and unvented (b) teats ..................................... 117
20. Data capture window ......................................................................................... 120
21. Data review window ......................................................................................... 121
22. Loading files for analysis .................................................................................. 121
23. Data review window ......................................................................................... 122
24. Data analysis window ...................................................................................... 124
25. Data analysis window ...................................................................................... 126
26. Identification of sucking bursts ........................................................................ 127
27. Marking the beginning and end of a sucking burst .......................................... 127
28. Automatic analysis window ............................................................................. 129
29. Illustration of swallow sounds identified in sucking burst (taken from (Vice et al., 1990)) .............................................................................................. 131
30. Example of accelerometer trace used in the identification of Bolus Transit Sounds (BTS) .................................................................................. 132
31. GOSMIF summary screen, showing bolus in pharynx and simultaneous accelerometer trace, taken during simultaneously GOSMIF and videofluoroscopic assessment of swallowing ........................................ 134
32. GOSMIF summary screen showing accelerometer traces where swallows were confirmed with videofluoroscopy ........................................ 135
33. Scatter plot of “length of sucking bursts” of non-cleft infants (3 per infant) ........................................................................................................ 141
34. Scatter plot of “average pressure generation” of non-cleft infants .................. 142
35. Scatter plot of “% pressure generated above baseline pressure in bottle” of non-cleft infants .................................................................................. 143
36. Scatter plot of “rate of sucking” of non-cleft infants ........................................ 144
37. Scatter plot of "length of sucks or peak to peak intervals" of non-cleft infants ................................................................. 145
38. Scatter plot of "suck swallow ratios" of non-cleft infants ................................................................. 146
39. Beginning of sucking bursts for each rater .............................................................................. 148
40. End of sucking bursts for each rater .................................................................................. 149
41. Scatter plot showing the length of sucking bursts selected by each rater ................................................................. 151
42. Scatter plot showing the rate of sucking for each sucking bursts and raters .............................................................................. 153
43. Scatter plot showing the peak to peak intervals for sucking bursts and raters ................................................................. 154
44. Scatter plot showing the average pressure generation for sucking bursts and raters .............................................................................. 155
45. Scatter plot showing the % of pressure generated above baseline pressure in bottle for sucking bursts and raters .............................................................................. 156
46. Impression tray and Optosil ................................................................................................. 165
47. Dental cast ........................................................................................................................................ 165
48. Timetable for CLPC appointments for infants with UCLP, in relation to surgery .............................................................................. 167
49. Timetable for CLPC appointments for infants with ICP in relation to surgery .............................................................................. 168
50. Diagram showing technique for measuring recumbent length (Gibson, 1990) .............................................................................. 171
51. Diagrammatic representation of method of measuring head circumference (Gibson, 1990) .............................................................................. 172
52. Timetable of data collection points and assessment tools used in relation to surgical management .............................................................................. 177
53. (a): Graph showing the standardised difference in means that can be detected with 80, 90 and 95% power at 5% significance with specified numbers per group .............................................................................. 184
   (b): Graph showing percentage reduction detected with 80, 90, 95%
power at 5% significance level for different control percentages and
two groups of 25 infants.....................................................................................185

54. (a) Diagram showing the flow of participants with UCLP through
each stage of the trial; based on template recommended by Altman et
al (2001) .................................................................................................................192

(b) Diagram showing the flow of participants with ICP through each
stage of the trial; based on template recommended by Altman et al
(2001)......................................................................................................................193

55. Length of feeding over time for UCLP NO PSO group .......... 222

56. Length of feeding over time for UCLP PSO group .......... 222

57. Length of feeding over time for ICP NO PSO group .......... 223

58. Length of feeding over time for ICP PSO group .......... 223

59. SOMA scores for (bottle) UCLP No PSO (6 months) .......... 243

60. SOMA scores (bottle) UCLP PSO (6 months) .......... 243

61. SOMA scores (bottle) ICP No PSO (6 months) .......... 244

62. SOMA scores (bottle) ICP PSO (6 months) .......... 244

63. SOMA scores (puree) UCLP No PSO (6 months) .......... 245

64. SOMA scores (puree) UCLP PSO (6 months) .......... 245

65. SOMA scores (puree) ICP No PSO group (6 months) .......... 246

66. SOMA scores (puree) ICP PSO group (6 months) .......... 246

67. GOSMIF study, pattern 1 .................................................. 249

68. GOSMIF study pattern 2 .................................................. 249

69. Length of first 3 rateable sucking bursts at neonatal assessment .... 252

70. Length of first 3 rateable sucking bursts at 3 months .......... 252

71. Length of first 3 rateable sucking bursts at 6 months .......... 252

72. Changes in median length of sucking bursts for individual infants
within UCLP No PSO group ................................................................. 253

73. Changes in median length of sucking bursts for individual infants
within UCLP PSO group ................................................................. 253

74. Changes in median length of sucking bursts for individual infants
within ICP No PSO group ................................................................. 254
75. Changes in median length of sucking bursts for individual infants
   within ICP PSO group ................................................................. 254
76. Peak to peak intervals at neonatal assessment .................................. 256
77. Peak to peak intervals at 3 months .................................................. 256
78. Peak to peak intervals at neonatal assessment 6 months ................. 256
79. Changes in peak to peak interval for individual infants over time,
   within UCLP No PSO group ....................................................... 257
80. Changes in peak to peak interval for individual infants over time,
   within UCLP PSO group ............................................................ 257
81. Changes in peak to peak interval for individual infants over time,
   within ICP No PSO group .......................................................... 258
82. Changes in peak to peak interval for individual infants over time,
   within ICP PSO group ............................................................... 258
83. Rate of sucking at neonatal assessment .......................................... 260
84. Rate of sucking at 3 months ......................................................... 260
85. Rate of sucking at 6 months ........................................................ 260
86. Changes in rate of sucking over time, for individual infants, within
   UCLP No PSO group ................................................................. 261
87. Changes in rate of sucking over time, for individual infants, within
   UCLP PSO group ................................................................. 261
88. Changes in rate of sucking over time, for individual infants, within
   ICP No PSO group ................................................................. 262
89. Changes in rate of sucking over time, for individual infants, within
   ICP PSO group ................................................................ 262
90. Suck swallow ratios at neonatal assessment ..................................... 264
91. Suck swallow ratios at 3 months .................................................... 264
92. Suck swallow ratios at 6 months .................................................... 264
93. Changes in suck swallow ratios over time, for individual infants,
   within UCLP No PSO group ...................................................... 265
94. Changes in suck swallow ratios over time, for individual infants,
   within UCLP PSO group .......................................................... 265
95. Changes in suck swallow ratios over time, for individual infants,
within ICP No PSO group ................................................................................. 266
96. Changes in suck swallow ratios over time, for individual infants,
within ICP PSO group .................................................................................... 266
97. % pressures generated above baseline pressure in bottle at neonatal
    assessment ..................................................................................................... 268
98. % pressures generated above baseline pressure in bottle at 3 months .... 268
99. % pressures generated above baseline pressure in bottle at 6 months .... 268
100. Changes in % pressure generated above baseline pressure in bottle,
     over time, for individual infants, within UCLP No PSO group .............. 269
101. Changes in % pressure generated above baseline pressure in bottle,
     over time, for individual infants, within UCLP PSO group ................... 269
102. Changes in % pressure generated above baseline pressure in bottle,
     over time, for individual infants, within ICP No PSO group ................. 270
103. Changes in % pressure generated above baseline pressure in bottle,
     over time, for individual infants, within ICP PSO group ....................... 270
104. "Z" scores for weight over time for UCLP No PSO group ....................... 274
105. "Z" scores for weight over time for UCLP PSO group ............................. 274
106. "Z" scores for weight over time for ICP No PSO group ........................... 275
107. "Z" scores for weight over time for ICP PSO group ............................... 275
108. "Z" scores for height over time for UCLP No PSO group ....................... 276
109. "Z" scores for height over time for UCLP PSO group ......................... 276
110. "Z" scores for height over time for ICP No PSO group ......................... 277
111. "Z" scores for height over time for ICP PSO group ............................... 277
112. "Z" scores for head circumference UCLP No PSO group ....................... 278
113. "Z" scores for head circumference UCLP PSO group ............................... 278
114. "Z" scores for head circumference ICP No PSO group .......................... 279
115. "Z" scores for head circumference ICP PSO group ............................... 279
116. "Z" scores for bmi UCLP No PSO group ................................................ 280
117. "Z" scores for bmi UCLP PSO group ....................................................... 280
118. "Z" scores for bmi ICP No PSO group ..................................................... 281

-xiv -
119. "Z" scores for bmi ICP PSO group ........................................................... 281
120. "Z" scores for weight in UCLP no PSO ..................................................... 288
121. "Z" scores for weight in UCLP PSO group taking into consideration PSO compliance group .............................................................. 288
122. "Z" scores for height in UCLP no PSO group ............................................ 289
123. "Z" scores for height in UCLP PSO group taking into consideration PSO compliance ........................................................................ 289
124. "Z" scores for head circumference in UCLP no PSO group ...................... 290
125. "Z" scores for head circumference in UCLP PSO group taking into consideration PSO compliance ............................................................. 290
126. "Z" scores for bmi in UCLP no PSO group ................................................ 291
127. "Z" scores for bmi in UCLP PSO group taking into consideration PSO compliance ............................................................................. 291
128. "Z" scores for weight in ICP no PSO group ................................................. 292
129. "Z" scores for weight in ICP PSO group taking into consideration PSO compliance .................................................................................. 292
130. "Z" scores for height in ICP no PSO group ............................................... 293
131. "Z" scores for height in ICP PSO group taking into consideration PSO compliance .................................................................................. 293
132. "Z" scores for head circumference in ICP no PSO group .......................... 294
133. "Z" scores for head circumference in ICP PSO group taking into consideration PSO compliance ............................................................. 294
134. "Z" scores for bmi in ICP no PSO group .................................................. 295
135. "Z" scores for bmi ICP PSO group taking into consideration PSO compliance respectively ................................................................. 295
136. Graph showing the standardised difference in means that can be detected with 80. 90 and 95% power at 5% significance with specified numbers per group ............................................................................. 297
137. GOSMIF data review screen for non cleft infant ..................................... 307
138. GOSMIF data review screen showing pattern 1 for infants with CL and/or P ..................................................................................... 308
139. GOSMIF data review screen showing pattern 2 for infants with CL and/or P ................................................................................................................308

140. GOSMIF data review screens showing differences in feeding patterns between neonatal and 3 month assessments for infants with UCLP........310

141. Scatterplots showing length of sucking bursts for non-cleft infants and all groups of infants with CL and/or P at neonatal assessment .......... 312

142. Scatterplots showing rate of sucking for non-cleft infants and all groups of infants with CL and/or P at neonatal assessment .............. 312

143. Scatterplots showing length of individual sucks (peak to peak intervals) for non-cleft infants and all groups of infants with CL and/or P at neonatal assessment ........................................................................ 313

144. Scatterplots showing percent pressure generated above baseline pressure for non-cleft infants and all groups of infants with CL and/or P at neonatal assessment ........................................................................... 314

145. Scatterplots showing suck swallow ratios for non-cleft infants and all groups of infants with CL and/or P at neonatal assessment ........... 315
<table>
<thead>
<tr>
<th>Number</th>
<th>Table</th>
<th>Page</th>
</tr>
</thead>
<tbody>
<tr>
<td>1.</td>
<td>Table taken from Clarren et al (1987)</td>
<td>39</td>
</tr>
<tr>
<td>2.</td>
<td>Demographic details for “non cleft” cohort</td>
<td>139</td>
</tr>
<tr>
<td>3.</td>
<td>Percents of variance attributed to bursts, raters and patients for selected measurements obtained with GOSMIF</td>
<td>157</td>
</tr>
<tr>
<td>4.</td>
<td>Number of infants assessed at each data collection point according to cleft type and PSO status</td>
<td>189</td>
</tr>
<tr>
<td>5.</td>
<td>Table of ages of infants (months) at data collection points</td>
<td>191</td>
</tr>
<tr>
<td>6.</td>
<td>Summary of infants recruited, distribution across groups; sex, referral site, gestational ages, birth-weights and birth order</td>
<td>195</td>
</tr>
<tr>
<td>7.</td>
<td>Risk factors for developmental delay and/or feeding problems</td>
<td>199</td>
</tr>
<tr>
<td>8.</td>
<td>Infant medical problems</td>
<td>200</td>
</tr>
<tr>
<td>9.</td>
<td>Orthodontic visits and problems</td>
<td>205</td>
</tr>
<tr>
<td>10.</td>
<td>Summary of feeding methods used at neonatal assessment</td>
<td>207</td>
</tr>
<tr>
<td>11.</td>
<td>Summary of modifications to teats at neonatal assessment</td>
<td>208</td>
</tr>
<tr>
<td>12.</td>
<td>Feeding methods used at 3 months of age</td>
<td>209</td>
</tr>
<tr>
<td>13.</td>
<td>Bottles used at 3 months of age</td>
<td>210</td>
</tr>
<tr>
<td>14.</td>
<td>Summary of modifications to teats at 3 months</td>
<td>211</td>
</tr>
<tr>
<td>15.</td>
<td>Feeding methods used at 6 months of age</td>
<td>212</td>
</tr>
<tr>
<td>16.</td>
<td>Bottles used at 6 months of age</td>
<td>213</td>
</tr>
<tr>
<td>17.</td>
<td>Adaptations made to bottle at 6 months of age</td>
<td>214</td>
</tr>
<tr>
<td>18.</td>
<td>Textures accepted at 6 months of age</td>
<td>215</td>
</tr>
<tr>
<td>19.</td>
<td>Feeding methods used at 12 months of age</td>
<td>216</td>
</tr>
<tr>
<td>20.</td>
<td>Bottles/beakers used at 12 months of age</td>
<td>217</td>
</tr>
<tr>
<td>21.</td>
<td>Summary of modifications to teats at 12 months</td>
<td>218</td>
</tr>
<tr>
<td>22.</td>
<td>Textures accepted at 12 months</td>
<td>219</td>
</tr>
</tbody>
</table>
23. Reported length of solid feeds ................................................................. 225
24. Summary of reported feeding characteristics at neonatal assessment ...... 226
25. Summary of reported feeding characteristics at 3 months ....................... 227
26. Summary of reported feeding characteristics at 6 months ....................... 228
27. Frequency and extent of episodes of nasal regurgitation during feeding 6 months ........................................................................................................ 229
28. Summary of reported feeding characteristics at 12 months ..................... 230
29. NOMAS ratings at neonatal assessment ................................................... 232
30. NOMAS ratings at 3 month assessment point ........................................... 233
31. Comparison of NOMAS scores for GOSMIF and routine adapted bottle-feeds at neonatal assessment ................................................................. 234
32. Comparisons of NOMAS ratings with routine adaptive bottle vs. GOSMIF for UCLP No PSO, at neonatal assessment .................................................. 236
33. Comparisons of NOMAS ratings with routine adaptive bottle vs. GOSMIF for UCLP PSO, at neonatal assessment .................................................... 236
34. Comparisons of NOMAS ratings with routine adaptive bottle vs GOSMIF for ICP No PSO, at neonatal assessment ................................................... 237
35. Comparisons of NOMAS ratings with routine adaptive bottle vs GOSMIF for ICP PSO, at neonatal assessment ....................................................... 237
36. Comparison of NOMAS scores for GOSMIF and routine adapted bottle feeds at 3 month assessment ........................................................................ 238
37. Comparisons of NOMAS ratings with routine adaptive bottle vs. GOSMIF for UCLP No PSO, at 3 months of age ....................................................... 240
38. Comparisons of NOMAS ratings with routine adaptive bottle vs. GOSMIF for UCLP PSO, at 3 months of age ......................................................... 240
39. Comparisons of NOMAS ratings with routine adaptive bottle vs. GOSMIF for ICP No PSO, at 3 months of age ......................................................... 241
40. Comparisons of NOMAS ratings with routine adaptive bottle vs. GOSMIF for ICP PSO, at 3 months of age ......................................................... 241
41. SOMA raw scores for cracker .................................................................. 248
42. Mean length of sucking bursts, p values and confidence intervals for No PSO and PSO groups for UCLP and ICP at neonatal, 3 and 6 month assessments .............................................................. 251

43. Mean peak to peak intervals, p values and confidence intervals for No PSO and PSO groups for UCLP and ICP at neonatal, 3 and 6 month assessments ............................................................................................................ 255

44. Mean rate of sucking, p values and confidence intervals for No PSO and PSO groups for UCLP and ICP at neonatal, 3 and 6 month assessments ............................................................................................................ 259

45. Mean suck swallow ratios, p values and confidence intervals for No PSO and PSO groups for UCLP and ICP at neonatal, 3 and 6 month assessments ............................................................................................................ 263

46. Mean % positive pressure generated above baseline, p values and confidence intervals for No PSO and PSO groups for UCLP and ICP at neonatal, 3 and 6 month assessments ........................................................... 267

47. Oral and pharyngeal behaviours rated as abnormal on videofluoroscopy ................................................................................................................................. 272

48. Table showing the last assessment points used in comparisons of anthropometric measures ........................................................................................................ 282

49. Means, p values and confidence intervals for differences between UCLP (No PSO and PSO) and ICP (No PSO and PSO) for weight, height, head circumference and body mass index z scores at last assessment point ........................................................................................................... 284

50. Means, p values and confidence intervals for differences between UCLP (No PSO and PSO) and ICP (No PSO and PSO) for weight, height, head circumference and body mass index z scores at 12 month assessment point ........................................................................................................... 285
# Appendices

<table>
<thead>
<tr>
<th>Number</th>
<th>Page</th>
</tr>
</thead>
<tbody>
<tr>
<td>1.</td>
<td>346</td>
</tr>
<tr>
<td>2.</td>
<td>348</td>
</tr>
<tr>
<td>3.</td>
<td>350</td>
</tr>
<tr>
<td>4.</td>
<td>352</td>
</tr>
<tr>
<td>5.</td>
<td>353</td>
</tr>
<tr>
<td>6.</td>
<td>364</td>
</tr>
<tr>
<td>7.</td>
<td>365</td>
</tr>
<tr>
<td>8.</td>
<td>369</td>
</tr>
<tr>
<td>9.</td>
<td>370</td>
</tr>
<tr>
<td>10.</td>
<td>371</td>
</tr>
<tr>
<td>11.</td>
<td>372</td>
</tr>
<tr>
<td>12.</td>
<td>378</td>
</tr>
<tr>
<td>13.</td>
<td>379</td>
</tr>
<tr>
<td>14.</td>
<td>384</td>
</tr>
<tr>
<td>15.</td>
<td>388</td>
</tr>
<tr>
<td>16.</td>
<td>389</td>
</tr>
<tr>
<td>17.</td>
<td>390</td>
</tr>
<tr>
<td>18.</td>
<td>395</td>
</tr>
<tr>
<td>19.</td>
<td>400</td>
</tr>
<tr>
<td>20.</td>
<td>403</td>
</tr>
<tr>
<td>21.</td>
<td>404</td>
</tr>
<tr>
<td>22.</td>
<td>408</td>
</tr>
<tr>
<td>23.</td>
<td>409</td>
</tr>
</tbody>
</table>
24. Feeding history update form, data collection point 4 ..................................... 415
25. Feeding history update form, data collection point 5 ..................................... 421
One of the greatest early concerns for parents of infants born with cleft lip and/or palate is the establishment of safe and efficient feeding (Trenouth and Campbell, 1996a; Oliver and Jones, 1997). A thorough understanding of the physiological basis and nature of the feeding problems is essential if effective management is to be instituted. Many management techniques are advocated to facilitate feeding, however few of these have been scientifically proven. The use of pre-surgical orthopaedics is one such technique. The purpose of this thesis is to report the findings of an investigation into the effect of pre-surgical orthopaedics on feeding in infants with cleft lip and/or palate.

This chapter sets the scene for this investigation. The incidence, causes and presentation of cleft lip and palate are summarised. The early management of infants with cleft lip and/or palate is outlined with particular reference to the early establishment of feeding and the use of pre-surgical orthopaedics. The outline of this thesis is described.

1.1 Cleft lip and/or palate overview

Cleft lip and palate deformities are the most common congenital craniofacial malformations (World Health Organisation Expert Committee, 2002). The prevalence of cleft lip and palate is difficult to determine accurately with little international consistency. The literature suggests that the incidence is highest in American Indians where an incidence of 3.7/1000 live births is reported, followed by an incidence of 2.7/1000 in the Japanese population, 2.0/1000 in
the Chinese and Maori populations, 1.7/1000 in Caucasians and 0.4/1000 in “Blacks” (Vanderas, 1987; World Health Organisation Expert Committee, 2002). Among the British population an incidence of 1 in 700 live births is reported with 1000 new cases each year (World Health Organisation Expert Committee, 2002; Craniofacial Anomalies Network, 2003).

1.1.1. Causes of cleft lip and/or palate

Clefts of the lip and palate result from incomplete fusion of the palatal shelves, around 4-9 weeks gestation (Watson, 2002). In normal embryology, fusion of the median nasal processes, the lateral nasal processes and the maxillary prominences occurs resulting in the formation of the upper lip and premaxilla known as the primary palate. The hard and soft palate is formed by fusion of the palatine processes and is known as the secondary palate. The palatine processes also fuse with the primary palate, posterior to the incisive foramen, along the premaxilla structures, and the nasal septum and vomer bone superiorly. The nasal septum grows downwards, fuses with the palate and separates the nostrils. The process of fusion is viewed as highly complex and is not fully understood (Burdi and Silvey, 1969; Humphrey, 1969; Humphrey, 1971; Diewert, 1974; Kjaer et al., 1993). When fusion does not occur clefts of the lip and/or palate result. Several causative factors have been proposed to account for the breakdown in fusion including hypoplasia of the facial shelves, variation in facial geometry, inability of the epithelium to fuse, excessive mesenchymal seam cell death and the tongue becoming positioned so it interferes with movement of the palatal shelves (Fara, 1971; Mato et al., 1972; Mitts et al., 1981; Bagatin and Zajc, 1985; Ferguson, 1987a; Kitamura, 1991; Watson, 2002).

Cleft lip and/or palate (CL and/or P) is increasingly recognised as a group of conditions associated with other anomalies and syndromes (Cohen, 1978; Winter and Baraitser, 1998). Some of these syndromes are caused by an abnormal gene. In Treacher Collins syndrome the abnormal gene is 5q32-
Similarly, in non-syndromic CL and/or P, some authorities suggest the cause maybe a single abnormal gene as yet unknown (Fukuhara, 1965; Melnick et al., 1968; Coccia et al., 1969; Chabora and Horowitz, 1974; Melnick and Sheilds, 1976; Bixler, 1981; Marazita et al., 1984; Melnick, 1986; Marazita et al., 1986; Mitchell and Risch, 1992; Ray et al., 1993). In the absence of an abnormal gene or chromosomal disorder, a multi-factorial model involving genetic and environmental factors is hypothesised (Fraser and Pashayan, 1970; Fraser, 1971; Fraser, 1977; Fraser, 1978; Johnston and Millicovsky, 1985; Chung et al., 1986; Czeizal and Nagy, 1986; Ferguson, 1987b; Johnston et al., 1990; Ray et al., 1993). Many environmental factors have been implicated including exposure to pesticides (Gordon and Shy, 1981; Owens et al., 1985; Coupland and Coupland, 1988) nitrate compounds (Dorsch et al., 1984), maternal epilepsy and the medications used to manage this (Meadow, 1968; Hanson et al., 1976; Owens et al., 1985; Czeizal and Nagy, 1986; Friis, 1989) maternal diet and vitamin intake (Tolarova, 1982; Fernhoff and Lammer, 1984; Tolarova, 1987), alcohol, (Jones et al., 1973; Hanson and Smith, 1975; Clarren and Smith, 1978), and cigarettes (Khoury et al., 1977; Ericson et al., 1979; Khoury et al., 1989; Werler et al., 1990).

1.1.2 Presentation of cleft lip and/or palate

The presentation of CL and/or P malformations at birth is highly variable. Within the UK about 43% of reported clefts are of the palate only, 21% of the lip only and about 30% of the lip and palate. The remaining 6% are “other clefts” or “missing data” (Craniofacial Anomalies Network, 2003).

Clefts of the lip (CL) may vary from a small notch in the lip to a complete cleft extending through the lip, the floor of the nostril and the alveolus. The extent of clefts of the alveolus may be minimal varying from a small notch or extending completely through the arch. They may be unilateral or bilateral (Figure 1a).
Cleft palate may occur in isolation or in conjunction with cleft lip. Isolated cleft palate (ICP) ranges from minimal defects of the soft palate to complete defects forward to the incisive foramen. They also vary in width, from a narrow slit to a large horseshoe shaped defect (Figure 1b). A diagnosis of Pierre Robin Sequence (PRS) is given where there is a large horseshoe or U shaped cleft associated with micrognathia, glossoptosis and airway compromise (Figure 1b).

There is some variation in the distribution of cleft types reported worldwide (World Health Organisation Expert Committee, 2002). The most common presentation involving both the lip and palate is unilateral cleft lip and palate (UCLP). In these cases the defect extends through the lip, alveolus, hard and soft palate. Newborn infants with UCLP present with varying degrees of distortion of the greater and lesser segments and their relationship to each other. Usually the anterior part of the major segment rotates outwards whilst the anterior part of the lesser segment is contracted inwards. The overall configuration results in a somewhat distorted and asymmetrical appearance of the palate and the alveolar segments (Figure 1c). In bilateral cleft lip and palate (BCLP) the lip defect extends though the lip and alveolus on both sides and through the hard and soft palate. With the premaxilla attached only to the columella of the nose, the premaxilla usually protrudes (Figure 1d).
Figure 1: Diagrammatic representation of cleft lip (a), cleft palate (b), unilateral cleft lip and palate (c) and bilateral cleft lip and palate (d)
1.1.3 Classification of cleft lip and/or palate

The variable presentation of this condition has resulted in numerous approaches to its classification (Kemahan and Stark, 1958; Harkins et al., 1960; Pfeifer, 1964; McCabe, 1966; Kemahan, 1971; Elsahy, 1973; Millard, 1976; Friedman et al., 1991; Davison et al., 1998). In the UK, data are currently collected using the system devised for the Craniofacial Anomalies Register (CARE), recently renamed the Craniofacial Anomalies Network (CRANE) (Figures 2 and 3) (Craniofacial Anomalies Network, 2003).

This system describes the cleft as complete or incomplete in relation to its position and extent. For example Figure 2 shows an infant with a left complete unilateral cleft of the lip and palate (UCLP). In contrast Figure 3 shows an infant with complete cleft of the hard palate and soft palate (ICP).
CLASSIFICATION OF CLEFT: (I - Incomplete, C - Complete. Please circle as appropriate)

LIP
ALVEOLUS
HARD PALATE
SOFT PALATE
VOMER ATTACHED TO HARD PALATE

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SUBMUCOUS CLEFT:

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PIERRE ROBIN
SIMONARTS BAND
FORME FRUSTE

SUMMARY OF CLEFT TYPE:

| COMPLETE UNILATERAL CLEFT LIP AND PALATE |

Figure 2: Photographs and CARE classification of an infant with complete unilateral cleft of the lip and palate.
CLASSIFICATION OF CLEFT: (I - Incomplete, C - Complete. Please circle as appropriate)

RIGHT | MID LINE | LEFT
--- | --- | ---
LIP | I | C | I | C | I
ALVEOLUS | I | C | I | C | I
HARD PALATE | I | C | I | C
SOFT PALATE | I | C
VOMER ATTACHED TO HARD PALATE | Y | N
SUBMUCOUS CLEFT | Y | N | Type:
PIERRE ROBIN | Y | N
SIMONARTS BAND | L | R | FORME FRUSTE | L | R
SUMMARY OF CLEFT TYPE
ABNORMALITIES/SYNDROME

isolated complete cleft of the hard and soft palate

Figure 3: Photographs and CARE classification of an infant with an incomplete cleft of the hard palate and complete cleft of the soft palate.
1.2 Management of cleft lip and/or palate

Cleft lip and palate has far reaching implications for the child, from birth through to maturity. A range of difficulties may be experienced over the child's life including medical based problems such as impaired facial growth, dental anomalies, middle ear problems and hearing impairment (Habel et al., 1996). General body growth may be affected, with failure to thrive frequently reported (Spriesterbach et al., 1973; Paradise et al., 1974; Ranalli and Mazaheri, 1975; Avedian and Rubery, 1980; Bryan Jones, 1988; Felix-Schollaart et al., 1992). Speech development is also at risk and language development may be delayed (Bzoch, 1989; McWilliams et al., 1990; Chapman and Hardin, 1990; Golding-Kushner, 1995; LeBlanc, 1996; Neiman and Savage, 1997; Russell and Harding, 2000). Speech problems associated with velopharyngeal insufficiency may also occur and require repeated surgical intervention (McGrath and Anderson, 1991; Leeper and Charles, 1993; Golding-Kushner et al., 1995). Developmental problems (Starr et al., 1977b; Fox et al., 1978; Jocelyn et al., 1996; Neiman and Savage, 1997) and poor educational achievement are reported (Spriesterbach et al., 1973; Lahti et al., 1974; Richman, 1976; Richman, 1978b; Richman, 1978c; Broder and Strauss, 1993; Broder et al., 1998). In isolation or in combination, these problems may have significant psychosocial consequences for the child and their family, not least impacting on psychosocial wellbeing (Richman and Harper, 1978; Richman, 1978a; Richman, 1978b; Broder and Strauss, 1991; Kapp-Simon et al., 1992; Bradbury, 2001).

The management of the child with CL and/or P is lengthy, with both surgical and non surgical interventions requiring the specialist involvement of a large interdisciplinary team, and with considerable disruption to the child and their family's life (Sandy et al., 1998; Bearn et al., 2001; World Health Organisation Expert Committee, 2002).
There are many areas of controversy with regards to management and best practice. One such area is the highly emotive area of feeding (Adams et al., 1999; Oliver and Jones, 1997), which is the purpose of the investigation presented in this thesis.

1.3 Feeding issues

As many as 63% of infants born with CL and/or P experience feeding difficulties (Clarren et al., 1987). Among the consequences of feeding difficulties are poor growth and development (Paradise et al., 1974; Ranalli and Mazaheri, 1975; Avedian and Rubery, 1980; Jones, 1988; Felix-Schollaart et al., 1992; Lee et al., 1997). Altered mother-child bonding (Speltz et al., 1994; Speltz et al., 1997; Coy et al., 2002), decreased emotional well-being of the family, and increased burden of care (Adams et al., 1999) are further risks. Another serious consequence although not widely reported in children with CL and/or P, is aspiration which may lead to recurrent chest infections (Zickefoose, 1957; Taniguchi and Moyer, 1994).

In the United Kingdom surgical intervention to repair CL and/or P usually occurs within the first year of life (CSAG Report, 1998). Infants with CL and/or P are usually fed with an unrepaired cleft palate for at least the first six months of their life. Despite cleft teams having protocols for the establishment and monitoring of feeding, parents continue to report difficulties feeding their infants (Trenouth and Campbell, 1996b; Oliver and Jones, 1997).

Given the many medical, psychological and social implications of impaired feeding, and with parents of infants with CL and/or P reporting ongoing feeding difficulties in the period up to palate repair, prompt and appropriate management of feeding is a priority.

Although the literature contains little information about the physiological basis of feeding in infants with CL and/or P, many techniques are used to
facilitate feeding. The techniques used include the use of adaptive equipment (Brophy, 1923; Brophy, 1927; Kelly, 1971; Paradise and McWilliams, 1974; Styer and Freeh, 1981; Clarren et al., 1987; Barone and Tallman, 1998; Scheuerle, 1998), adaptive positioning (Lifton, 1956; Zickefoose, 1957; Tisza and Gumpertz, 1962; Paradise and McWilliams, 1974; Wolf and Glass, 1992; Arvedson, 1993; Glass and Wolf, 1999) and strategies designed to control the pace of feeding (Richard, 1991; Brine et al., 1994).

An additional technique used to facilitate efficient feeding is pre-surgical orthopaedics (Lifton, 1956; Williams et al., 1968; Razek, 1980; Balluff and Udin, 1986; Osuji, 1995).

1.4 Pre-surgical orthopaedics

Pre-surgical orthopaedics (PSO) or baby plates have been used since the seventeenth century. The process involves realigning the characteristically distorted and misplaced alveolar segments of infants with UCLP by external forces. Early methods involved applying external pressure to the cleft maxilla with the aid of facial binding, taping or the use of strapping attached to bonnets (Winters and Hurwitz, 1995). Brophy (1927) described the use of a more invasive technique, whereby wire was placed through both ends of the alveolus and was gradually tightened. The modern day technique involves the fitting of acrylic plates (McNeil, 1950; Burston, 1958; Robertson, 1983). These plates are custom made and fabricated on plaster models from dental impressions (Figure 4). There may be additional use of strapping (Brogan, 1986), internal fixation of the plate with screws (Mylin et al., 1968), or regular adjustment or activation of the plate (DiBiase and Hunter, 1983; Hotz, 1990; Ball and DiBiase, 1995). Plates where adjustment or activation is undertaken are known as active plates (Figure 4). In infants with BCLP, where distortion of the pre-maxilla is prominent, PSO may be used to expand the posterior segments in preparation for retraction of the premaxilla. Retraction is achieved by elastic strapping, applying pressure to the prominent premaxilla.
(Ross and MacNamera, 1994). Infants with ICP do not present with the alveolar distortion seen in those with UCLP. Consequently plates fitted for these infants differ from the active plates by simply occluding the cleft and lying passively supported by the alveolar ridges (Figure 5) (DiBiase and Hunter, 1983; Hotz, 1990; Ball and DiBiase, 1995). The basis of using these passive plates is to prevent the tongue from sitting up in the cleft and possibly discouraging lateral shelf growth.

Figure 4: “Active” baby plate (DiBiase and Hunter, 1983)
The proponents of the PSO technique claim that it has several benefits including minimising the risk of surgical breakdown by reducing the strain on newly operated tissues (Winters and Hurwitz, 1995), retracting the premaxilla in cases of BCLP (Winters and Hurwitz, 1995), narrowing the cleft size (Brophy, 1927) and realigning the maxillary alveolar process, thus encouraging more normal facial growth and dental arch development (McNeil, 1950; Burston, 1958). It is also suggested that PSO may contribute to non-surgical closure of palatal defects by stimulating the “growth impulse” and reducing the need for orthodontic treatment (McNeil, 1950; McNeil, 1956; Burston, 1958). Furthermore it is hypothesised that PSO may facilitate speech development (Stuffins, 1983; Konst et al., 1999) and improve feeding (Lifton, 1956; Burston, 1958; Williams et al., 1968; Balluff and Udin, 1986; Goldberg et al., 1988; Winters and Hurwitz, 1995).

Several studies have attempted to investigate these suggestions (Fish, 1972; O’Donnell et al., 1974; Huddart and Crabb, 1977; Mars et al., 1992; Ross and MacNamera, 1994; Hathorn et al., 1996; Chate et al., 1997; Kuijpers-Jagtman
and Prahl-Andersen, 1997; Konst et al., 1999). These studies were however scientifically flawed (Winters and Hurwitz, 1995). Only one study (Dutchcleft) has evaluated the effect of PSO using scientifically sound design. This multi-centre randomised control trial investigated the effects of PSO on surgical, orthodontic, feeding, growth and speech and language outcomes in children with UCLP. Feeding outcomes have not been reported yet but improved speech development and temporarily improved maxillary arch dimensions prior to surgical repair of the soft palate are shown (Kuijpers-Jagtman and Prahl-Andersen, 1997; Severens et al., 1998; Konst et al., 1999; Konst et al., 2000; Kuijpers-Jagtman et al., 2001; Konst, 2002).

Pre-surgical orthopaedics may be beneficial but alternatively they may cause unnecessary and unproductive disruption (Winters and Hurwitz, 1995; World Health Organisation Expert Committee, 2002). At present, use of PSO is dependent on individual preference and varies from unit to unit.

Since 1995 there has been one surgeon operating at St Andrews Centre for Plastic Surgery, Mid Essex NHS Trust (St Andrews) and Great Ormond Street Hospital NHS Trust (GOSH). Each centre had differing practices with regards to the use of PSO; St Andrews used PSO routinely and GOSH rarely. Given that all other aspects of care were standardised across the two sites and that one surgeon performed all primary surgery at both sites, there was an ideal opportunity for a multidisciplinary study investigating the effects of PSO. Both sites were amenable to changes in protocol to allow randomisation of infants to PSO or not, rather than following the standard protocol. The two centres therefore implemented a randomised control trial designed to investigate the effects of PSO on facial growth, facial appearance, surgical outcomes, speech development and feeding. Subsequently the two centres were amalgamated and recognised as the North Thames Regional Cleft Unit (NTRCU). This thesis reports the first stage in this investigation: the effects of PSO on infant feeding patterns and efficacy during the first year of life in infants with either UCLP or ICP.
1.4 Thesis Outline

The structure of the thesis is as follows.

Chapter 1 provides the background of the investigation and introduces the arguments regarding the use of PSO.

Chapter 2 is a review of the impact of poor feeding, the understanding of the physiological basis of the feeding patterns and management of feeding in the cleft lip and/or palate population. Factors affecting feeding in normal infants are discussed. While there is broad knowledge about normal infant feeding, there are areas which are poorly investigated because of the ethical implications of using invasive assessment techniques in normal healthy infants. It is generally accepted that extrapolating information from normal adult studies to paediatrics is sometimes appropriate and this is discussed.

Chapter 3 describes approaches to the assessment of feeding and swallowing, providing a basis for the assessments used in this investigation.

Chapter 4 covers the development of the Great Ormond Street Measurement of Infant Feeding (GOSMIF) and the results of it’s application to a normal cohort.

Chapter 5 documents the research questions and hypotheses addressed by the proposed trial and outlines the trial methodology.

Detailed results of the study are presented in Chapter 6.

A discussion of the results of the study and possible explanations for these are given in Chapter 7. Problems encountered during the study are also presented here. This is followed by a discussion of the clinical implications of the findings of the trial and conclusions.
2.1 Feeding in the cleft lip and palate population

Feeding difficulties in the cleft lip and palate population were first reported in 1619 by Fabricius of Aquapendente who recognised that infants with cleft lip and palate were unable to suck and often died of malnutrition (Jones, 1988).

Most of the literature focuses on the management of feeding difficulties rather than the underlying physiology (Zickefoose, 1957; Tisza and Gumpertz, 1962; Pashayan and McNab, 1979; Styer and Freeh, 1981; Martin, 1983; Clarren et al., 1987). The first published descriptions of the nature of feeding problems were by Zickefoose (1957) and Tisza and Gumpertz (1962), and since then there have been only three further reports (Shelton et al., 1966; Brogan et al., 1987; Clarren et al., 1987). In addition there are several summary articles providing an overall description of the feeding patterns (Spriesterbach et al., 1973; Wolf and Glass, 1992; Glass and Wolf, 1999; Arvedson and Brodsky, 2002).

The study of Zickefoose (1957) was designed to evaluate the eating habits and problems of children with CL and/or P and was carried out by a nutrition consultant. Parents of 58 children aged between one and a half and six years of age with CL and/or P were interviewed about their child's feeding and diet history. The main problems in infancy, which were reported included: insufficient suction to extract milk from the nipple with most children learning to "chew" the nipple; excessive air intake resulting in the need for "several burplings during the feeding session"; difficulty for both babies and
older children in taking liquids without choking; mothers' fear of feeding; "annoyance from acid and spicy foods"; refusal of fizzy drinks; trouble with sticky or pasty foods" getting stuck in the cleft palate; difficulty chewing, reportedly due to poor teeth; nasal regurgitation; altered social interaction with some families refusing to eat in public due to embarrassment; prolonged use of soft foods and refusal of puree foods; long feeding times with some children "giving up and refusing to finish their meals". The authors reported that the number of chest infections in these children decreased after cleft repair. No information as to the frequency or extent of each of the problems is given.

The validity of this study is questionable. The parent interviews were relatively ad hoc with no set of questions reported. The interpretation of the parental responses might therefore be biased. Research suggests that maternal ability to recall feeding events accurately is dependent on the time since the occurrence (Launer et al., 1992). These parents were being asked to recall feeding difficulties which had occurred up to six years previously. In addition there was no control group.

Tisza and Gumpertz (1962) interviewed parents of infants and observed the infants feeding (numbers not specified). These infants were unable to generate the negative pressure required to feed from a bottle. The authors discussed this inability to suck in terms of "squeezing and milking". They stated that this process is "obviously different from normal sucking" and commented on the changed function of the lip and intra-oral structures, especially the tongue. It is reported that the infants used the back of their tongue to occlude the cleft and that they required frequent pauses during their feeds to "prevent nasal regurgitation" which interfered with the rhythm of feeding and led to increased frustration. Although Tisza and Gumpertz described that they observed infants feeding, they did not present a formalised way of evaluating their observations. No information about the
questions used in the interviews or the number of people involved in the data collection was given.

Some years later Clarren et al (1987) described clinical parameters of feeding in terms of cleft type and the use of successful feeding devices or techniques. They assessed 143 infants with varying cleft types over a 5-year period and reported the results of 113 of these (CL & P, n=53; ICP, n=17; Submucous Cleft, n=6; Mild PRS not requiring airway intervention, n=23; CL, n=14). All infants were assessed by informal observation with regard to their ability to generate negative intra-oral pressure and to move the tongue against the nipple. It was reported that children with clefts generally swallow normally but do not suck normally. These observations were related to cleft type and summarised in Table 1.

<table>
<thead>
<tr>
<th>Cleft type</th>
<th>Ability to generate negative intra-oral pressure during sucking*</th>
<th>Ability to produce mechanical movements/compression during sucking*</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cleft lip &amp; palate</td>
<td>-</td>
<td>-/+</td>
</tr>
<tr>
<td>Cleft palate</td>
<td>-/+</td>
<td>+</td>
</tr>
<tr>
<td>Cleft soft palate</td>
<td>-/+</td>
<td>+</td>
</tr>
<tr>
<td>Pierre Robin Sequence</td>
<td>-/+</td>
<td>-</td>
</tr>
<tr>
<td>Cleft lip</td>
<td>-/+</td>
<td>+</td>
</tr>
</tbody>
</table>

* + = present, - = absent, +/- = partial

Table 1: Table taken from Clarren et al (1987)
The findings given in Table 1 were reportedly based on clinical observations. However the generation of negative intra-oral pressure is not visible clinically. Similarly, while some of the mechanical aspects of feeding such as lip seal and jaw elevation are visible, compression of the teat by the tongue is not. No details are given about the staff involved in the feeding assessments, the ages of the children at assessment or the tools used to describe the feeding patterns. As data were collected over a five-year period, staff may have changed and may have had differing levels of expertise and experience.

Videofluoroscopic assessment of dysphagia allows visualisation of the oral and pharyngeal stages of feeding, and is widely regarded as the “gold standard” assessment of dysphagia (Section 3; 4). It has been reported in the assessment of children with CL and/or P in two studies only. The first (Shelton et al., 1966) assessed feeding in school age children with CL and/or P and the second (Brogan et al., 1987) looked at infants with cleft lip and palate.

Shelton et al (1966) used videofluoroscopic assessment to evaluate swallowing patterns in school-aged children. The study cohort consisted of 32 children aged between 6.5 years, and 12.11 years. Seventeen children had surgically repaired CL and/or P, 4 had palatal incompetence (no further details given), 1 child had a repaired cleft palate and 10 children had unrepaired clefts of the palate, which were managed with an obturator. Each child was given a 55-ml bolus of diluted barium solution to swallow. The studies were reviewed by an investigator who judged whether the bolus entered the nasopharynx and the pattern of oral and pharyngeal stage swallowing. Each pattern was characterised by “a period of freefall of the bolus” and by an interruption of the stripping action of the tongue against the roof of the mouth. The bolus was lifted to the alveolar ridge and forced back to the middle of the hard palate by the occlusion of the tongue against the hard palate. The tongue movements then ceased and the bolus fell over the posterior tongue into the valleculae and in several cases over the tip of the epiglottis suggesting
laryngeal penetration. Three distinct swallow patterns were identified in relation to transfer of the bolus through the pharynx. Pattern 1 was considered a near normal pattern with the tongue contacting the pharyngeal wall at the level of cervical vertebrae 1 and 2 (C1/C2). Pattern 2 was characterised by a similar movement pattern except that the pharyngeal wall actively moved to meet the tongue at the level of C1/C2. In contrast, pattern 3 was characterised by a lack of contact between the tongue and hard palate (or palatal obturator) but similar movement to patterns 1 and 2 with the pharyngeal wall meeting the tongue but at a lower level (C2/C3). Unfortunately the trial cohort was heterogeneous with a variety of cleft types, structural status and a wide range of ages. The authors do not report the number of children demonstrating each pattern, nor do they relate the pattern types to structural status (e.g. post surgical repair of the cleft, managed with obturators). However this study was the first to objectively describe the physiological process of feeding in children with repaired clefts of the lip and/or palate and in those fitted with obturators.

Some years later, Brogan et al (1987) investigated 7 infants aged 4 days to 48 weeks with cleft lip and palate and 2 non-cleft infants using videofluoroscopy. The study did not consider feeding patterns as a whole but focused on tongue positioning. Four groups were studied. The first consisted of 2 non-cleft infants who were being investigated for bowel problems. The second included 2 infants with unrepaired clefts of the lip and palate (UCLP and BCLP), neither of whom had been managed with PSO. Group 3 consisted of 1 infant with unrepaired cleft lip and palate, who was being managed with PSO and the fourth group included 4 infants (3 UCLP, 1 BCLP) who had undergone PSO treatment and lip and alveolus repair. Tongue positioning was rated from videofluoroscopic studies of the infants during a short feed. The approach to rating tongue positioning was not described but it was reported there was no significant difference between any of the groups. The authors concluded that infants with cleft lip and palate have normal tongue movements. The main weaknesses of this study were the small sample size,
the lack of homogeneity within the cohort in terms of age, the lack of consideration of developmental status, cleft type or use of PSO. In addition there was a lack of detail about the methodology for rating tongue positioning, such as who rated the studies and reliability checks.

To summarise, there is little information about the physiology of feeding in the cleft population. Much of what is available is based on parent report, subjective observations and anecdotal experience. Interviews have lacked structured formats and clinical observations are poorly described. When more objective assessment techniques, such as videofluoroscopy have been used there have been limitations particularly with regard to the homogeneity of the groups (mixed ages of subjects and cleft types) and few evaluations of the reliability of measures used.

2.2 Consequences of impaired feeding in the cleft lip and palate population

The consequences of feeding problems in infants in the general population are well described and may include developmental repercussions such as impaired growth and general development, psychological and social problems such as altered mother child bonding and behavioural feeding problems, and medical complications for example recurrent chest infections, potentially leading to chronic respiratory problems and in severe cases death. Within the CL and/or P population, these problems are also evident (Zickefoose, 1957; Tisza and Gumpertz, 1962; Spiesterbach et al., 1973; Paradise et al., 1974; Ranalli and Mazaheri, 1975; Di Scipio et al., 1978; Avedian and Rubery, 1980; Di Scipio and Kaslon, 1982; Felix-Schollaart et al., 1992; Speltz et al., 1994; Lee et al., 1997) and have been suggested to be linked with poor feeding (World Health Organisation Expert Committee, 2002).
2.2.1 Growth

The most widely reported consequence of feeding difficulties in the cleft lip and palate population is impaired general body growth. Several studies have examined the growth patterns of children with CL and/or P (Spriesterbach et al., 1973; Paradise et al., 1974; Ranalli and Mazaheri, 1975; Avedian and Rubery, 1980; Felix-Schollaert et al., 1992; Lee et al., 1997). The results have been conflicting, with some identifying failure to thrive and others disputing this. While some studies address the variable of feeding management, others do not therefore making it difficult to compare findings across the studies.

There is some discrepancy in reports of the birth weight of infants with CL and/or P. They have been reported to be below the norm (Avedian and Rubery, 1980), of near average birth weight (Lee et al., 1997), or slightly longer and heavier than the norm (Ranalli and Mazaheri, 1975).

Several studies have reported that children with CL and/or P may not grow as well as the non-cleft population. It has been suggested this may be the result of feeding technique (Ross and Johnson, 1972; Ranalli and Mazaheri, 1975; Avedian and Rubery, 1980); increased incidence of infections (Ross and Johnson, 1972; Seth and McWilliams, 1988); surgical procedures (Ranalli and Mazaheri, 1975; Jensen et al., 1983; Seth and McWilliams, 1988) and/or growth hormone deficiency (Rudman et al., 1978; Bowers et al., 1988).

Avedian and Ruberg (1980) published the preliminary results of a retrospective study of 37 children (22 boys and 15 girls) in which they examined growth in the first six months of life. Only children with CL and P were included. Data were collected from the parents who were asked to provide weight records and the medical history for their child's first 6 months of life. The median birth-weight for the cohort was at the 30th percentile. By the first month the median had dropped to the 20th percentile and at three, four and five months of age had returned to the 30th percentile. However,
only 17 of the 37 children followed this pattern. Eight children did not drop percentiles. Another 12 remained consistently “below the norm of the 50th percentile” and failed to return to their birth percentile at 6 months of age. The results of this study may be explained by the teams’ management of the feeding problems where the protocol was to intervene only in cases where children were experiencing feeding difficulties. Unfortunately how they defined a feeding difficulty is not described. The number of children who required intervention and the nature of this are not reported. Similarly, the extent of the cleft deformity and the presence of syndromes are not reported.

It has also been suggested that cleft type and extent may influence growth (Ranalli and Mazaheri, 1975; Jensen et al., 1983; Jones, 1988; Lee et al., 1997). Ranalli and Mazaheri (1975) examined the relationship in a study of growth patterns in 279 children with different cleft types. The study population was taken from serial patient data files, with subjects ranging from 6 months to 6 years of age. They found no significant differences between cleft types with regard to growth, and no significant differences between the cleft subjects and non-cleft controls. They concluded that infants born with CL and/or P are born a little heavier and longer than the control group, but following birth begin to show a lag. However by 3 years of age the children with CL and/or P, tended to “catch up” to the norm.

In contrast, Lee et al (1997) found cleft type had the greatest influence on the degree of failure to thrive. They studied 83 children including those with syndromic clefts. Data were collected by interview or postal questionnaire and included history of feeding problems, feeding management techniques, and background information. Medical notes were reviewed to retrieve medical and surgical information and measurements of parents’ weight and height were taken. Sixty three percent of the children experienced feeding difficulties at some point, with a higher incidence in children with cleft palate only (CP). Growth was considered from the perspective of the “thrive index” which showed that all groups (cleft palate only (CP), cleft lip only (CL) and unilateral
cleft lip and palate (UCLP)) had decreases just prior to surgery, with the CP group showing the largest fall. Similarly, a significantly higher proportion of the CP group manifested failure to thrive than the other CL and CLP groups. The authors concluded that while all children with clefts of the lip and/or palate showed disrupted early growth, those with isolated clefts of the secondary palate and/or associated syndromes were more severely affected. However they failed to acknowledge that the higher proportion of poor growth in the infants with isolated clefts of the palate might be explained by increased incidence of associated syndromes, known to be particularly common in infants with cleft palate only.

Felix-Schollaart et al (1992) carried out a study to evaluate the growth patterns of children with and without cleft lip and palate and factors which might explain any observed differences. They compared 45 children with non-syndromic clefts, (12 cleft lip, 20 cleft lip and palate, 13 cleft palate only) and 50 healthy infants born at the same hospital. They were examined on a 3 monthly basis for the first year and 6 monthly for the following 18 months. Data were also collected on gastroenterological feeding difficulties, intestinal disorders, occurrence of airway infections, surgery, gestational age and birth order. There were few differences in growth between the cleft type groups CL, CLP, CP and the controls. The occurrence of gastroenterological feeding difficulties, intestinal disorders and airway infections did not differ significantly across the four cleft groups. The authors concluded that these conditions do not account for the differences in growth reported in this population.

The available research shows that children with CL and/or P have delayed growth in the early years but by 3 years of age the majority have “caught up” to the norm. However there is little agreement about the factors contributing to this delayed growth, with feeding method, metabolic variability, growth hormone deficiency and airway infections being implicated.
2.2.2 General development

General development in children with cleft lip and palate may be different to that of their peers (Fox et al., 1978; Jocelyn et al., 1996; Neiman and Savage, 1997).

Although the relationship between feeding and general development has not been directly investigated in the cleft population a link has been shown in other groups including developing countries and social deprivation (Chase, 1970; Chavez et al., 1975; Lasky et al., 1981; Waber et al., 1981; World Health Organisation, 1983; Soewondo et al., 1989; Grantham-McGregor et al., 1991; Husaini et al., 1991; Lozoff et al., 1991; Engle et al., 1992; Martorell et al., 1992; Idjradinata and Pollitt, 1993; Pollitt, 1993; Pollitt et al., 1993; Grantham-McGregor et al., 1994; Pelletier, 1994; Gorman, 1995; Grantham-McGregor, 1995; Levitsky and Strupp, 1995; Meeks-Gardner et al., 1995; Pollitt, 1996; Grantham-McGregor et al., 1997; Martorell, 1997; Walker et al., 1998; Department of Child and Adolescent Health and Development, 1999; Arvedson and Brodsky, 2002). It might therefore be suggested that there may be a connection between the reported failure to thrive in infants with CL and/or P and developmental delay (World Health Organisation Expert Committee, 2002).

Several groups have evaluated the development of children with CL and/or P in the pre-school years. The results have been conflicting and inconclusive. Some suggest there may be trends associated with cleft type (Neiman and Savage, 1997) but others dispute this (Starr et al., 1977a). These discrepancies may be due in part to a greater awareness of and earlier diagnosis of syndromes associated with CL and/or P in recent years thus making comparisons between studies less valid. Several cross-sectional studies have been carried out using standardised, published instruments. Plotkin et al (1970) found no significant differences in the development of children with CL and/or P. Similarly Starr et al (1977a) and Plotkin et al (1970) found that
children with CL and/or P were functioning within normal limits at 6 and 12 months of age. Fox et al. (1978) assessed 24 infants with CL and/or P, between the ages of 2 and 33 months, and compared them to an aged matched control group. They found the children with CL and/or P tended to have a developmental lag of 1 to 3 months.

Neiman and Savage (1997) recently carried out the largest study by examining the development of 186 infants and toddlers with non-syndromic clefts. Parents completed questionnaires (response rate 78%) and comparisons were made with an unmatched control group. A greater proportion of infants with CL and/or P demonstrated at-risk or delayed development at 5 months of age. Infants with cleft lip only demonstrated at-risk or developmental delay for the full-scale score and motor domain scores. Infants with cleft lip and palate demonstrated at-risk or delayed full-scale, motor, self-help and cognitive scores. Infants with isolated cleft palates exhibited delayed or at-risk motor, self-help and language scores. However by 13 months of age, the majority of the delays had resolved, with just over a third of toddlers with isolated cleft palate demonstrating at-risk or delayed development and all others being within normal limits.

Jocelyn et al. (1996) carried out a prospective, longitudinal controlled study of 16 infants with cleft lip and palate (CL & P), compared with controls matched for age, sex, race, socio-economic status and birth order. They found the children with CL & P had significantly lower scores on tests of cognition, comprehension and expressive language compared with the control group at the ages of 12 and 24 months.

IQ testing becomes more practical and reliable as children approach school age (Sattler, 1992), and several studies have investigated the general intelligence of children with CL and/or P. These reports suggest that although IQ levels fall within the normal range they are significantly lower on
average than in their control-matched non-cleft peers (Goodstein, 1961; Ruess, 1965; Smith and McWilliams, 1968).

Children with CL and/or P obtain lower school achievement scores than their control, IQ-matched peers (Richman, 1976; Richman and Harper, 1978; Broder and Strauss, 1993). Spristerbach et al, (1973) found that 31% of children with CL and/or P were older when they started school compared with 16% of the controls. Forty three percent of the children with CL and/or P were reported to be behind in their grade placement compared to 20% of the control subjects. The parents of children with CL and/or P were reported to have lower expectations of their children continuing formal education at college. More recently Broder et al (1998) found that in a US population, 46% of the school-aged subjects had learning disability (where national rates for non cleft children are 20-25%). Similarly it has been reported that fewer children with CL and/or P completed secondary education and university study than the national average (Lahti et al., 1974).

It has been suggested this lower school achievement may be associated with children with CL and/or P being perceived by their teachers as being intellectually delayed and inhibited (Richman, 1978a; Richman, 1978b). Similarly it has been suggested the negative reaction of the peers of children with cleft lip and palate may undermine their self-esteem which may in turn contribute to underachievement (Schneiderman and Harding, 1984; Kapp-Simon et al., 1992; Broder, 1994).

Many possible explanations have been suggested to account for, or at least, contribute to developmental problems in the cleft population. These include hearing problems, speech and language problems, altered interaction patterns, low self-esteem and confidence and lowered expectations of children with cleft. However although it is known early feeding problems contribute to failure to thrive, malnutrition and poor growth in this population, and although the literature suggests malnutrition in the first years of life
contributes to developmental delay (Pollitt et al., 1993; Gorman, 1995; Pollitt, 1996; Pollitt et al., 1996; Department of Child and Adolescent Health and Development, 1999), there are no studies investigating the relationship between these factors in the cleft population.

2.3 Management of feeding problems in the cleft lip and palate population

Given the known feeding difficulties in the cleft lip and palate population and their suspected consequences, management of feeding is a high priority. Because of the poor understanding of the basis of the feeding problems numerous techniques designed to provide “symptomatic relief” have been described.

The literature in this area can be grouped into four main areas: positioning strategies, use of assistive feeding devices, breastfeeding and the use of presurgical orthopaedics.

2.3.1 Positioning

Some of the earliest strategies relate to the use of positioning (Lifton, 1956; Zickefoose, 1957; Tisza and Gumpertz, 1962; Styer and Freeh, 1981; Glass and Wolf, 1999). Both Tisza et al (1962) and Zickefoose (1957) advocated the feeding of infants in an upright position to minimise nasal regurgitation. Glass and Wolf (1999) suggested that feeding in the upright position allows gravity to facilitate transfer of the bolus through the oral cavity and pharynx. A third possible advantage is that the upright position may foster bolus movement away from the Eustachian tubes, minimising the risk of otitis media. Upright positioning may facilitate frequent burping and minimise the frequency of episodes of positing which are thought to expose the eustachian tubes to refluxed materials. These practices have no objective validation but
nevertheless are frequently recommended (Wolf and Glass, 1992; Arvedson, 1993; Glass and Wolf, 1999).

2.3.2 Assistive Feeding Devices

The use of “assistive” feeding devices is based on the assumption that infants with CL and/or P are “unable to achieve adequate lip seal around the nipple or teat” and are “unable to generate adequate intra-oral pressures to take milk from the breast or bottle” (Zickefoose, 1957; Tisza and Gumpertz, 1962; Glass and Wolf, 1999). These devices therefore focus on facilitating delivery of milk into the infant's mouth. No attention is paid to the pharyngeal and oesophageal stages of the swallow, which are assumed to be unaffected. Some techniques allow the infant to bypass the oral stage of sucking completely, while others supplement this sucking process.

Historically, and still today in the developed world, infants with CL and/or P are fed with an assortment of equipment including pipettes, syringes, spoons and cups (Lang et al., 1994). Tisza (1962) and Zickefoose (1957) describe the use of equipment such as medicine droppers, the Brecht Feeder and a variety of “home-made” devices. All of these devices were means of delivering milk into the baby’s mouth (Zickefoose, 1957). Some years later, soft plastic bottles were advocated. These bottles come in various forms and facilitate feeding by allowing the feeder to assist the infant’s sucking by squeezing the bottle and reducing the need for generation of negative intra-oral pressure to deliver milk into the infant’s mouth. The variations of the soft bottles described included the "Mead Johnson" feeder and “Soft Plas” feeder (Kelly, 1971; Paradise and McWilliams, 1974; Styer and Freeh, 1981; Clarren et al., 1987; Scheuerle, 1998). The Playtex or Evenflo feeders utilise a rigid shell with a collapsible inner bag, allowing the mother to squeeze the inner collapsible bag in much the same way as a soft bottle (Paradise and McWilliams, 1974; Barone and Tallman, 1998; Scheuerle, 1998).
Haberman (1988) developed a feeding system designed to express milk into the infant's mouth with compression of the teat only, i.e. without the generation of negative intra-oral pressure. The bottle consists of an extended teat or nipple and a one-way valve system, which allows milk to flow into the teat but not back into the bottle. As a result the need for generation of negative intra-oral pressure, to facilitate transfer of milk into the infant's mouth is reduced.

There has also been debate about the types of teats or nipples that may facilitate feeding in the cleft population (Brophy, 1923; Pashayan and McNab, 1979; Styer and Freeh, 1981).

Teats designed for premature babies have been used based on the principle that they generally have larger holes and thus require less generation of intra-oral pressures to transfer milk (Styer and Freeh, 1981).

Martin (1983) described a “gravity flow” nipple, which drops milk into the infant's mouth, bypassing the need for sucking.

The use of “cross cut” teats has also been advocated, but there is debate about their safety. Cross cut teats work by remaining closed until the teat is compressed. It is hypothesised that by creating a cross cut in a teat rather than one or more simple holes it is possible to allow the infant more control over the flow rate of milk. Milk can be ejected into the mouth by purely compressing the teat (Pashayan and McNab, 1979; Styer and Freeh, 1981). Such teats are reported to prevent “flooding” when the flow rate of the milk is faster than the infant is able to cope with (Pashayan and McNab, 1979; Styer and Freeh, 1981). On the other hand, it has been suggested that these teats are difficult for infants to control. The amount of milk expelled on compression of the teat can be variable making it difficult for the infant to coordinate sucking, swallowing and respiration (Glass and Wolf, 1999). When used in conjunction with soft bottles the infant can be “flooded” if
compression of the bottle is not accurately timed with compression of the
teat resulting in coughing, choking and in extreme cases aspiration.

Pashayan and McNab (1979) carried out a study to evaluate the effectiveness
of cross cutting teats and to develop guidelines for the cross cutting
procedure. The study included a group of infants with different cleft types (4
infants with UCLP, 2 infants with BCLP, and 6 infants with CP). The growth
of these infants was monitored over a 6 month period. All infants with UCLP
maintained their growth (as measured on standard growth charts), 1 of the 2
infants with BCLP also maintained growth along their birth weight percentile
and 5 out of the 6 infants with CP maintained growth. The authors
concluded the technique of modifying teats with a crosscut is a useful way of
feeding infants with CL and/or P. However these conclusions must be
regarded with caution as they present the findings of a survey of a small
group rather than a specifically designed study with a control group.

Others have suggested the modification of teats by either enlarging the holes
or creating additional holes (Kelly, 1971; Pashayan and McNab, 1979; Styer

There have been several studies which have evaluated the efficacy of “soft
bottles” to facilitate feeding with infants with CL and/or P.

Brine et al (1994) undertook a trial of 31 infants, 22 with UCLP and 9 with
CP and randomised them to one of two treatment arms. Eighteen infants
were fed with a soft bottle with long cross cut teat and thirteen were fed with
a standard bottle and standard cross cut teat. Outcome measures included
growth and nutritional assessment of energy and protein intake. Their results
showed that both techniques were useful in maintaining growth within
normal limits with no significant difference between the 2 groups. Inclusion
and exclusion criteria and the methodology were well documented. No
reliability information was reported. The researcher was not blinded to the
treatment arm, and it was assumed the carer had interpreted instructions
regarding the manner of feeding appropriately without the observer assessing this. It would have been interesting if rather than comparing these two feeding methods (with differing bottles and teats) the study had controlled for one of these variables. If, for example, the same teats were used in both groups information about whether the bottle type influenced success of feeding would have been available.

In 1999, Shaw et al presented the results of another, larger randomised controlled trial evaluating the usefulness of “squeezy bottles” compared with a standard “rigid bottle”. This study recruited 101 infants with cleft palate (+/- cleft lip) and randomised them to one of two groups, rigid bottle or squeezy bottle. The exclusion criteria and the randomisation procedure were well described. A group of the cohort were also allocated to be managed with PSO. Details of this allocation are not presented. Outcome was measured by growth measurements (up to 12 months) and the number of modifications to the teats required. It was reported infants in the “rigid bottle” group required significantly more modifications to teats (48%) than infants in the “squeezy bottle” group (8%). Anthropometric data suggested the growth patterns in the “squeezy bottle” group were significantly better than those in the “rigid bottle” group. The researchers concluded that the use of “squeezy bottles” might be a more effective management technique than “standard rigid bottles” for the cleft palate population. Unfortunately it was not reported whether all aspects of care were standardised, e.g. in terms of the amount, place or time spent with mother and infant and the amount and type of advice given. There were no reliability checks for outcome measurements and the researcher was not blinded to the treatment technique, allowing room for bias. There is no data reported about the outcome of this group of infants managed with PSO, however the authors suggested that PSO did not improve feeding efficiency.
2.3.3 Breast Feeding

The World Health Organisation (2001a; 2001b) recommends breast-feeding as the ideal feeding method for infants. Breast milk is said to be more easily digestible than formula, to provide protection against infection (including otitis media) and to minimise allergic responses. Breast-feeding is said to help develop the muscles in the facial area, leading to proper development and growth (La Leche News, 1972). It is also believed to encourage optimal growth (Danner, 1992), benefit neurodevelopment (Vestergaard et al., 1999) and promote better speech development (Broad, 1972). Breast-feeding facilitates maternal-infant bonding (Zetterstrom, 1999) and gives the mother feelings of satisfaction and reward. Additionally, it has been suggested breast feeding of infants with cleft lip and palate disguises the cleft to some extent, making the infant look "more normal" (Zetterstrom, 1999). As a result of all these factors many mothers of infants with cleft lip and palate want to breastfeed.

There is much debate about the efficacy of breast-feeding in CL and/or P, yet there are no studies that scientifically investigate it. While some teams report they have not found breast feeding to be an efficient means of ensuring adequate nutrition (Clarren et al., 1987; Glass and Wolf, 1999) others report the reverse with as many as 80% of mothers succeeding to some extent (Willis, 2000; Styer and Freeh, 1981; Clarren et al., 1987). However the majority of these latter reports are anecdotal. In addition, successful breast-feeding is poorly defined in these papers. While some authors imply the use of assistive techniques such as hand expression of breast milk into the infant's mouth, constitutes "successful" breastfeeding (Willis, 2000) it might better be described as "assisted" breast feeding. In addition, while it is acknowledged that the measurement of successful breast-feeding is difficult, there was no attempt to evaluate objectively the success of the techniques advocated in these studies by for example, comparing growth with non breastfed infants.
2.3.4 PSO and feeding

In common with much of the preceding literature about feeding problems in infants with CL and/or P, the literature regarding the use of pre-surgical orthopaedics in feeding is often anecdotal, or based on case studies. In addition, several authors make claims about the benefits of PSO without data to support their claims.

A number of groups report improved feeding efficiency (both bottle- and breast-feeding) with the use of PSO (Lifton, 1956; Williams et al., 1968; Balluff and Udin, 1986; Goldberg et al., 1988). Claims are made that the use of PSO eliminates slow and frustrating feeds (Jones et al., 1982; Balluff and Udin, 1986; Osuji, 1995) reduces choking episodes (Jones et al., 1982), improves growth (Goldberg et al., 1988) and improves parents psychosocial well-being (Razek, 1980; Jones et al., 1982). Others dispute this, reporting that the use of PSO does not improve feeding (Berkowitz, 1978; Pashayan and Lichtenstein, 1975; Choi et al., 1991) or that there is insufficient evidence to support any of the benefits (Pruzansky, 1964; Paradise and McWilliams, 1974).

Several hypotheses have been proposed as to why PSO improves feeding. It is suggested PSO provides a rigid opposing surface allowing compression of the teat or nipple (Osuji, 1995) and reduces ulceration of the nasal septum by teats, which can lead to pain (Huddart and Ziberman, 1977). In addition it is claimed PSO corrects the tongue position and improves the airway (Huddart and Ziberman, 1977; Osuji, 1995), restores the infant’s ability to generate intraoral pressures required for sucking (Schwenzer and Grimm, 1981; Hotz, 1983; Komposch, 1986; Kogo et al., 1997; Trankmann, 2000) and minimises food residue and stagnation of feed in the cleft (Osuji, 1995) thus reducing the unpleasantness of nasal regurgitation (Huddart and Ziberman, 1977; Jones et al., 1982). It is hypothesised that it prevents the tongue from exploring and widening the cleft (Razek, 1980; Osuji, 1995). Others claim
some infants do not like the feel of the cleft in the mouth and prefer the sensation of an intact palate (Huddart and Ziberman, 1977). Finally Razek (1980) suggested parents are reassured some course of active treatment has been undertaken.

The majority of these claims are made with no scientific basis.

Lifton (1956) reported seven cases studies of infants with CL and/or P, who were fitted with feeding plates. All infants were reported to have experienced feeding difficulties together with weight loss or poor weight gain. The infants wore their plates for feeding only. All infants were reported to gain weight immediately following fitting of the plates and gained sufficient weight to be discharged from hospital. It was hypothesised that the plates provided an opposing surface against which the infant could compress the teat during sucking.

Markowitz et al (1979) evaluated the use of PSO with regard to the length of hospital admission required to establish feeding. They presented a case series of 11 infants with CLP (6 UCLP, 1 BCLP, 4 ICP), seen over a 4 year period. It was reported that after PSO fitting, the infants were able to feed "normally" with a "standard nipple". They could be discharged within 4–7 days as compared to the routine 5-8 weeks for a healthy cleft infant, hence minimising costs, avoiding extended hospitalisation and inconvenience for the parents. It is important to note that this reported length of hospital stay for infants not fitted with PSO would no longer be considered acceptable practice and that the shortened hospital stay might also have been achieved with other assistive feeding options.

Balluf and Udin (1986) investigated whether PSO improved infant growth and whether parents felt the use of PSO had helped their infants' feeding. They evaluated 7 infants with either UCLP or BCLP, all of whom had feeding difficulties even after the use of positioning strategies and adaptive equipment (but not PSO). Following fitting of PSO, parents were asked to complete a
short questionnaire about the length of feeding, ease of feeding, their child's growth, benefits PSO provided and any problems experienced and their opinion about whether infants with CL and/or P should be fitted with plates. The average daily weight gain was also recorded. Following fitting of plates all infants showed improved daily weight gain. Five out of the 7 sets of parents reported feeding became easier. The parents reported the plate resulted in improved sucking, less nasal regurgitation and the elimination of NG feedings. All parents recommended the plate be fitted for children with CL&P. Although these results superficially look impressive, the numbers were very small and there may have been some bias in recruiting the infants to the study. Additionally, the questions asked of the parents were general and subjective, there was no control group, no recognition of parental or researcher bias or that infants and their parents often take some time to establish a feeding pattern and technique. For example, it may have been that the observed improvement occurred coincidentally at the time of plate fitting.

In all three of these studies however there was no control group and it is possible that improvement in growth was coincidental with plate fitting. Since infants were selected for plates at the time of poor growth there could be expected to be some effect of regression to the mean (Bland and Altman, 1994a; Bland and Altman, 1994b).

Choi et al (1991) evaluated infants’ ability to generate oral pressures during sucking when fitted with PSO. They evaluated 26 infants between the ages of 2 weeks and 8 months with CL and/or P and a control group of 7 non-cleft infants. Of the 26 infants with CL and/or P, 7 had UCLP, 8 ICP, 2 CL and 4 had repaired CL & P. They developed equipment that allowed them to measure the intra-oral pressures generated during milk feeds. Two teats were used, a NUK standard orthodontic teat and a NUK cleft palate teat. They found infants with CLP and CP were unable to generate any intra-oral pressures and there was no significant difference between pressure generation with PSO in situ versus without PSO in situ. The inability of infants with
unrepaired CL&/or P and ICP to produce any intra-oral pressures, with or without PSO in situ, suggests that PSO did not compensate for the structural defect during feeding. This study disproves the hypotheses that PSO restores infants’ ability to generate the intra-oral pressures required for sucking. It would be interesting to know whether the amount of feed taken by the infants increased with fitting of PSO. If so, it may suggest PSO facilitates compression of the teat and contributes to more efficient feeding. This study is the only reported use of objective assessment measures to evaluate the claim that PSO restore infants’ ability to generate negative intra-oral pressures during feeding. Unfortunately the age range of the infants introduces variables associated with developmental changes and the development of compensatory feeding behaviours. In addition, infant co-operation and tolerance of the assessment is not reported.

More recently, a multi-centre trial (Dutchcleft study) has been undertaken investigating the effects on PSO on several outcomes including feeding efficiency prior to surgical intervention (personal communication Konst, 2001; Prahl, 2001). Although these results have yet to be published it is reported that of the 49 infants recruited to the trial, 24 were randomised to receive PSO. All infants were bottle-fed with the Haberman bottle; a valved bottle which allows the infant to receive nutrition by simply compressing the teat without the need for generation of negative intra-oral pressures. Feeding efficiency was measured in terms of time taken to complete a feed. It was reported that there were no significant differences in the time taken to complete feeds whether infants were receiving PSO or not. Unfortunately details of the study methodology are not yet published.

There are also case reports of PSO being useful in facilitating both bottle and breast feeding (Lifton, 1956; Williams et al., 1968; Goldberg et al., 1988).

Williams et al (1968) reported a case study of an infant with Pierre Robin Sequence and additional complex medical problems. The infant had complex
feeding difficulties with episodes of apnoea and aspiration of vomit. While being able to take small amounts of feed orally, nasogastric supplementary feeds were required. After fitting the plate the infant could tolerate more feed orally but remained unable to take full oral intake, requiring a gastrostomy tube. The authors suggest that the increased amount of feed taken was related directly to the PSO but failed to acknowledge that it might also have been related to the maturation.

Some years later Goldberg et al (1988) reported a case demonstrating the effectiveness of a feeding plate on feeding efficiency in an infant with isolated cleft palate. The infant's medical history was unremarkable. He was fed initially by nasogastric tube but progressed on to oral feeds within 24 hours of birth. His feeding was assessed to be adequate and he was discharged from hospital at 6 days of age. Seven weeks later, however, he was readmitted with severe dehydration and malnutrition. His feeding regime was modified to include high calorie feeds and an alternative teat. After 5 weeks on the ward he had gained weight and was again discharged. However at 26 weeks of age he was readmitted with failure to thrive. During this admission a feeding plate was fabricated. The plate was well tolerated and the feeding improved with resultant weight gain. While the authors use this case study to demonstrate successful use of a feeding plate in a non-syndromic infant with ICP, they also suggest that reduced parental compliance with the feeding routine, while at home in an unsupported environment, may have contributed to the episodes of malnutrition and dehydration.

In summary, the adequacy of studies evaluating the use of PSO in feeding management is poor. The studies are often based on small numbers and lack, or have poorly matched control groups. They express subjective opinion rather than report objective measures, and lack evaluation of reliability of observations and potential for bias. They are prone to neglect aspects of the feeding process (e.g. physiological, psychological, behavioural), do not
consider issues related to growth in this population and fail to take account of
developmental issues such as change in feeding patterns with age.

In order to fully appreciate the effects of CL and/or P on feeding, and then
to evaluate the effect of PSO, a brief overview of normal feeding and the
changes in anatomy associated with CL and/or P is given. The following
sections summarize the relationship between normal anatomy and the process
of infant feeding, and suggest how feeding may be affected by the abnormal
anatomy of cleft lip and palate.

2.4 Normal Feeding

Infants' and children's feeding skills change over time with developmental
maturation. These changes are closely related to anatomical changes, the
disappearance of oral reflexes, sensori motor integration of swallowing and
respiration, normal posture and tone development, and psychosocial
maturation. By 3 years of age children's swallowing patterns closely resemble
those of adults. Much is known about swallowing in the adult population but
appreciably less of infant/paediatric feeding. There are areas of
infant/paediatric feeding where, due to ethical reasons, normative data
collection has not been possible. Extrapolations from the adult literature have
been made and are applied to infants and children (Arvedson, 1993). The
following discussion therefore includes a review of the adult and paediatric
literature with similarities, differences and extrapolations highlighted.

Infant feeding involves a highly co-ordinated sequence of oral, pharyngeal,
oesophageal and respiratory events. At the centre of the infant feeding
process is the suck, swallow and breathe triad. The ability to perform this
relies not only on an intact neurological system but also developmentally
appropriate anatomy. Furthermore, feeding may be affected by many
intrinsic, health and environmental factors, such as bonding, maternal skill or
experience with infant feeding. Other factors include the level of sedation or
Figure 6: Diagrammatic representation of differences between infant, older child and adult anatomy (Arvedson and Brodsky, 2002)
pain relief the mother required during delivery (Kron et al., 1966), the infant’s
behavioural state or level of arousal (Wolff, 1972), the taste (Crook and
Lipsitt, 1976), consistency and fat content of the feed (Nysenbaum and
Smart, 1982) and the feeding equipment used, for example the flow rate of
the nipple or teat (Mathew, 1988; Mathew and Bhatia, 1989; Mathew, 1990;
Mathew, 1991a; Mathew, 1991b; Mathew, 1991c; Mathew et al., 1992; Wolf
and Glass, 1992).

2.4.1 Anatomy

The infant’s oropharyngeal anatomy differs from that of an adult and relates
to the functional requirements of sucking and swallowing. These differences
predominate until the age of three or four months, when the anatomy
changes and becomes more “adult-like” (Figure 6).
The upper aero-digestive tract consists of the nasal cavity, mouth, pharynx, larynx and oesophagus. The nose, mouth, pharynx, airway and oesophagus provide a conduit for the passage of both air and food. At the proximal end of this conduit are two distinct channels, the nasal and oral cavities. These cavities meet at the level of the pharynx where there is a combined channel. At the level of the larynx this divides into the oesophagus and the trachea (shown in Figure 7). In terms of swallow efficiency this is a crucial area. While it is perfectly safe for air to enter the oesophagus there can be significant consequences if food/fluid enters the trachea. Food/fluid entering the trachea may result in chest infections and aspiration pneumonia leading in extreme cases to death.

Figure 7: Simplified diagram demonstrating the principles of the pharynx acting as a "combined channel" (Wolf and Glass, 1992)
• The nasal cavity

The nasal cavity is the primary passage for air. It is bordered anteriorly by the cartilaginous portions of the nose, inferiorly by the palatine process of the maxilla and the palatine bone (forming a rigid boundary between the nasal cavity and the oral cavity) and superiorly by the nasal, frontal and ethmoid bones. Internally it is divided vertically by the nasal septum, ethmoid bone and vomer. The nasal choanae run horizontally through each section and direct airflow through the nasal cavity. Posteriorly, the nasal cavity narrows, forming the choanae and marking the junction with the pharynx. Tissue extending beyond the hard palate forms the soft palate, which elevates during swallowing completing the separation of the nasal and oral cavities (Bosma, 1972; Sommerlad, 2000). The structure of the infant nasal cavity is essentially the same as that of the adult, however a major difference is that infants are obligate nasal breathers (Figure 6). The infant's ability to co-ordinate sucking, swallowing and respiration relies on this nasal breathing.

There are significant anatomical differences with cleft lip and palate. Whereas in non-cleft infants the nose is supported in the midline by the nasal septum, in infants with complete clefts of the lip and palate, the nasal septum is deviated away from the midline as shown in Figure 8. In addition the lower cartilaginous septum and vomer are angled upwards from the vertical septum. In CLP, the nasal bones are broad. Nasal obstruction affecting the infant's ability to breathe through the nose is not uncommonly seen in infants with CLP (Sommerlad, 2000).
Deviated nasal septum

Figure 8: Diagram showing the distortion of the alar cartilages and nasal septum in cleft lip (Sommerlad, 2000)

- The lips

A circular ring of muscle, the obicularis oris, forms the lips. They provide the seal with the breast or bottle, thus allowing generation of intra-oral pressures required during sucking (discussed further in Section 2.4.2). The lips also prevent anterior loss of fluids and solids during eating and drinking.

In contrast to the lip musculature in non-cleft infants, the obicularis oris of infants with cleft lip is interrupted and the muscle fibres turn upwards at the cleft (Figure 9). Other muscle fibres extend up towards the anterior nasal spine medially and the alar base laterally. Where there is an incomplete cleft of the lip there may be varying degrees of muscle continuity. Lip seal and containment of liquids and solids within the oral cavity may be adversely affected.
Figure 9: A schematic drawing of the lip musculature in unilateral cleft lip as compared to normal (Sommerlad, 2000)

- The mouth/oral cavity

The boundaries of the oral cavity are the palatine processes of the maxilla and the palatine bone, which merge into the soft palate and uvula (forming the roof of the mouth), the bony mandible, the mylohyoid and geniohyoid muscles, the anterior belly of the diagastric muscle (forming the floor of the mouth), the buccinator and masseter muscles (defining the cheeks) and the obicularis oris muscles (forming the lips). The tongue has attachments to the mandible, hyoid bone and styloid process of the cranium by the genioglossus, hypoglossus and styloglossus muscles.

There are several differences between the adult and infant oral cavity. The infant tongue fills the infant's oral cavity and is almost entirely positioned in the mouth, contacting the floor and roof of the mouth simultaneously (Figure 6). The mandible is smaller and is retracted in relation to that of the adult (Figure 6). Additionally the infant has deposits of fatty tissue in the cheeks (sucking pads), which help stabilise the cheeks. They are also reported to
have an inner ridge of specialised mucosa, known as the pars villosa which is said to contribute to the lip seal around the nipple (Bosma, 1972; Donner et al., 1985).

Clefts of the lip and palate disrupt the hard palate to varying degrees. In contrast to non-cleft anatomy, where the nasal septum is joined to the hard palate along its length, in UCLP the vomer is joined to the palatal shelf on the non-cleft side. (Sommerlad, 2000).

Several authors (Veau, 1931; Kriens, 1969; Fara and Dvorak, 1970) have described the abnormal velar musculature in cleft palate. The muscles of the palate, including the levator, palatopharyngeus and palatoglossus are unable to meet in the midline, instead inserting into the margins of the cleft, the oral and nasal mucosa, the abnormal palatal aponeurosis, and the back of the hard palate (Figure 10) (Sommerlad, 2000).

Figure 10: Diagram of the cleft muscular anatomy (Sommerlad, 2000)
The palate has several functions during feeding. The hard palate provides a rigid opposing surface allowing the tongue to compress the nipple or teat during breast or bottle-feeding. The soft palate elevates during swallowing separating the oral and nasal cavities and preventing nasal regurgitation. In addition soft palate elevation and resultant velopharyngeal closure contributes to the “closed system pressure generation” required for efficient sucking and swallowing. Where the anatomy and function are affected, as with cleft palate, nasal regurgitation may be evident, sucking less efficient and the closed system required for swallowing compromised (Wolf and Glass, 1992).

- The pharynx

The pharynx can be considered as three different anatomic areas; the nasopharynx, the oropharynx and the hypopharynx.

The nasopharynx is the area of the pharynx extending from the nasal choanae to the soft palate. The eustachian tubes originate in the nasopharynx. The faucial arches act as a bridge between the mouth and the oropharynx. The oropharynx is defined anteriorly by the junction and the base of the tongue (Figure 6). It consists of the area between the elevated soft palate and the epiglottis and includes the valleculae. The hypopharynx extends from the base of the epiglottis to the cricopharyngeal sphincter. It incorporates the laryngeal inlet, pyriform sinuses and the cricoid cartilage.

The shape of the pharynx changes significantly from infancy, through childhood and into adulthood. In infancy the pharyngeal wall slopes gently from the nasopharynx to the hypopharynx. However as the child matures and moves to a more adult diet, the angle between the nasopharynx and oropharynx gradually increases until it reaches near 90 degrees in adulthood (Figure 6).
While the oropharynx and nasopharynx of infants with cleft palate will be affected by the abnormal palatal anatomy as discussed in the previous section, the hypopharynx is reported to be unaffected.

- **The larynx**

The larynx is a cartilaginous structure suspended by muscular ligamentous attachments to the hyoid bone and cervical vertebrae. The infant larynx is superiorly and anteriorly positioned making it closer to the base of the tongue than in the adult (Figure 6). The larynx provides the airway protective mechanisms during swallowing thus preventing aspiration. The primary protective mechanism is the epiglottis, which folds downwards, sealing against the aryepiglottic folds and closing the laryngeal inlet. In addition the false vocal folds and the true vocal folds adduct during swallowing adding another level of airway protection (Figure 11).

The larynx is considered to be unaffected anatomically in non-syndromic cleft lip and palate (Figure 12).
Figure 11: Diagrammatic representation of the levels of airway protection (Arvedson and Bradsay, 2002)

Figure 12: Diagrammatic representation of larynx in relation to the tongue and soft palate
The oesophagus

The oesophagus is a musculomembranous tube that extends from the pharynx at the level of the cricoid cartilage to the stomach at the level of the eleventh vertebral body. The pharyngo-oesophageal segment (upper oesophageal sphincter) forms the junction between the pharynx and the oesophagus. The oesophagus is made up of three anatomic segments; the cervical, thoracic and abdominal.

The oesophagus is considered to be unaffected anatomically in non-syndromic cleft lip and palate.

2.4.2 Stages of swallowing

The feeding-swallowing process is usually considered in 4 stages; the oral preparatory, the oral, the pharyngeal and the oesophageal stages. These stages are interdependent and highly co-ordinated. The oral preparatory and oral stages are largely under voluntary control. The pharyngeal stage is though to be under both voluntary and involuntary control and the oesophageal stage under involuntary control (Logemann, 1983; Groher, 1984).

The following diagram (Figure 13) taken from Groher (1984) summarises these main stages and highlights their cross-over and interdependence. For the purposes of this diagram the oral preparatory and oral stages have been combined as one group. Figure 14 show the sequential nature of the oral and pharyngeal stages.
Figure 13: Diagrammatic representation of interdependence of the oral, pharyngeal and oesophageal stages of swallow (Groher, 1984)
Figure 14: Diagrammatic representation of the oral and pharyngeal stage sequence (Grober, 1984)
The oral preparatory stage/bolus formation

Adults

Prior to the commencement of the oral preparatory stage, there must be sensory recognition of food approaching and being placed in the mouth. Once there, the lips seal to prevent loss of food or liquid. During this stage the palate is pulled down and forward, sealing the oral cavity from the pharynx. Nasal breathing continues (Shedd et al., 1960; Fletcher, 1974; Storey, 1976; Wildman, 1976; Logemann, 1983).

The patterns of manipulation and control of the bolus used are dependent on it's texture and volume (Dantas and Dodds, 1990; Dantas et al., 1990; Kahrilas and Logemann, 1993; Kahrilas et al., 1996). Dodds et al (1989) investigated oral preparatory patterns in normals and reported two distinct positions in which the bolus can be held. They described these patterns as "tippers", where the bolus is held between the midline of the tongue and the hard palate with the tongue tip elevated and contracting the alveolar ridge, and "dippers", where the bolus is held on the floor of the mouth in front of the tongue.

Liquid boli initially are held between the tongue and the anterior hard palate. This is generally achieved as the tongue cups around the bolus, with the sides of the tongue laterally sealed against the alveolar ridge. Some individuals may move the bolus around the mouth prior to triggering the pharyngeal stage resulting in the bolus being spread throughout the oral cavity. However prior to triggering of pharyngeal stage the liquid is gathered, reformed into a bolus and held in either the tipper or dipper position (Logemann, 1983; Dodds et al., 1989).

A similar pattern is observed when taking thicker textures. As the viscosity of food increases the bolus size tends to decrease. Depending on the texture of foods, the boli may simply be positioned in the tipper or dipper position in
preparation for swallowing or may be manipulated, moved laterally, masticated, reformed into a bolus and repositioned in the tipper or dipper position (Dodds et al., 1989).

The process of mastication begins with the transfer of food onto the teeth. The tongue and mandible then move in a rotary fashion and the upper and lower teeth meet, crushing the food. The masticated food then falls on the tongue and is moved back on to the teeth as the mandible opens. As this process is repeated the food is mixed with saliva (Lowe, 1981).

**Infants and children**

As with adults, during this stage, the soft palate is lowered, preventing the bolus from entering the pharynx prior to initiation of a swallow. The airway is open and nasal breathing continues (Arvedson, 1998).

During infancy it is difficult to differentiate the oral preparatory stage from the oral stage. However the primitive reflexes which occur in preparation for nutritive sucking, such as rooting and latching onto a nipple or teat, are generally considered preparatory (Arvedson, 1998). This pattern changes as the infant matures. Primitive reflexes diminish and the infant moves from sucking to spoon-feeding at approximately 4 to 6 months of age. As children develop and move on to more solid textures the oral preparatory stage resembles more closely the adult patterns described above. As this stage of the swallowing process is largely voluntary, behavioural responses become more significant (discussed further in Section 2.5).

**The oral stage**

*Adults*

The oral stage commences as the tongue begins to move posteriorly. If the more common "tipper" position is used to hold the bolus the tongue tip moves forward smoothly and lifts the bolus on to the tongue (or "dipper"
position). The middle of the tongue moves in a stripping or rolling action moving sequentially from the hard or anterior palate towards the soft or posterior palate, thus transferring the bolus towards the pharynx (Shawker et al., 1984; Kahrilas and Logemann, 1993). During this time the sides and tip of the tongue remain in contact with the alveolar ridge and a central groove forms contributing to posterior bolus transfer (Shedd et al., 1960). It has also been suggested that generation of negative pressure within the oral cavity, created by slight inward movement and increased tension of the buccal musculature may contribute to bolus transfer (Shedd et al., 1961). The oral stage is usually completed in less than 1.5 seconds, but increases slightly with viscosity (Dantas and Dodds, 1990; Dantas et al., 1990; Reimers-Neils et al., 1994).

**Infants and children**

The oral stage remains reflexive for infants until an age of approximately 3 months. After this time it comes under greater voluntary control. In children over the age of 2 years the oral stage resembles that of an adult pattern.

In infants the oral stage lasts approximately 0.5 seconds. This may however be affected by bolus volume.

Breast and bottle fed infants initiate the oral stage as they begin anterior posterior tongue movements on the teat or nipple (Arvedson and Brodsky, 2002).

The key event in the oral stage for infants is the suckling/sucking process. There are 2 distinct developmental phases in infant sucking. The first, known as suckling, develops in utero and persists until an approximate developmental age of 6 months. Suckling is characterised by backward and forward movements of the tongue. Milk is transferred from a nipple or teat by a rhythmic licking action of the tongue combined with pronounced opening and closing of the jaw (Flowers and Morris, 1973; Bosma, 1986;
Evans Morris and Klein, 1987). During this time the lips are loosely approximated. In contrast, a sucking pattern develops at an approximate developmental age of 6 months following a period of facial growth, which permits increased oral cavity size and therefore more room for tongue movements. Sucking is characterised by upward and downward movements of the body of the tongue and firm lip approximation (Evans Morris and Klein, 1987). The main difference between these 2 phases is therefore the direction of tongue movements used to transfer milk from the breast or teat. Between six and nine months of age a mixture of suck and suckle patterns is seen (Arvedson and Brodsky, 2002).

In spite of the distinction between suckling and sucking the term “sucking” has become generic and will be used in this thesis to refer to “the rhythmic movements of the infant’s mouth and tongue on either the bottle or breast to obtain nourishment or on a pacifier, hand or other object to modulate state or explore the environment” (Wolf and Glass, 1992).

**Biomechanics of sucking**

It is thought there are two main pressures required for efficient nutritive sucking: positive and negative pressure generation (Logan and Bosma, 1967). Positive pressure generation or compression “pushes” fluid out of the teat or nipple and negative pressure or suction “draws” fluid out. Positive pressure is generated as the tongue compresses the nipple or teat against the opposing surface or palate and liquid is expelled into the mouth (Ardran et al., 1958a; Ardran et al., 1958b) as seen in Figure 13. Negative pressure can only be generated when the oral cavity is sealed by the lips anteriorly and the soft palate and tongue posteriorly. With the oral cavity sealed the jaw and tongue drop enlarging the oral cavity. This has the effect of creating negative intra-oral pressure and producing suction, which draws fluid into the mouth (Colley and Creamer, 1958; Wolf and Glass, 1992) as seen in Figure 15.
Motoric components of sucking

Several specific oral movements are required during sucking. The tongue body forms a central groove or cups around the nipple or teat enabling positioning and stabilisation of the teat. Anteriorly the tongue is involved in sealing the oral cavity against the nipple or teat. Posteriorly it is sealed against the soft palate, until it is lifted during the swallow. Once the teat or nipple is in the mouth the anterior tongue elevates and compresses it against the opposing surface of the hard palate (positive pressure generation). Almost simultaneously, the posterior tongue lowers and enlarges the oral cavity contributing to negative pressure generation. The jaws, cheeks and the fat pads provide a stable bases for the tongue, lips and cheeks during sucking. The lips are involved in facilitating the anterior seal around the nipple/teat. The soft palate elevates sealing the nasal cavity, preventing nasal regurgitation.
and contributing to the sealed oral cavity (Bosma, 1972; Wolf and Glass, 1992; Glass and Wolf, 1999; Arvedson and Brodsky, 2002)

Rhythm and co-ordination of sucking

When all the oral structures work together in a co-ordinated way they produce the smooth, rhythmic movements that characterise sucking. There are two different types of sucking; nutritive and non-nutritive. Non-nutritive sucking occurs in the absence of nourishment and occurs typically with a pacifier/dummy (Bosma, 1972; Wolf and Glass, 1992; Glass and Wolf, 1999; Arvedson and Brodsky, 2002). It provides comfort, and opportunity for exploration and state regulation (Bosma, 1972; Glass and Wolf, 1999; Arvedson and Brodsky, 2002). Nutritive sucking on the other hand occurs to obtain nourishment. It is characterised by slower and longer sucking bursts, compared with the faster and shorter bursts of non-nutritive sucking (Figure 16). There are several differences between the patterns of nutritive and non-nutritive sucking. These differences will be described in detail below. Two hypotheses have been proposed to account for the differences between nutritive and non-nutritive sucking. Lipsitt et al (1976) suggest the slower rate of nutritive sucking, reflects the infant savouring the fluid whereas other authors suggest this might be due to the swallowing activity associated with nutrition (Dubignon and Campbell, 1969; Wolff, 1972; Burke, 1977).
Nutritive sucking (NS)

Sucking patterns are characterised by bursts of continuous sucking and pauses (Figure 16). These have been evaluated extensively in terms of gross and fine structures of sucking (Lau and Hurst, 1999). The gross structures of sucking include the number and duration of sucking bursts and pauses. More detailed analysis of "within burst" or fine structures includes number of sucks per burst, the rate of sucking, the intervals of the sucks and the amplitude or strength of the sucking. Many of these measures change with maturity (Pollitt et al., 1981; McGowan et al., 1991) and the emergence of a regulatory mechanism that modulates the pattern of intake (Pollitt et al., 1981) as described below.

In nutritive sucking the length of sucking bursts varies within feeds and with age. Sucking bursts at the beginning of a feed are reported to last between 30 and 80 seconds and as the feed progresses the duration of these bursts decreases and becomes erratic. Similarly the pauses following the sucking bursts become inconsistent in length (Qureshi et al., 2002).
The number of sucks within each burst and the rate of sucking is reported to vary with age (Wollf, 1968; Qureshi et al., 2002). Qureshi et al (2002) report the average number of sucks within a burst for neonates to be 10.14 (SD 8.61) increasing to 20.99 (SD 22.65) at one month of age.

The rate of sucking is measured as sucks/second or sucks/minute. Reports are consistent with a rate of approximately 1 (range 0.8 to 1.2) suck/second reported within the first few days of life (Wollf, 1968; Burke, 1977; Anderson et al., 1982; Jain et al., 1987; Bu'Lock et al., 1990; McGowan et al., 1991). As the infant matures, sucking rates increase slightly to approximately 1.3 sucks per second, with the lowest reported being 1.1 and the highest 1.5 at approximately 2 months of age (McGowan et al., 1991; Qureshi et al., 2002). The sucking rate continues to increase slightly and is reported to be as high as 1.8 sucks/second at 9 months of age (McGowan et al., 1991; Qureshi et al., 2002).

The number of sucks utilised before triggering a swallow is referred to as the suck:swallow ratio (S:S). This measure varies with age and other variables such as feeding method (see 2.5). In the past it has been widely accepted that infants use a suck:swallow ratio of 1:1 (Wolf and Glass, 1992; Glass and Wolf, 1999; Arvedson and Brodsky, 2002). However recent studies suggest this may not be the case and that while suck:swallow ratios of 1:1 are evident in the majority of new-born infants at birth (78.8%, SD 20.1), this is not so at one month of age when only 57.5% of infants use the 1:1 ratio, alternatively using ratios of 2:1 or 3:1 (McGowan et al., 1991; Qureshi et al., 2002). This highlights the significance of developmental changes over the neonatal period and the rapidity of these changes.

The volumes of feed taken have been measured in terms of total amount taken during a feed (Qureshi et al., 2002), ml taken per minute (Pollitt et al., 1981; McGowan et al., 1991) and ml taken per suck (McGowan et al., 1991). While the volume of feed per suck decreased with age (0.40 ml at 2 months
of age and 0.26 ml at 10 months of age) this paralleled the increased rate of sucking previously reported (McGowan et al., 1991). Pollitt et al (1981) obtained similar results in terms of the volume of feed consumed per minute. Qureshi et al (2002) evaluated the volume of feed taken based on the amount of feed divided by the total number of sucks used. They reported an increase of 75% during the first month of life paralleling the finding that the rate of sucking increases with age.

Pressures generated during sucking are highly variable. Pressures of —15 to —130 mm Hg have been reported (Kron et al., 1963; Sameroff, 1968; Ellison et al., 1979; Dubignon and Cooper, 1980; Pollitt et al., 1981; Mathew and Bhatia, 1989). Unfortunately the techniques used to obtain these measurements have varied, and have been poorly reported and confounded by the many variables that can affect infant sucking. Nevertheless pressure generation is reported to be the most consistent measure for individual infants across different feeds (Kron et al., 1968). Pressure generation (mean peak intra-oral pressure) has been reported to be unaffected by age (Pollitt et al., 1981; McGowan et al., 1991).

Non-nutritive sucking (NNS)

In contrast non-nutritive sucking is characterised by short sucking bursts, with an average of 7.76 (range 4.3-12.2) sucks per burst followed by a rest period of approximately 6.61 (range 3.0-10.6) seconds. The rate of non-nutritive sucking is approximately twice as fast as that of nutritive sucking. It varies considerably with age, from 2.1 sucks per second (range 1.9-2.3) at 4-6 days of age increasing slightly to 2.5 (range 2.0-2.6) at 14 to 60 days of age and 2.7 (range 2.4-2.7) at 7-9 months of age (Wolff, 1968).
The pharyngeal stage

**Adults**

The pharyngeal stage begins with triggering of the swallow. It is suggested that this is dependent on sensory feedback from a number of areas including the faucial arches, uvula, soft palate, posterior tongue, pharynx, epiglottis and larynx (Hollshwander et al., 1975; Storey, 1976; Miller, 1986). There is some difference in where the swallow is triggered in normal adults possibly depending on age. In younger adults the swallow has been found to be triggered when the bolus reaches the anterior faucial arches (Logemann, 1983; Fujiu et al., 1994), base of tongue, and valleculae (Derkay and Schechter, 1998). Adults over the age of 60 years have been found to trigger the swallow slightly later as the bolus head reaches the middle of the tongue base (Tracy et al., 1989; Robbins et al., 1992).

On triggering of the swallow a series of closely linked events occurs within 1 second (Logemann, 1983; McGrath and Anderson, 1991; Logemann et al., 1998; Robbins et al., 1999).

Velopharyngeal closure takes place as the result of elevation and retraction of the soft palate and inward movement of the posterior and/or lateral pharyngeal walls. Velopharyngeal closure contributes to the closed system required for pharyngeal clearance (discussed below).

During the swallow the larynx and hyoid bone elevate, contributing to glottic closure and airway protection. Anterior movement of the larynx and hyoid contribute to relaxation of the cricopharyngeus or upper oesophageal sphincter. In the adult male the expected laryngeal elevation is 2 cm (Jacob et al., 1989; Tracy et al., 1989). Data for females are not available.

Glottic closure is achieved at several levels (Figure10). The vocal folds adduct and the laryngeal vestibule compresses, clearing any contents and any laryngeal penetration (Logemann, 1983). Simultaneously the arytenoid
cartilages move downwards, forwards and inwards, narrowing the laryngeal opening (Ardran and Kemp, 1967) and the larynx is elevated and pulled forward assisting closure of the laryngeal vestibule (Ohmae et al., 1995). Complete vocal cord closure is achieved when the larynx is elevated to about 50% of its maximum elevation. The duration of this airway closure is 1/3 to 2/3 second (Logemann et al., 1998) for a single bolus. However in continuous drinking airway closure may be maintained for 5 seconds or more (Martin et al., 1994).

There has been debate about what constitutes normal airway protection and to what extent healthy individuals have incomplete airway protection and aspiration without complications. To address this problem Robbins et al (1999) evaluated airway protection in 98 normal adults of different ages and genders using videofluoroscopy and the laryngeal penetration/aspiration scale (Rosenbek et al., 1996). It was found that high laryngeal penetration (epiglottic undercoating but no contact with the vocal folds) was present in all age groups, however aspiration was only evident in one elderly male. This episode of aspiration was spontaneously cleared from the trachea. The results of this paper suggest aspiration in healthy young adults is not normal.

Several events contribute to pharyngeal clearance. The first occurs as the tail of the bolus reaches the base of tongue. The tongue base retracts and the lateral and posterior pharyngeal walls contract (Kahrilas et al., 1992). Following the base of tongue and pharyngeal wall contact, pharyngeal wall contraction moves progressively through the pharynx applying pressure to the tail of the bolus. As the bolus moves through the pharynx it divides at about the level of the valleculae with approximately 50% of the bolus moving down each side of the pharynx. The two portions of the bolus then meet again at the cricopharyngeus (Ardran and Kemp, 1951).

In addition it has been proposed that bolus transfer through the pharynx is partly achieved by forces generated within a closed system created by closure
at the lips, the velopharyngeal port and the cricopharyngeus or upper oesophageal segment. These forces are described as being generated by the tongue base and pharyngeal wall contraction. As the tongue forces the bolus into the pharynx, increasing pressures, the cricopharyngeus relaxes, and the bolus is in effect “sucked” into the upper oesophagus, moving from an area of high pressure in the pharynx to lower pressure in the oesophagus (McConnel et al., 1983).

At the conclusion of the pharyngeal stage it is reported there is normally very little residual food in the pharynx (Logemann, 1983; Groher, 1984).

Infants and children

Due to the ethical and safety issues of irradiating infants and children, very little normative data about the pharyngeal stage of swallowing in paediatrics exists (Lefton-Greif and Arvedson, 1997), in contrast to adults.

Arvedson and Lefton Greif (1998, 2002) maintain that the pharyngeal stage in infants and children closely resembles that of adults but there are differences in terms of airway protection related to infant anatomy. The hyoid bone and larynx are much higher in infants and young children, thus providing more natural airway protection (Bosma, 1986; Newman et al., 1991) and laryngeal elevation is reduced. It is also suggested that during infancy the posterior pharyngeal wall may move further anteriorly than in adulthood (Logemann et al., 1998). Newman et al (1991) also found that the duration of the pharyngeal stage is similar to that reported for adults, 0.60 sec (range 0.46-0.89).

The oesophageal stage

Adults, infants and children

The oesophageal stage is initiated as the pharyngo-oesophageal (PE) segment relaxes and the bolus enters the upper oesophagus. The amount and duration of PE segment relaxation in adults increases with greater bolus size. Once in
the oesophagus peristaltic movements transfer the bolus to the stomach. The process of transferring the bolus through to the lower oesophageal segment takes approximately 7-10 seconds in adults (Ingelfinger, 1958; Dodds et al., 1981; Clark, 1993). Lower oesophageal sphincter relaxation occurs within 2 seconds of swallowing and lasts 8-10 seconds allowing the peristaltic wave and bolus to pass through. The oesophageal stage occurs following every discrete pharyngeal swallow. If 2 or more swallows occur within quick succession, such as in continuous drinking or in bottle-feeding, there may be inhibition of the oesophageal stage until the continuous burst of drinking or sucking is complete. The final swallow in the series will be then be followed by one peristaltic wave that clears the oesophagus (Dodds et al., 1973; Arvedson and Lefton-Greif, 1998).

2.5 Factors which affect the feeding process

A multitude of factors can affect the swallowing/feeding process. These can be broadly categorised into intrinsic factors of the bolus (such as bolus size, viscosity and temperature), the method of taking the bolus and equipment used, medical factors, and mechanical factors such as the presence of nasogastric feeding tubes. When considering infant feeding there are also environmental factors such as the sex and birth order, pre- and perinatal history, social issues and behavioural aspects to be considered.

2.5.1 Intrinsic factors of the bolus

Bolus volume is well described as affecting the oro-pharyngeal swallow in adults (Kahrilas et al., 1993; Kahrilas and Logemann, 1993; Shaker et al., 1993; Kahrilas et al., 1996). It is reported that small boli (1-3mL) result in sequential initiation of the oral and then pharyngeal stages, whereas larger boli (10-20mL) trigger simultaneous oral and pharyngeal activity in order to clear the large bolus safely (Kahrilas et al., 1993; Kahrilas and Logemann, 1993; Shaker et al., 1993; Kahrilas et al., 1996). Overall, oral and pharyngeal transit times remain unchanged (Dantas and Dodds, 1990). While changes in the
timing of individual oral and pharyngeal stage events are changed with bolus volume, the range of movements and duration remain unaltered. However within the oral stage the onset of individual events such as velopharyngeal elevation occur sooner relative to bolus transfer but the range of velopharyngeal elevation does not change.

Viscosity of fluids and foods on swallowing is reported to contribute predominantly to pharyngeal clearance. More viscous bolus lead to increased generation of pressures by the tongue, tongue base and pharyngeal walls, all of which contribute to pharyngeal clearance (Dantas and Dodds, 1990; Dantas et al., 1990; Reimers-Neils et al., 1994). Similarly, the duration of valving functions, such as velopharyngeal closure, cricopharyngeal relaxation and laryngeal closure, is increased with increased viscosity (Kendall et al., 2000).

There is controversy about whether increasing sucrose concentration affects the number of sucks generated. Crook and Lipsitt (1976) reported no significant difference in the number of sucks generated with increased sucrose concentration but Burke et al (1977) reported an increase in the number of sucks. There is however agreement that the rate of sucking decreases inversely to sucrose concentrations. Crook and Lipsitt (1976) also reported that the length and number of sucking bursts increases with increasing sucrose concentration.

Similarly the fat content of milk has been shown to affect sucking patterns. When the fat content of feed is increased, the overall sucking and resting time during feeds remains unaltered but the rhythm is altered with longer sucking bursts and post-sucking burst pauses (Woolridge et al., 1980).

### 2.5.2 Feeding method/equipment

Adult studies have shown that the method of fluid presentation affects the subsequent pattern of swallowing. When adults use continuous cup drinking
the pharyngeal stage begins early, as the cup approaches the lips, and is characterised by early laryngeal elevation and airway closure. As drinking continues the lips remain sealed, the velopharyngeal port remains closed and the tongue repeatedly propels consecutive swallows from the oral cavity. The tongue base and posterior pharyngeal wall make contact with the tail of each bolus and the cricopharyngeal sphincter is relaxed for each bolus. Throughout this time the airway remains closed (Martin et al., 1994). An alternative pattern of continuous drinking known as “chug a lug” is also described (Logemann, 1983). This differs from sequential drinking in that the larynx is pulled forward, opening the cricopharyngeal sphincter, the breath is held and the bolus “dumped” with the assistance of gravity through the oral cavity, pharynx and oesophagus (Logemann, 1983).

In infant bottle feeding, the type of teat used has been shown to affect sucking patterns. When large hole teats are used and there is therefore a faster flow rate of fluid, fewer sucks are used, the rate of sucking decreases and the amount of suction decreases (Christensen et al., 1976; Eishima, 1991; Mathew, 1991a). Conversely, when teats contain small holes and the flow rate is therefore slowed, infants use rapid and vigorous tongue and jaw movements (Ardran et al., 1958a; Mathew, 1991a). It is also suggested infants use different movement patterns when fed with large or small hole teats. When a small hole is used the tongue surrounds the teat completely. In contrast, when a large hole is used, the tongue flattens and does not surround the teat at all (Eishima, 1991). When teats with a small hole are used, vigorous tongue and jaw movements are seen (Eishima, 1991). It is suggested that “if a nipple with a large hole is given repeatedly then the movements of the tongue, lips, jaws and cheeks and throat would become lazy” (Ardran et al., 1958a; Eishima, 1991).

2.5.3 Mechanical factors

The presence of nasogastric tubes has been associated with altered patterns of oro-pharyngeal swallowing, among which are delayed triggering of the
swallow, delayed pharyngeal transit times and increased duration of cricopharyngeal relaxation (Huggins et al, 1999). Additional complications which may also result in altered oro-pharyngeal swallowing include gastro-oesophageal reflux (Curtis and Langmore, 1997) and tube displacement (Bastow, 1986; Finucaine et al., 1991).

2.5.4 Demographic and social factors

It has been suggested that feeding behaviour of male and female infants may differ with regards to responsiveness to oral and cutaneous stimuli (Korner, 1973), taste (Dubignon et al., 1969) and tongue movements during feeding (Balint, 1948). However this has not been found consistently (Hendry and Kessen, 1964; Korner et al., 1968; Dubignon et al., 1969; Nisbett and Gurwitz, 1970).

Birth order is reported to be a factor in infant feeding efficiency. First time mothers are reported to spend significantly longer feeding their infants (Thoman et al., 1970) and to use more feeding and non-feeding intervals for resting and winding (Thomas et al., 1971). Further, their infants take less formula (Thoman et al., 1970).

2.5.5 Prenatal and perinatal history

Although prenatal factors such as alcohol and nicotine exposure have been found to affect infant feeding patterns (Stock et al., 1985) perinatal factors are more significant. The type of delivery has been shown to affect sucking efficiency, with those requiring assisted delivery such as forceps, producing lower sucking times and number of sucks (Dubignon et al., 1969). Maternal sedation has been shown to affect infant feeding patterns up to 4 days of age (Bosma, 1990). Infants of mothers who had not received barbiturates during the delivery of their infant use more continuous sucking bursts and stable sucking rates (Kron et al., 1968). In contrast, infants of mothers who received barbiturates demonstrate altered sucking patterns, with slower sucking rates and pressure generation and consume less nutrients during the feeding
session (Kron et al., 1966; Dubignon et al., 1969). Babies who are heavier at birth and/or more mature have been found to have a higher daily intake of feed in volume and calories (Dubignon et al., 1969).

2.5.6 Psychological factors

The impact of psychological factors on infants’ feeding has been investigated extensively in infants and children with non-organic failure to thrive (Gordon and Jameson, 1979; Kotelchuck, 1980; Call, 1984; Crittenden, 1987; Brinich et al., 1989; Ward et al., 1993; Chatoor et al., 1998). The relationships between these factors and organic feeding disorders are often difficult to separate. Non-organic failure to thrive has been associated with maternal deprivation, parental neglect and physical abuse (Kotelchuck, 1980; Call, 1984). Between 49% and 92% of infants with non-organic failure to thrive demonstrate insecure attachment patterns (Gordon and Jameson, 1979; Crittenden, 1987; Brinich et al., 1989; Ward et al., 1993). Chatoor (1998) suggests environments of maternal deprivation and inconsistent caregiving lead to lack of interest in food and physiological dysregulation.

2.5.7 Infant factors

Other factors which have been shown to affect feeding include the infant’s behavioural state, such as wakefulness or distress (Levin and Kaye, 1964) and when the infant is next due a feed (Levin and Kaye, 1964; Kron et al., 1968). Factors such as the infant’s hunger and time of feed do not appear to affect the characteristics of sucking, only the amount of feed taken (Kron et al., 1968; Rybski et al., 1984).

2.5.8 Behavioural aspects

Behavioural feeding disturbances are common in the paediatric population and are reported to occur in between 25 to 40% of healthy toddlers and early school age children (Forsyth et al., 1985; Marchi and Cohen, 1990; Mayes and Volkmar, 1993). The more common problems are: inappropriate mealtime
behaviours (e.g. throwing food), food selectivity or only eating a limited range of foods, failure to move from puree foods to family foods and food refusal. These problems are generally transitory and resolve spontaneously or with appropriate behaviour management (Kerwin, 1999). More serious behavioural feeding problems, such as gagging, vomiting, and persistent refusal of feeds, occur in 3 to 10% of the paediatric population but are more common in children with physical or intellectual disabilities, complex medical problems or prematurity (Arvedson and Brodsky, 2002).
The highly complex nature of infant feeding has resulted in the continuing development of many different assessment methods. However, no single technique comprehensively assesses all aspects of the feeding process. This chapter reviews feeding assessment techniques as a background to the selection of procedures used in this study.

The approaches to assessing feeding can be categorised as:

1. Feeding history
2. The clinical feeding evaluation
3. Diagnostic tests and procedures

3.1 Feeding History

Information relating to feeding informs assessment, prognosis and management. This is collected informally during interview with the parents/caregivers and/or on review of the child’s medical notes. The following should be included (Evans Morris and Klein, 1987; Wolf and Glass, 1992; Arvedson, 1993; Arvedson and Brodsky, 2002).

1. Medical history and medications
2. Social factors

3. Parental description of the infant's feeding

Several questionnaires have been developed to guide this process, however none are universally accepted (Wolf and Glass, 1992; Arvedson and Brodsky, 2002).

3.2 Clinical feeding evaluation

The clinical feeding evaluation includes an examination of the infant or child, and observation of his or her feeding. This includes assessment of behavioural state, neuro-motor control, oral motor and reflexes examination, and the infant's or child's behavioural responses to tactile input (Wolf and Glass, 1992; Arvedson and Brodsky, 2002).

An advantage of the clinical evaluation is that it allows the clinician to replicate the child's feeding routine as much as possible. However one major disadvantage is that the pharyngeal stage of the swallow cannot be accurately assessed during clinical assessment. Adult patients' reports of the severity of their own dysphagia has been shown to be unreliable (McCullough et al., 2000). Several studies have investigated whether experienced dysphagia clinicians are able accurately to detect aspiration in adults using clinical assessment techniques (Linden and Siebens, 1983; Splaingard et al., 1988; Linden et al., 1993; Arvedson et al., 1994; Murray et al., 1996; Daniels et al., 1997; Logemann et al., 1999; Warms and Richards, 2000). They have revealed very poor reliability for detecting aspiration with as few as 30% of episodes of aspiration being detected clinically (Linden and Siebens, 1983; Splaingard et al., 1988; Nathadwarawala et al., 1992; DePippo et al., 1992; Linden et al., 1993; Arvedson et al., 1994; Murray et al., 1996; Daniels et al., 1997; Logemann et al., 1999; Warms and Richards, 2000). There are no such studies for the paediatric population. Given the similarities between the pharyngeal stage of the swallow in adults and infants Arvedson and Lefton
Grief (1993; 2002) argue these findings can be extrapolated to the paediatric population. So while the clinical evaluation provides information on some aspects of infant feeding it relies on descriptions of behaviours and is likely to be unreliable in assessing the pharyngeal stage of the swallow.

3.3 Paediatric assessment scales

A number of groups have proposed systematic approaches to reporting observations during the clinical feeding evaluation. The focus of paediatric studies has been on the oral stage, in particular sucking.

3.3.1 Leaf and Gisel Observational System

Leaf and Gisel (1986) developed a system that assesses the sucking component of infant feeding. They suggest the number of sucks used in the time taken to consume 25 ml of infant formula reflects feeding efficiency and can be rated by counting the number of sucks and the length of the sucking time. Reliability of this scoring system was evaluated by scoring the live assessments of 47 normal healthy infants, and comparing these scores with those obtained from video recorded assessments of the same 47 infants. The measures obtained were significantly correlated. In developing the system the number of intervals or sucking bursts required to provide a valid assessment was investigated. The majority of infants completed 3 intervals and for those infants who completed 4 or 5 intervals there was no significant difference in their performance (number of sucks) across the intervals. The authors advocated that these findings might be considered clinically representative of normal feeding.

3.3.2 Neonatal Oral-Motor Assessment Scale

The Neonatal Oral-Motor Assessment Scale, NOMAS (Braun and Meyer Palmer, 1985; Braun and Meyer Palmer, 1990) was developed in 1985 in order to identify and quantify normal and deviant oral motor patterns in neonates. The scale is based on a short observation of non-nutritive sucking followed
by a two-minute observation of a routine feed, whether this is by bottle or breast. The scale consists of 28 items (14 tongue movements and 14 jaw movements), which are coded as normal, disorganised or dysfunctional (Appendix 12). An overall severity rating of normal, disorganised or dysfunctional is given based on the most impaired coding of these individual tongue and jaw items. For example if there is a dysfunctional rating on at least one item the infant is categorised as having a dysfunctional oral motor pattern for feeding. A reliability study of the scale was carried out on 35 infants between post-conceptual ages of 35-49 weeks (without a need for supplemental oxygen and without anatomical anomalies). The assessments were carried out according to a specified protocol and were video-recorded. Experienced therapists, who were trained in the administration of the scale, independently rated the studies. The percent agreement between observers was determined for each of the tongue and jaw items. Eighty to one hundred percent was achieved for 17 of the 28 items. Where agreement was less than 80% the descriptors for these items were modified. The revised scale (Braun and Meyer Palmer, 1990) has not however been subjected to further reliability studies. It was recommended that users of this scale received training and are 80% reliable for individual items and 90% reliable for overall severity rating. Although the scale has been specifically designed for use with premature infants (Case-Smith et al., 1989; VandenBerg, 1990; Palmer, 1993), it has been used with infants with neuromuscular disorders and cerebral palsy (Braun and Meyer Palmer, 1985; Braun and Meyer Palmer, 1990).

3.3.3 Schedule for Oral Motor Assessment

The Schedule for Oral Motor Assessment (SOMA) (Reilly et al., 1995; Skuse et al., 1995) was developed to record objectively oral motor skills in infants aged between 8 and 24 months and is the only validated, standardised, published rating scale. It aims to identify areas of dysfunction that contribute to feeding difficulties. The procedure involves a structured feeding assessment in which a variety of different foods are presented to the infant in
a standardised manner. Six textures are assessed: liquid (breast or bottle, trainer beaker, cup and straw drinking), puree, semi-solid, and two different solids (crackers and dried fruit). Each of these textures, referred to as oral motor challenge categories, is rated in terms of **functional areas**, referring to the muscle group or structure being investigated, **functional units**, referring to the activity the muscle group(s) or structure(s) perform, and **discrete oral motor skills**, referring to the individual oral motor movements that prevent food loss. The SOMA has been shown to be a reliable and valid assessment of infant oral motor function (Reilly et al., 1995; Skuse et al., 1995). A shortened screening assessment has been developed and shown to have positive predictive validity greater than 90% and sensitivity greater than 85% for the detection of infants with significant oral motor dysfunction. The SOMA has the advantage that it can be used in children with mild to severe oral motor dysfunction and in infants with a variety of disorders, including those with non-organic failure to thrive (Mathisen et al., 1999; Reilly et al., 1999).

### 3.3.4 Oral Motor/Feeding Rating Scale

The Oral Motor/Feeding Rating Scale systematically reports observations made during feeding in children of twelve months of age and over (Jelm, 1990). The scale provides a structured approach to evaluating lip/cheek, tongue and jaw movements for drinking (breast, bottle, cup and straw) and eating (spoon, biting soft and hard cookies and chewing). Oral motor function is scored on a scale of 0-5. These scores are allocated by the clinician based on what percentage of the child's feeding pattern is atypical or abnormal. A score of 0 indicates normal function, whereas a score of 5 would indicate that abnormal movement patterns are consistently observed and interfere with feeding function in more than 75% of observations. While the scale is recommended for use with children who are 12 months of age and over, it assesses breast and bottle-feeding and might therefore be used with infants. No normative data are available, and validation and standardisation
have not been carried out. The scale relies highly on the therapist's knowledge of normal development and is subjective in nature.

3.3.5 Paediatric Oral Skills Package

The Paediatric Oral Skills Package (Brindley et al, 1996) was developed to provide a checklist for use with infants and children between birth and 16 years of age. It allows description of oral problems, changes in the function areas assessed over time, and indicates areas requiring therapy or management. Twelve function areas can be assessed including posture, reflexes and reactions, breathing, voice, orofacial structure and function, eating, drinking, movement, articulation of sounds, contrast of phonetic features, teeth/dentition and saliva control. Within each of these areas a level of assessment is selected (a) where the child is not attempting specific oral tasks, (b) where the child is attempting oral tasks and (c) in a situation where the child is eating or drinking. Skills within each area are evaluated using a combination of carer questions, observations and specified tasks. Specific instructions and guidelines are provided ensuring consistency in scoring. Guidelines regarding normal development are provided as appropriate and instructions for scoring are generally explicit. Unfortunately reliability and validity studies have not been carried out. The scale has not been used in any published studies to date.

3.4 Diagnostic tests and procedures

Several diagnostic procedures are routinely used to supplement the clinical evaluation. In isolation these techniques do not provide definitive information about the infant's feeding status but in conjunction with the clinical evaluation can provide information about function and physiological changes.
3.4.1 Pulse oximetry

Oximetry, the measurement of haemoglobin saturation with oxygen, has been used in the assessment of feeding and swallowing. The usual technique is pulse oximetry in which the level of saturation is determined in capillary blood in a finger or toe by means of an external sensor. The technique can be particularly useful when working with infants. It can be particularly difficult to judge oxygen saturation in infants as they may look “pink”, yet have low oxygen saturation (Hays, 1987). The system has the advantage of being a transportable, non-invasive technique. Sensors can be left in place for extended periods of time allowing ongoing monitoring. Pulse oximetry has been found to monitor oxygen saturation levels in infants reliably (Hay et al., 1989). However it can be sensitive to movement artefact (Hays, 1987; Moyle, 1996) and can be affected by jaundice (Hays, 1987), skin pigmentation (Emery, 1987), and external machines such as infrared heating sources (Hays, 1987).

Oxygen saturation is expressed as a percentage. Although measures of 95% are considered normal it is essential to collect a baseline measure for individual infants before feeding is commenced. The literature suggests a drop of 4% or more below the baseline is significant (Sullivan, 1985).

Several studies have investigated the usefulness of pulse oximetry in identifying aspiration in adults and there remains controversy (Brown et al., 1983; Rogers et al., 1993a; Rogers et al., 1993b; Zaidi et al., 1995; Collins and Bakheit, 1997; Sellars et al., 1998; Sherman et al., 1999; Colodny, 2000; Leder, 2000).

Sellars et al (1998) reported a study in which 6 adults, considered to be at risk of aspiration and who were undergoing videofluoroscopic assessment (Section 3.4.7) of swallowing, underwent simultaneous pulse oximetry during and for two minutes after the assessment. Five of the 6 patients had medical histories of respiratory difficulties or lesions in the respiratory centres in the
brainstem. A control group of healthy adults underwent pulse oximetry assessment, but not videofluoroscopy as it was assumed they did not aspirate. No significant differences in average oxygen saturation levels were found between the patient and control groups on baseline measures. However, a small but significant decrease in oxygen saturation was found after videofluoroscopic assessment of the patient group. There was no decrease in oxygen saturation of the control group after oral intake. Of the 6 patients assessed, 4 demonstrated aspiration and 2 of these also showed evidence of oxygen saturation drops of greater than 4%, which is considered significant (Sullivan, 1985). However, both the patients who did not aspirate also showed small changes in oxygen saturation levels. The results were therefore inconclusive with desaturation observed in 2 of 4 cases where aspiration was confirmed but also in the 2 cases where aspiration was not evident. While this study did have a control group for comparison the numbers were small and the results should be regarded with caution.

Following this report Sherman et al (1999) undertook a similar study on a larger group of 46 dysphagic patients with suspected aspiration. Following videofluoroscopic assessment, the patients were grouped according to degree of aspiration. Group 1 included patients who aspirated, group 2 patients who demonstrated laryngeal penetration but did not clear it from their airway (e.g. with coughing), group 3 demonstrated laryngeal penetration but were able to clear it and group 4, where no laryngeal penetration was evident. A significant association of oxygen desaturation with aspiration status was observed. Those patients found to aspirate showed a significantly greater decline in oxygen saturation than those patients who demonstrated laryngeal penetration and cleared it or who did not show laryngeal penetration. Similarly those who demonstrated laryngeal penetration without clearance had a significantly greater decline in oxygen saturation than those who did not show penetration. These results supported those of Sellars et al (1998), that pulse oximetry was useful in detecting patients who aspirated or who demonstrated laryngeal penetration without clearance.
Leder (2000) studied 60 consecutive adult cases referred for dysphagia assessment. All were inpatients on Intensive Care Units and were receiving nil by mouth. All patients underwent Fibreoptic Endoscopic Evaluation of Swallowing Safety (FEESS) (see below) with simultaneous oximetry, heart rate and blood pressure monitoring. Following assessment patients were allocated to one of four groups. Group 1 consisted of those patients who required no supplementary oxygen, and who did not aspirate; Group 2, those who did not require supplementary oxygen but who did aspirate; Group 3, those who required supplementary oxygen and did not aspirate and Group 4 included those who required supplementary oxygen and who did aspirate during the assessment. They found all patients had significantly higher heart rates and blood pressure during FEESS, both of which lowered following the procedure, but there was no significant difference in the oxygen saturation levels whether aspiration was present or not. In contrast to the previous two studies, they concluded that use of oximetry, heart rate and blood pressure monitoring as indirect markers of aspiration were not useful.

Pulse oximetry has been applied to the paediatric population by Rogers et al (1993a) who reported 5 cases of children with severe cerebral palsy and feeding difficulties. All 5 children underwent clinical assessment, including monitoring by pulse oximetry. Four of the children had a history of and demonstrated clinical signs suggestive of risk of aspiration. These children showed drops in their oxygen saturation. Videofluoroscopic assessment of feeding was undertaken. None of the children assessed with videofluoroscopy were found to aspirate during assessment but all demonstrated patterns of dysphagia that have been associated with aspiration (Griggs et al., 1989; Dodds et al., 1990). The authors concluded that the episodes of hypoxemia evident in their cohort were probably related to aspiration. This study suggests pulse oximetry may be clinically useful in the paediatric population, but warrants further research.
In summary, there is controversy about the usefulness of pulse oximetry as an indicator of aspiration in adults. There is only one study evaluating it's usefulness in paediatrics and while the numbers in this study are small, the preliminary results suggest that it might be a useful technique for identifying children at risk of aspiration.

3.4.2 Cervical auscultation

Cervical auscultation is another technique used frequently to enhance the clinical evaluation of infant and paediatric feeding (Vice et al., 1990; Selley et al., 1990d; Vice et al., 1995) and adult dysphagia (Selley et al., 1990c; Zenner et al., 1995). It involves listening with a stethoscope or microphone placed on the throat, to the sounds generated during swallowing. This technique can be used for different purposes. At a clinical level it is used to aid clinicians in identifying swallows and to aid in identifying pharyngeal "noises" that might be associated with aspiration and pharyngeal residue (Austin, 2001; Zenner et al., 1995). However in research studies swallow sounds are analysed in greater detail as is discussed below.

The stethoscope is the instrument most frequently used (Austin, 2001; Hamlet et al., 1988; Hamlet et al., 1994; Zenner et al., 1995), but hearing aid microphones (Lear et al., 1965) standard microphones (Mackowiak et al., 1967; Takahashi et al., 1994a) and accelerometers (Vice et al., 1990; Hamlet et al., 1992; Qureshi et al., 2002) have also been applied. The use of both microphones and accelerometers has the advantage of allowing auditory recording or visual display of the signal (Vice et al., 1990; Selley et al., 1990c; Selley et al., 1990d). Differences have however been reported between auditory and visually represented signals. Vice et al (1990) report visual representation of the signals provides better detail as it captures signals that may not be heard. With computerised synthesiser equipment, swallow sounds can be analysed in terms of discrete sounds of the swallow, bolus transit sounds and breath sounds. Measures, such as the length of swallow and post-swallow expiration breath sounds, can be determined. Visual
representation provides measurements of duration of signal, frequency
characteristics of the signal and amplitude of the waveform (Lear et al., 1965;
Logan and Bosma, 1967; Hamlet et al., 1992; Takahashi et al., 1994a). Normative
data for both adults and infants are available, although sample sizes are small (Vice et al.,
1990; Selley et al., 1990a; Selley et al., 1994).

There is some debate about whether accelerometers (recording information on
vibrations) or microphones (recording acoustic information) provide clearer
information about swallow sounds (Vice et al., 1990; Vice et al., 1995; Qureshi et al.,
2002). While there is no definitive evidence either way, recent studies suggest that
accelerometers might provide more accurate information than microphones as they do not pick up background noise (Qureshi et al., 2002).

Another source of controversy is the site of placement of the acoustic
detector. Different sites have been shown to alter the signal to noise ratio and thus affect amplitude components of the signal. Takahashi et al (1994b) reported the best placement site to be the lateral borders of the trachea, immediately inferior to the cricoid cartilage.

There is however, no clear or well-accepted theory as to the physiological basis of swallow sounds. It has been suggested that swallow sounds may be the result of (a) movement of the mucous membranes as the bolus flows into the pyriform sinuses (Lear et al., 1965) (b) movement of the lower oesophagus as the bolus approaches the PE segment, (c) movement of the hyoid, larynx or epiglottis (Mackowiak et al., 1967; Hamlet et al., 1990; Takahashi et al., 1994b) or (d) bolus movement through the pharynx (Hamlet et al., 1992). Cichero and Murdoch (1998) proposed a theory that swallow sounds are generated through vibrations set up by a pump and valve system within the vocal tract. They suggest the perceptual characteristics of swallow sounds may therefore change in accordance with alterations of the configurations of the pharynx. This suggests that where there are structural
abnormalities comparisons with norms may be inappropriate. It also raises
the question as to whether structural abnormalities such as cleft palate might
alter the perceptual characteristics of swallowing sounds.

3.4.3 Exeter Dysphagia Assessment Technique

The Exeter Dysphagia Assessment Technique (EDAT) (Selley et al., 1990a) is
a non-invasive objective assessment method involving simultaneous
recording and timing of respiratory patterns and swallow sounds during
drinking. The timing of the bolus entering the mouth is recorded using an
electronic switch activated by contact of a spoon with the lip. Swallow
sounds are recorded by a microphone positioned below the angle of the
mandible. These are confirmed by observer visualisation of laryngeal
elevation associated with the swallow. Respiratory airflow direction is
recorded using a nasal catheter. These simultaneous recordings allow the
measurement of swallow events including the pattern of inspiration/exhalation before, during and after the swallow, and oral and
pharyngeal transit times. Selley et al (1990a) presented data on healthy
children (25, aged between 2 and 11 years and 23 senior school aged children
aged between 11 and 18, 15 young adults and 18 elderly adults). These were
compared with data on 16 adults with neurological impairment. Reliability
and validity data were not provided. In summary, this technique provides
useful information about the co-ordination of swallow and respiration for
spoon feeding and drinking, however its reliance on the observers visual
confirmation of swallows would make it difficult to use with small children
and may decrease its reliability.

Selley et al (1990a) adapted their initial version of the EDAT to allow
assessment of infant feeding. By replacing the spoon with a modified bottle
incorporating a sensitive pressure transducer positioned just through the teat
they could monitor intra-oral pressures and volumes of fluid taken.
Additionally a stretch transducer was attached between the infant’s forehead and chin allowing monitoring of jaw movements. Synchronous data of nasal flow timing and volume of milk taken, intra-oral pressures during sucking, swallow sounds, and jaw movements were therefore available in printed form and on tape. The procedure was cumbersome but was reported to be well tolerated by the study cohort of 20 normal newborn infants. The adapted EDAT provides detailed information about timing and synchronisation of the suck, swallow and breathe triad, but the measurements and analysis of the length of sucks or swallows and suck swallow ratios, are not automated and must therefore be time-consuming with potential for error.

3.4.4 Intra-oral visualisation during suckle feeding

In assessing the oral stage of infant bottle-feeding, Eishima (1991) used a camera positioned inside an infant feeding bottle allowing intra-oral movements to be recorded during nutritive and non-nutritive sucking. Simultaneous video recordings of the infant were also made allowing visualisation of lip movements and seal, tongue protrusion, tongue grooving around the teat, “peristaltic” movements of the tongue and the synchronous movements of the jaws and tongue. Based on the findings, Eishima (1991) formulated hypotheses about the generation of negative pressure. This technique was used on 50 infants who were considered to be normal. While it provided visualisation of the oral stage no measurements were taken and therefore results were subjective.

3.4.5 Measurement of sucking using pressure transducers

An alternative way of evaluating the oral stage objectively is an examination of the rhythm, rate and strength of sucking.

As early as 1865, Herz used a mercury manometer attached to a teat to measure the negative pressures generated by infants during sucking. Basch (1893) modified this technique to allow continuous monitoring of sucking using a water manometer and kymograph. Jenson (1932) measured changes
in pressure in an inverted infant's feeding bottle as he sucked. Others have continued this work and have developed modified dummies, bottles and teats to measure the intra oral pressures generated during non-nutritive and nutritive sucking (Colley and Creamer, 1958; Halverson, 1938; Kron et al., 1963; Wolff, 1968; Drake and Wilson, 1983; Jain et al., 1987; Weaver and Anderson, 1988; Choi et al., 1991).

Some of these techniques have been further modified allowing researchers to manipulate different stages of the feeding process and thereby to assess more closely infants' abilities to compensate and modify their participation in this process. Kron et al (1963) in a study of the measurement of intra-oral pressures generated during sucking, modified an apparatus to control the flow of fluid as the infants sucked. Building further on this work, Medoff-Cooper et al (1993) reported the development of a similar system incorporating a modified teat (connected to a pressure transducer, thus allowing measurement of intra-oral pressure) with a calibrated capillary tube which allowed control of the flow of fluid into the teat. These measurements were then linked to vital sign recordings (heart rate, blood pressure and oxygen saturation) to provide a more complete assessment of the infants' state and stress during feeding. The co-ordination of respiration, measured with chest impedance bands or nasal flow meters and intra-oral pressure generation, has also been investigated (Wolff, 1968; Mathew et al., 1985a; Mathew et al., 1985b; Mathew et al., 1985c; Koenig et al., 1990; Craig et al., 1999).

This early work has been fundamental in setting the scene for research in this area. These laboratory based systems provide detailed information about the patterns of sucking but require specialist equipment, which is not commercially and therefore not routinely available. A lack of normative data obtained with these systems limits their clinical and research potential at this stage but with ongoing research and development, normative data will be available in the future (Personal communication; Gewolb, 2002).
3.4.6 Whitney strain gauge

Sucking behaviours have also been measured using stretch sensitive gauges (Whitney Gauge) placed under the infant's chin, thus allowing measurement of jaw movements as the infant lowers and raises the jaw during sucking. This technique has the advantage of being able to be used with both breast and bottle-fed infants (deMonterice et al., 1992; Ramsay and Gisel, 1996). However its application is limited to the evaluation of sucking patterns only and does not allow evaluation of the co-ordination of sucking, swallowing and/or breathing.

3.4.7 Videofluoroscopic assessment of swallowing

Videofluoroscopy (VF) is used widely in the assessment of dysphagia in paediatrics and adult populations. Many different videofluoroscopic assessment protocols and procedures exist, yet few have undergone validation or reliability studies. Despite this, the technique has been adopted as the gold standard. VF is used extensively clinically and for research (Griggs et al., 1989; Morton et al., 1993; Friedman and Frazier, 2000). This radiographic technique was first described by Ardran et al in 1958 and is also known as Cineradiography, Cookie Swallow and Modified Barium Swallow (Ardran et al., 1958a; Ardran et al., 1958b; Logemann, 1983; Arvedson and Lefton-Greif, 1998).

The procedure involves the infant or child being fed liquids and/or solids as appropriate. In order to ensure that the feeding behaviours seen are typical for the child, the usual feeding environment is simulated as closely as possible with respect to positioning and feeding techniques. Liquid barium is mixed with an appropriate fluid or food to replicate the textures being assessed. Although there is some debate about the validity of replicating textures, since barium is more viscous than everyday fluids and foods, at this stage there are no alternatives (Arvedson and Lefton-Greif, 1998). The study images are
collected via a fluoroscope, allowing real-time video images to be recorded for later review and analysis.

VF allows observation of patterns of movement of oral and pharyngeal structures. At the oral stage these include the extent of lip closure, tongue configuration and range of movement. At the pharyngeal stage they include movement patterns related to pharyngeal clearance, base of tongue retraction, pharyngeal wall contraction, cricopharyngeal relaxation, airway protection including the extent of laryngeal elevation, epiglottic movement, the amount and extent of aspiration and whether aspiration occurs before, during or after triggering of the swallow.

Observations regarding the amount and place of residue are also made. It is suggested these include an estimate of the amount of residue as a percentage of the bolus (Logemann, 1993).

Measures of the duration of the oral stage (oral transit time) and pharyngeal stage (pharyngeal transit time) can be taken. More specifically the duration of specific oral movements such as palatal elevation can be measured. At the pharyngeal stage these include measurement of the delay time for triggering of the swallow, duration of laryngeal elevation and duration of cricopharyngeal relaxation (Logemann, 1993).

Based on these observations it is possible to hypothesize reasons for the impairment and to test and evaluate the effectiveness of management strategies (Logemann, 1993).

Documentation and rating of videofluoroscopic studies varies. Scales may report selected patterns of movement and timing measures as described above, or may provide a more general score of degree of impairment.

In adult studies, detailed scales allowing standardised reporting of individual movement patterns have been reported (Logemann, 1983; Ekberg et al.,
1988; Palmer et al., 1993; Wilcox et al., 1996; Kuhlemeier et al., 1998; Scott et al., 1998; McCullough et al., 2000). The inter-rater reliability of these scales varies (Rosenbek et al., 1996; Kuhlemeier et al., 1998; Scott et al., 1998; Wooi et al., 2001).

Although there is a published format for rating paediatric videofluoroscopic studies which are based on adult scales (Arvedson and Lefton-Greif, 1998) there are currently no standardised or validated scales. There is however a multi-centre trial in progress investigating the reliability of experienced clinicians rating videofluoroscopic studies (personal communication; Reilly 2002).

The Dysphagia Outcome and Severity Scale (DOSS) (1999) is an example of a scale that provides an overall score of degree of impairment based on videofluoroscopic studies. The seven-point scale systematically rates the functional severity of dysphagia. Instead of reporting individual movement patterns the scale allocates the patient to 1 of 7 levels, where level 7 places the patient as requiring a normal diet, with no extra time or strategies required, to Level 1 where the patient demonstrates severe dysphagia, is unable to take anything safely orally and may exhibit features such as "severe retention in the pharynx or silent aspiration".

Ryan et al (2001) applied this scale to the paediatric population. Four experienced dysphagia-trained speech and language therapists rated VF studies of 20 consecutive patients of mixed aetiology and severity. Seventy-percent reliability was achieved with all therapists agreeing in 40% cases, and 3 out of 4 therapists agreeing in 30% of the remaining cases. There were no significant differences related to the age of the child or the speech and language therapist rating the study. There was some inconsistency in rating specific items. These items were related to extent and management of pharyngeal residue. The items are now being modified and a repeat reliability study is planned.
3.4.8 Fibreoptic Endoscopic Evaluation of Swallowing Safety

An alternative dysphagia assessment technique used widely in the adult population (Langmore et al., 1988; Bastian, 1993) and becoming more popular in the paediatric population (Hartnick et al., 2000) is the use of flexible nasendoscopy or Fibreoptic Endoscopic Evaluation of Swallowing Safety (FEESS). In this procedure a flexible nasendoscope is inserted to either the level of the uvula and/or the level of the epiglottis allowing visualisation of the pharynx, larynx and airway before and after a swallow. The patient is given appropriate foods or fluids, which are dyed with blue or green food colouring making them easier to visualise. The study is video-recorded for later analysis and feedback. Unfortunately as the swallow occurs and the pharynx contracts the view is obscured. However patterns of movement, presence and extent of aspiration and residue before and after the swallow can be viewed.

Protocols described by Bastian (1993), Hartnick (2000) and Langmore (1991) include observations of the pharyngeal and laryngeal structure and the range and co-ordination of their movements (Langmore et al., 1991; Bastian, 1993; Hartnick et al., 2000). Assessment using one or more trial swallows with observations of pharyngeal function can be made. Aspects of the pharyngeal stage that can be commented on include (a) the point at which the swallow is triggered, (b) laryngeal penetration/aspiration prior to triggering of the swallow, (c) extent and place of pharyngeal residue, (d) clearance of pharyngeal residue, (e) laryngeal penetration/aspiration post swallow and (e) clearance of laryngeal penetration and/or aspiration. Management strategies can be trialled and evaluated. This technique provides useful biofeedback for cognitively able patients and carers.

There has been debate about the reliability of FEESS in assessing the pharyngeal stage of swallowing. Comparative studies of FEESS and VF have found it to be a reliable technique for identifying the presence or absence of
abnormal swallow events, and identification of pharyngeal residue, laryngeal penetration and aspiration (Langmore et al., 1991; Madden et al., 2000).

FEESS has been applied successfully to the paediatric population (Hartnick et al., 2000). This study evaluated 643 assessments performed on 568 children. The median age of the children was 2.5 years (range 3 days to 21 years). There was a wide range of medical aetiologies including structural abnormalities of the upper aero-digestive tract or airway, neurologic diagnoses, gastro-oesophageal diagnoses, genetic syndromes, pulmonary dysfunction, prematurity, cardiovascular abnormalities and metabolic problems. Unfortunately the swallowing diagnoses reported were medically based rather than physiologically based. Tolerance and child co-operation was not reported, but the authors did comment that the procedure was time-intensive and that some children would not co-operate for the feeding trial.

There are several areas of concern with regard to the use of this technique in the adult population. These include patient tolerance of the scope in situ, the effect of the scope on the swallow mechanism, the effect of the local anaesthetic on the swallow and the effect of the procedure on the patient in terms of physiological measures (Leder et al., 1997). The procedure also requires expensive equipment and training of the operator.

In recent years it has been suggested that laryngopharyngeal sensory testing (FEEST) is useful in determining risk of aspiration in adults. This has been investigated in adults by Aviv et al (2000a; 2000b) and in a paediatric population by Link et al (2000). The procedure involves delivering sensory stimuli in the form of short air-bursts, at variable intensities through equipment especially designed for this purpose. The air pulse stimuli were delivered to the mucosa innervated by the superior laryngeal nerve. Aviv et al reported the procedure was well tolerated and there were no serious complications associated with the 500 studies they completed. Similarly Link et al found that sensory testing was possible in 92.5% of their paediatric
subjects. The remaining studies could not be administered due to altered anatomy or excessive gagging or crying, and they suggested structural abnormalities may be a contra-indication.

One of the main areas of concern with regards to FEESS and FEESST is the safety of the procedure. The number of complications reported is few, but includes epistaxis, changes in heart rate and blood pressure, airway compromise and laryngospasm (Link et al., 2000; Aviv et al., 2000a). Tolerance of both FEESS and FEESST (in both adults and paediatrics) is reported to be high. In the adult population 81% of patients rated the level of discomfort as none or mild and 99% of patients agreed to repeat assessments if required (Aviv et al., 1999; Aviv et al., 2000a).

FEESS and FEESST provide descriptive information about aspects of the pharyngeal stage of the swallow including anatomy, velopharyngeal function, airway protection, laryngeal penetration and aspiration and pharyngeal residue. Although it has been shown to be tolerated in the paediatric population it is not routinely used. The results obtained have not been compared with VF and the question of the effect of the presence of the nasendoscope on swallowing and sucking patterns in particular has yet to be investigated.
This chapter is concerned with the development of the Great Ormond Street Measurement of Infant Feeding, a technique for assessing objectively several aspects of infant bottle-feeding. The hardware and software components and their use are described. The analysis package is demonstrated. The application of the technique to a healthy non-cleft (normal) cohort is described and the results obtained are reported. The reliability of the technique, as evaluated on this non-cleft cohort, is reported.

4.1 Reasons for development of the GOSMIF

As discussed in chapter 3, although there are numerous techniques available for the objective assessment of infant feeding, many are not readily transferable to everyday clinical use. Some of the techniques rely on the expertise of specialist staff and equipment, access to which may necessitate that the assessments be scheduled within specialist clinics such as X-ray/videofluoroscopy. If the appointment time does not coincide with a routine feed time, the infant may be unco-operative. An unfamiliar environment may further compound the problem. For example, the infant is likely to be positioned in an unfamiliar chair, fed an unfamiliar food/substance and could be intimidated by large noisy equipment used in the procedure. Infants might require several different assessment procedures in order to provide a comprehensive evaluation of the four stages of feeding.
Few of these procedures can be carried out simultaneously using currently available techniques, and consequently a series of investigations is required that could be protracted, intrusive and expensive in terms of demands on staff. Usually there are waiting lists for these investigations and there may be some time between appointments. Given the changes in infant feeding over time or associated with appetite or medication routine, a series of consecutive investigations may not provide an accurate or comparable picture of the infant’s feeding capabilities.

One aim of developing the GOSMIF was to objectively evaluate several aspects of infant feeding that are reported to contribute to feeding efficiency. Based on the literature review of infant feeding (Section 2.4) the aspects selected were:

- objective measurement of pressures generated during bottle-feeding
- identification of features of sucking patterns including number of sucks per burst, length of sucking bursts, rate of sucking
- identification of swallows through auscultation
- calculation of suck:swallow ratios
- monitoring of respiration changes during sucking

The system for these measurements would ideally be transportable and easily set up, thus allowing assessments to be carried out in the infant’s home, maternity unit, or hospital clinic. Another essential aspect of the assessment was that it would allow mimicking of the infant’s usual feeding routine as closely as possible, for example, allowing the infant to be fed by his/her carer in their usual manner. It was planned that these assessments would be carried out by the researcher independently.
Given the large amounts of data to be generated by the tool automated analysis would be advantageous.

In order to ensure that complete and representative evaluations of the infants’ feeding patterns were obtained a standardised oral motor assessment rating scale, the NOMAS was to be administered (Section 3.2) and the infant’s behavioural state monitored throughout the assessment, based on video-recordings of the infant.

The GOSMIF was developed to meet these requirements.

4.2 Components of the GOSMIF

The basic design of the GOSMIF is schematically demonstrated in the block diagram in Figure 17 and photograph in Figure 18.
4.2.1 Hardware

The GOSMIF system incorporates a rigid feeding bottle, which has been modified by drilling a hole in the base, permitting installation of a pressure transducer (Honeywell 24PC 1psi piezoresistive sensor; Appendix 1). This measures pressure changes that are generated within the bottle during sucking. NUK medium flow orthodontic teats are used as the holes are laser cut and are therefore consistent in size (Mathew, 1988). The size of the teat used is matched as near as possible to the size used routinely by the infant. The teats are unvented (Figure 19) ensuring accurate monitoring of pressure changes within the bottle.
Swallow sounds are recorded with both an accelerometer (Knowles BU1771 ceramic vibration transducer; Appendix 2) and microphone unit (Knowles EK3132 Electret Microphone; Appendix 3), which is taped on the infant’s neck (Vice et al, 1990).

A standard respiration band (Respirtrace expanding impedance, Appendix 4) is placed around the infant’s chest allowing monitoring of respiration patterns.

Video recording is carried out using a Sony DCR-TRV900E Digital camera (mounted on a tripod).

The notebook computer used is a Dell Latitude 366XT Pentium 2 with the following cards installed:
- Nogotch Capture Vision PCMCIA type2 Video capture card; Maximum resolution 384x288

- Amplicon PCM DAS08 PCMCIA type 2 ADC Card, 12 bit resolution

Details of the electronic design are included in the paper by Veness et al. 2002 (Appendix 5).

4.2.2 Software

The software for capturing, displaying, recording and analysing data was written using Borland’s Windows development tool, Delphi (Veness et al 2002).

The software has four basic windows: the data capture, the data review, the data analysis and the automatic analysis windows.

- Data capture window

The data capture window (Figure 20) controls both PCMCIA cards for simultaneous data and video capture.

The capture window controls the collection of data over a specified period which is selected by the user (Figure 20; 1). Patient identification details (Figure 20; 2) are entered and the location for storage selected (Figure 20; 3). This screen also allows the user to monitor the signals (Figure 20; 4) and video image (Figure 20; 5) before recording is started. Once the user is confident that the signals are stabilised the recording is started with activation of the “start recording button” (Figure 20; 6). When a recording session is in progress the waveforms and video are simultaneously updated in Figure 20; 4 and 20; 5.
Recording can be stopped at any time using the “stop recording button” (Figure 20; 7). This window is exited using the “exit button” (Figure 20; 8).

On completion of the data acquisition, the data signals are streamed to files on the hard disc drive. The video data is saved to disc a frame at a time during the capture.

Five files are generated for each study and are named as follows:

PatientnameRES.Dat - respiration
PatientnameSWSD.Dat - swallow sounds
PatientnameSWVB.Dat - swallow vibrations
PatientnameSUPS.Dat - sucking pressures
Patientname.Avi - video clip
■ Data review window

Files for review are selected and opened using the file open button shown in Figure 21; 1. This displays a subwindow shown in Figure 22, allowing the user to select one of the four displayed files for the selected patient. On re-entry to the main window, the data from all 5 files (4 data files + video file) for the selected patient are opened.
Figure 21: Data review window

Figure 22: Loading files for analysis
The data review window with patient data is displayed (Figure 23). Signals from the entire study are displayed in the top half of the screen. The data within the purple band (Figure 23; 1) is magnified and displayed in the lower screen (Figure 23; 9). The simultaneous video image is displayed in the right hand side of the lower screen. The length of the feed is displayed in the top right hand corner (Figure 23; 2). The respiration trace is shown in red (Figure 23; 3), the swallow sound and vibration traces in green and blue (Figure 23; 4 & 5), and the sucking pressures in black (Figure 23; 6). In this screen, the test can be replayed in real time using the play (Figure 23; 7) and stop (Figure 23; 8) buttons, allowing verification, based on the video clip, that sections of the test being studied are those where the infant is feeding and not sections where the infants may be fidgeting or fussing. Alternatively a small amount of information can be selected by moving the purple band (Figure 23; 1) to the desired point. The information within the purple band is then shown in an expanded form in the lower window (Figure 23; 9).

Figure 23: Data review window
**Data analysis window**

When pressures and waveform measurements are required the analysis window (Figure 24) is entered by selecting the key shown in Figure 23;10. The part of the test displayed in this window is adjusted using the scroll bar at the top of the page (Figure 24; 1). This has the major advantage of allowing one or several entire sucking bursts to be viewed, as seen in Figure 24. In this screen, cursors can be dragged across to enable amplitude or frequency measurements (Figure 24; 2). The time point into the assessment is shown at Figure 24; 3. Numerical measurements at the cursor are displayed below (Figure 24; 4). The display gain can also be increased by 4 levels of magnification (Figure 24; 5) to allow amplified viewing of small signals.
Figure 24: Data analysis window

In order for the automated aspect of the software to run the user is required to manually select the beginning and end of the sucking burst to be rated. Two cursors (Figure 25; 1 & 2) can be manipulated to mark/select the sucking bursts. To ensure consistency specific rules were created for selection of the sucking bursts for analysis.
These rules were:

- Sucking bursts selected for rating would be the first three complete bursts during which the infant is not crying, fussing or wriggling and has the bottle in his/her mouth, confirmed using the video images.

- A sucking burst is identified as a sequence of at least 3 events (consecutive peaks/troughs) in the pressure tracing with no pauses longer than 2 seconds.

Three examples of clearly identified sucking bursts are shown in Figure 26.

- The beginning of each burst is defined as the first identifiable peak in a sucking burst.

- The end of the sucking burst is marked by the last peak in the sucking burst.

The beginning and end markers are demonstrated in Figure 27; 1 & 2.
Figure 25: Data analysis window
Figure 26: Identification of sucking bursts

Figure 27: Marking the beginning and end of a sucking burst
Before entering the automated analysis window the user is also required to select whether the sound or vibration signals will be used in identification of swallows (Figure 25; 3). Since the development of the GOSMIF, recent studies suggest that the accelerometer is the more reliable option and these are the signals used therefore in this study (Gewolb et al., 2001; Qureshi et al., 2002). However there may be instances where this is unclear or absent (due to equipment failure). In these cases the microphone (sound) signals will be used.

The automated analysis window is entered by clicking the “signal analysis button” (Figure 25; 3). This brings up a subscreen, where the user is asked “Is sucking burst selected between cursors?” When the user agrees the automatic analysis window is entered.
Automatic analysis window

The automatic analysis result window is shown in Figure 28.

This window displays a smoothed version of the previously selected portion of the sucking pressure waveforms. This is necessary because the pressure trace is affected by noise. This "noisy trace" is smoothed with a numerical low pass filter. This filter moves continuously along the trace averaging 50 adjacent data points. These 50 points cover 80 msecs, a much shorter period than a typical suck of 1 second. If however, further smoothing is required, the smoothing button (Figure 28; 1) can be selected, increasing or decreasing the smoothing. This allows a judgment to be made (in conjunction with...
video images) as to whether the identification of sucks is improved. For consistency in this study the smoothing level has been set at 10% for all infants. Ten percent was selected after examination of a subset of studies (both cleft and non cleft infants).

In order to check that this level was appropriate, further analysis was carried out with data collected on a non cleft cohort (Section 4.5). For each non cleft infant study a visible sucking burst both on the pressure trace and video image was selected. Within the chosen sucking burst, the clearly identified sucks were marked on a print out. Automated analysis (identification of sucks) was then carried out at the threshold levels of 25%, 10% and 5%. Identification of sucks was then compared with the clinically identified sucks. Complete (100%) agreement was achieved at the 10% threshold. Additionally, comparisons were made on individual suck measures (average peak to peak interval and rate of sucking) with a recently published paper evaluating these aspects (Qureshi et al., 2002). At the 10% threshold the average peak to peak intervals and rate of sucking were similar (100% within 1 SD) to those reported by Qureshi et al. In addition, an automarking algorithm places marks on each suck trough as seen in Figure 28; 2. The suck trough identification begins with an automatic scanning through all data points in the selected section of the pressure trace. This scanning identifies the highest and lowest points in the section. The range of pressure is then calculated by subtracting the lowest from the highest value. The peak threshold register is then set to a proportion of that range (determined by where the user has set the scroll bar control). Any troughs where the fall is at least 70% of the average wave peak are marked as sucks. A detailed algorithm flow diagram is provided in Appendix 6.

The swallow count is entered by the user (Figure; 28.11), based on the findings of Vice et al (1990). Vice et al report three main components to swallow sounds, these being the initial discrete sounds (IDS), the bolus transit sounds (BTS) and final discrete sounds (FDS). They suggest that the main
acoustic sounds detected during swallowing, related to the bolus transit sounds (BTS). This is demonstrated in Figure 29 (Vice et al., 1990).

Figure 29: Illustration of swallow sounds identified in sucking burst (taken from Vice et al., 1990)
Given that in this study the purpose of recording swallow sounds was to assist in the identification of when swallows occur, it was decided that their identification would be based on bolus transit sounds (BTS). Unfortunately there are no consistent normative measures of the amplitude of an accelerometer trace associated with BTS. There was however remarkable similarity between the accelerometer traces obtained with the GOSMIF and those published by several authors (Vice et al., 1990; Selley et al., 1990a; Selley et al., 1994; Heinz et al., 1994). The decision about which sounds are BTS is based on the example shown in Figure 30.

* BTS indicating swallows

Figure 30: Example of accelerometer trace used in the identification of Bolus Transit Sounds (BTS)

It is reported that BTS vary in amplitude and pattern, probably in relation to the physical character of the bolus and whether or not air is mixed with the bolus. Given the altered anatomy and feeding patterns of infants with CL and/or P, it could not be assumed that the BTS traces would be similar for this group of infants. In order to match the accelerometer trace to bolus transfer through the pharynx, the GOSMIF was wired into the X-ray
screening equipment used for videofluoroscopic assessments of feeding, rather than to the routinely used camera. As a result, an x-ray image is seen in the lower right hand corner of the data review screen instead of a video image of the infant (Figure 31). One such study was administered. While the findings of this study cannot be generalised they did suggest that the BTS were similar to those reported for non-cleft infants (Figure 29). Figure 31 shows a screen shot taken of the bolus in the pharynx and the simultaneous accelerometer trace, closely matching that reported for non-cleft infants. This pattern was consistent for all swallows seen on this videofluoroscopy study. All accelerometer traces that matched this pattern were confirmed as swallows with videofluoroscopy images. Figure 32 shows where BTSs were confirmed with videofluoroscopy.

Therefore the same model for identification of BTS was applied for both non-cleft infants and those with CL and/or P.
GOSMIF summary screen, showing bolus in pharynx and simultaneous accelerometer trace, taken during simultaneously GOSMIF and videofluoroscopic assessment of swallowing.

Figure 31
Figure 32: GOSMIF summary screen showing accelerometer traces where swallows were confirmed with videofluoroscopy
Automated calculations are made for a series of “sucking burst results” (Figure 28; 3) and “individual suck results “ (Figure 28; 4).

The “sucking burst measures” reported are:

- Where the measure burst began during the test (in minutes and seconds)
- The length of the sucking burst being rated (in seconds)
- The average pressure generated within the bottle during the burst (in mmH2O) shown numerically (Figure 28; 5) and by a dotted red line (Figure 28; 6)
- The percentage of the pressures generated that were above atmospheric pressure (in %). Atmospheric pressure is shown by the solid black line (Figure 28; 7)
- The percentage of pressures generated that were above the baseline pressure in the bottle at the beginning of the burst (in %). The baseline pressure is shown by the hatched pink line (Figure 28; 8)

The “individual suck results” reported are:

- The number of sucks per minute or rate of sucking (Figure 28; 9)
- The average length of individual sucks (Figure 28; 10) as measured by the time from one peak to the next (Qureshi et al., 2002).
- The number of swallows (Figure 28; 11) entered manually
- The suck swallow ratio (Figure 28; 12)

The automatic analysis ensures reproducibility between studies and enables data analysis to be carried out very swiftly.

The results from this screen can be printed using the button shown in Figure 28; 13.
4.3 Storage of data

The video data is stored in the computer and recorded onto tape. Data on sucking pressures, swallow sounds, and respiratory trace, are stored both graphically and on a spreadsheet that can be readily accessed. The data files are large and are therefore written to CD for storage purposes.

4.4 Modifications to the GOSMIF during development

Throughout the development of the GOSMIF only one significant change to the design was required. During development the GOSMIF was trialled on infants with CL and/or P. However, when data collection began with healthy non-cleft infants, they were found to generate intra-bottle pressures that exceeded the capabilities of the GOSMIF. Several changes to the hardware and software were made in an attempt to compensate for this, but a group of infants continued to exceed the scale available resulting in lost sucking burst data. An option was added to allow reduction of the amplitude of the signal by half where necessary. This was done by allowing the user the option of using the full signal amplitude (setting 1) or reducing it by half (setting 2). These settings are set using the switch located on the side of the signal conditioning and amplitude unit (Figure 17). It was also decided that in those cases where data was being lost, the unvented teats (Figure 19) would be replaced with vented teats, which allow airflow through the vent and preventing large negative pressures being generated within the bottle. While this has the disadvantage of not allowing direct comparisons of pressures generated it does allow comparison of patterns and coordination of sucking and swallowing.

4.5 Normative data

A small cohort of non-cleft healthy infants was studied using the GOSMIF to test this new tool and allow comparison of cleft and non-cleft infants.
Twenty infants were recruited from the West Middlesex University Hospital. The mean gestational age of the non-cleft cohort was 39.64 weeks and the mean birth-weight 3.2 kg (Table 2). Ethnic origin and father's occupational rating are presented in Table 2. The mean age at assessment was 3.33 weeks (SD 0.76, Range 1.56 to 4.68).
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Table 2: Demographic details for "non cleft" cohort
The following section summarises the results obtained for the non-cleft infants. Measurements for the 3 sucking bursts rated for each infant are shown graphically on the following scatterplots (Figures 33 to 38). The measurement from the first suck is represented by a square, the second by a circle and the third a cross. The 3 measurements for each infant are shown vertically. The median value of the 3 sucking bursts for each infant was calculated. When summarising values across the group, the median, mean and standard deviations (SD) of the infants' median measurements are reported.
Length of the sucking burst (in seconds)

The median length of sucking bursts was 7.26 seconds and the mean length of sucking bursts was 10.77 seconds (SD 7.46), ranging from 2.90 to 33.59. Qureshi et al (2002) in a similar cohort, report a mean length of sucking burst of 10.14 seconds (SD 8.61) immediately post-natally and 20.99 seconds (SD 22.65) at 1 month of age.

Figure 33: Scatter plot of “length of sucking bursts” of non-cleft infants (3 per infant)
- Average pressure generated within the bottle during the burst (in mmH2O)

The median pressure generated within the bottle was -4.28 mm H2O and the mean pressure generated within the bottle was -8.46 mm H2O (SD 8.60, Range -26.60 to 2.32). As this is a unique piece of equipment, measuring pressures generated in the bottle in contrast to intra-oral pressure measurements which are more commonly reported in the literature (Section 3.4) comparisons with the literature were not possible for this measure.

Figure 34: Scatter plot of “average pressure generation” of non-cleft infants
- Percentage of pressures generated that were above the baseline pressure in the bottle at the beginning of the burst (in %)

The median percent pressure generated within the bottle, above the baseline pressure, was 8.79 and the mean percent pressure generated within the bottle, above the baseline pressure was 26.80 (SD 32.77, Range 0 to 87.80). As previously, comparisons with the literature are not possible for this measure.

Figure 35: Scatter plot of “% pressure generated above baseline pressure in bottle” of non-cleft infants
Number of sucks per minute or rate of sucking

The median rates of sucking ranged from 41.00 to 105.00 sucks per minute with a median rate of 73.70 sucks per minute and a mean rate of 74.16 sucks per minute (SD 18.52). This is similar to those reported by Qureshi et al at 1 month of age (68 sucks per minute) but is different to those they reported at postnatal assessment (55 sucks per minute).

Figure 36: Scatter plot of “rate of sucking” of non-cleft infants
- Length of individual sucks or peak to peak intervals

The median length of individual sucks was 0.81 seconds and the mean length of individual sucks was 0.86 seconds (SD 0.26, Range 0.57 to 1.46). These were very close to Qureshi et al's reported means of 1.1 seconds (SD 0.15) in the immediate post natal period and 0.88 seconds (SD 0.15) at 1 month of age.

Figure 37: Scatter plot of "length of sucks or peak to peak intervals" of non-cleft infants
• Suck swallow ratios

The median and mean suck:swallow ratio was 1 (SD 0.07; range 1.00 to 1.30). There are several reports in the literature supporting this finding (Selley et al., 1990b; Wolff, 1968; Weber et al., 1986; Bu'Lock et al., 1990). Qureshi et al however suggest that this rate increases to 2 or 3 sucks to each swallow as the infant gets older.

![Legend](image)

**Figure 38:** Scatter plot of “suck swallow ratios” of non-cleft infants

NB: There was some overlap in measurements obtained in this scatterplot, giving the impression that 3 sucking bursts were not measured for each infant.
4.6 Reliability

Once sucking bursts have been identified for use in the evaluation, all measures (except for suck swallow ratios) obtained by the GOSMIF are automated thereby ensuring consistency. Rules were developed (Section 4.5) to maximise the likelihood of different users selecting the same sucking bursts and the reliability of these rules was assessed. Ten of the 20 non cleft studies were randomly selected (SPSS 10). Two Speech and Language Therapists, not involved in the trial, but experienced in the assessment of infant feeding problems, were trained to use the analysis aspects of the GOSMIF and to apply the rules for the selection of sucking bursts. Written guidelines were also provided. They, and the researcher, then independently selected sucking bursts for automated analysis from the randomly selected studies.

This data was then evaluated in terms of agreement of selection of sucking bursts.

The choice of start and end times for sucking bursts were compared between raters (Figures 39 and 40). Rater 2 assessed the start of the first sucking burst as being on average 1.1 seconds later than rater 1 (range 1 sec earlier to 12 seconds later), the second sucking burst as being on average 0.5 seconds earlier than rater 1 (range 14 seconds earlier to 7 seconds later) and the third sucking burst as on average 0.1 sec later (range 9 seconds earlier to 10 seconds later). Differences for rater 3 as compared to rater 1 were similar although the ranges were much wider, with the start time for the first sucking burst on average 5.6 seconds later (range 1 sec earlier to 30 seconds later), on average 0.4 seconds earlier for the second burst (range 55 seconds earlier to 26 seconds later) and on average 0.3 seconds later for the third burst (range 39 seconds earlier to 31 seconds later). Rater 2 assessed the end of the first sucking burst as on average 0.97 seconds later than rater 1 (range 0.61 seconds earlier to 7.84 seconds later), the second burst on average 0.26 seconds earlier (range 16.53 seconds earlier to 9.68 seconds later), and the
third burst on average 0.48 seconds earlier (range 11.86 seconds earlier to 7.6 seconds later). End times for the rater 3 were similar. The end point of the first burst was on average 1.19 seconds later than rater 1 (range 46.71 seconds earlier to 33.06 seconds later), the end point of the second burst an average 7.62 seconds earlier than rater 1 (range 38.48 seconds earlier to 28.21 seconds later) and the end point of the third burst an average of 1.19 seconds later (range 52.32 seconds earlier to 41.66 seconds later).

**Figure 39: Beginning of sucking bursts for each rater**
Figure 40: End of sucking bursts for each rater
The length of the sucking bursts also varied (Figure 41). Comparisons were made between measurements obtained by raters 2 and 3 against rater 1. On average the length of sucking burst 1 was rated as 4.53 seconds shorter by rater 2 (range 1.39 seconds longer to 45 seconds shorter) and 4.44 seconds shorter by rater 3 (range 3.06 seconds longer to 47.5 seconds shorter). The second sucking burst was rated on average as 0.25 seconds longer by rater 2 (range 2.68 seconds longer to 2.53 seconds shorter) and on average 2.74 seconds shorter by rater 3 (range 2.69 seconds longer to 30.25 seconds shorter). Similarly sucking burst 3 was rated as on average 0.58 seconds shorter by rater 2 (0.29 seconds longer to 2.4 seconds shorter) and 0.87 seconds longer by rater 3 (10.66 seconds longer to 2.63 seconds shorter).
Figure 41: Scatter plot showing the length of sucking bursts selected by each rater.
Given that there was some inconsistency in the selection of start and end times for sucking bursts between raters, further analyses were undertaken to determine whether any differences between assessment of selected sucking bursts were of clinical relevance. That is, did differences in siting of bursts result in clinically important differences in the automated outcomes generated. Figures 42 to 45 show the differences in rate of sucking, peak to peak intervals, average pressure generation, % pressure generation above baseline pressure and % pressure generation above atmospheric pressure, between raters for each infant. Multilevel models were used to investigate differences between rater and sucking bursts, after accounting for the hierarchical nature of the data (3 raters assessing 3 sucking bursts from each infant) (Goldstein, 1998). Analyses of variance were used to estimate components of variance. The between rater and between sucking burst estimates are presented in Table 3 (Hicks, 1982; Dunn, 1989; Streiner and Norman, 1995). After accounting for the hierarchical nature of the data, multilevel models revealed that the values for peak to peak intervals, average pressure generated within the bottle and percent pressure generated above the baseline pressure in the bottle, tended to increase for later bursts. The differences between the 1st and 3rd bursts were significant for average pressure generated within the bottle. There were no trends or significant differences between raters. No other differences between raters of suck burst number within infant were statistically different.

Overall while there was some discrepancy between the bursts selected by the 3 raters, there was good reliability for the automated measures between raters. Any discrepancies tended to be between bursts, with the 3rd burst yielding higher measures.

The finding that the 3rd bursts rated yielded significantly higher measures than bursts 1 and 2 supports the commonly held view that non cleft infants take several minutes to settle in to a feed. While there was variability in the measures obtained, the majority of the measures still fell within clinically
acceptable ranges and were comparable to results obtained in other studies (Section 4.5).

Figure 42: Scatter plot showing the rate of sucking for each sucking bursts and raters
Figure 43: Scatter plot showing the peak to peak intervals for sucking bursts and raters
Figure 44: Scatter plot showing the average pressure generation for sucking bursts and raters
Figure 45: Scatter plot showing the % of pressure generated above baseline pressure in bottle for sucking bursts and raters
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<td>Average pressure generation</td>
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</tr>
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</table>

*Table 3: Percents of variance attributed to bursts, raters and patients for selected measurements obtained with GOSMIF*
This chapter describes the randomised controlled trial designed to evaluate the effect of pre-surgical orthopaedics (PSO) on feeding.

5.1 Hypothesis

The null hypothesis was that there would be no significant difference in feeding patterns and growth of infants (with ICP or UCLP) managed either with or without PSO.

5.2 Aims and research questions

The primary aim of the study was to ascertain if PSO had an effect on feeding and growth at 12 months of age. In addition, given suggestions in the literature that PSO may have a more "mechanical" effect on feeding prior to surgical repair of the palate the study also aimed to evaluate if PSO had an effect on feeding just prior to this surgical intervention at 6 months of age.

The primary research questions addressed were:

a. Is there a significant difference in the oral motor skills of infants (with ICP or UCLP) managed with or without PSO post palate repair at 12 months of age?
b. Is there a significant difference in general body growth (weight, length, head circumference and body mass index) of infants (with ICP or UCLP) managed with or without PSO after palate repair at 12 months of age?

In addition, secondary research questions were:

c. Is there a significant difference in the oral motor skills of infants (with ICP or UCLP) managed with or without PSO, prior to surgical repair of the palate at 3 and 6 months of age?

d. Are there any significant differences in reported feeding characteristics (length of feed, nasal regurgitation, number of adaptations of feeding equipment) of infants (with ICP or UCLP) managed with or without PSO, prior to surgical repair of the palate at 3 and 6 months of age?

e. Are there any significant differences in physiological measures of feeding in infants (with ICP or UCLP) managed with or without PSO, prior to surgical repair of the palate at 3 and 6 months of age?

f. Is there any significant difference in the general body growth (weight, length, head circumference and body mass index) of infants (with ICP or UCLP) managed with or without PSO, prior to surgical repair of the palate at 3 and 6 months of age?

Additional data (parent report) was collected at 9 months of age to aid interpretation of any differences that might be found.
5.3 Outcome measures

Primary outcome measures

As the study was primarily concerned with any differences in oral motor skills and anthropometry at 12 months of age, the primary outcome measures at this data collection point were:

- Oral motor skills during feeding (SOMA)
- Anthropometry (weight, length, head circumference and body mass index)

Secondary outcome measures

The secondary outcome measures were studied in order to:

a. address the question of whether PSO had any effect on infant feeding prior to surgical repair of the palate at 6 months of age.

b. provide information contributing to an understanding of the nature and history of any differences found at 12 months of age.

These measures prior to surgical repair were (at birth, 3 and 6 months of age):

- Oral motor skills during feeding
- Physiological measures of bottle feeding/sucking
- Parent's reports about feeding methods, techniques and symptoms
- Assessment of the pharyngeal stage of swallowing using videofluoroscopic assessment of feeding at 3 months of age (in 20 consecutive cases)
5.4 Context of trial

This trial was based at the North Thames Regional Cleft Unit (NTRCU). The unit consists of 2 main centres, St Andrews Centre for Burns and Plastic Surgery (St Andrews), and Great Ormond Street Hospital for Children NHS Trust (GOSH).

5.5 Procedure

5.5.1 Patient selection

- Inclusion and exclusion criteria

Infants referred to GOSH and St Andrews who were diagnosed with unilateral cleft lip and palate or isolated cleft of the soft palate and at least two thirds of the hard palate were considered for inclusion in the study. These groups were selected as representative common cleft types where PSO are used.

Feeding can be affected by many factors, including medical conditions such as cardiac anomalies, prematurity and neurological impairment (Section 2.5). CL and/or P can also be a feature in syndromes where feeding and growth might be affected. Therefore infants who met the above criteria, but required cardiac surgery and/or were diagnosed at birth with neurological impairment and/or a syndrome known to adversely affect feeding and or growth, were excluded.

- Allocation to groups

Once identified as eligible for inclusion (at the routine first contact with a member of NTRCU, within 48 hours of birth), parents or carers were provided with verbal and written information about the study. In cases where the cleft was diagnosed pre-natally parents were informed about the study at
this stage, but were not formally recruited until the infant was born and the diagnosis was confirmed. Information was provided either by the named consultant plastic surgeon, the surgical cleft fellow, the lead consultant orthodontist or one of two clinical nurse specialists. A written information sheet was provided (Appendix 7). Parents or carers had 5 days in which to consider participating in the study. Written consent (Appendix 8) was obtained by any one of the previously listed personnel or the researcher.

Subsequent to obtaining consent, infants were randomised to receive PSO or not. Since it was thought that the effect of PSO might differ for UCLP and ICP, separate randomisation lists were used.

Feeding may be affected by parity and sex (Section 2.5). Minimisation was therefore used to ensure that the two groups contained similar numbers of first born, later born, male and female infants (Treasure and MacRae, 1998). Data for patient allocation was entered by the researcher on a dedicated computer using MINIM (Evans et al., 1990).

All aspects of care for infants recruited to the study, including early counseling, feeding management advice, attendances at clinics and surgery, were carried out according to the NTRCU’s standardised care package.

5.5.2 Prenatal contact

Parents referred after prenatal diagnosis of CL and/or P were seen in the Cleft Lip and Palate Clinic for counseling regarding general management. The parents were usually invited for a follow-up appointment where feeding management was discussed and feeding equipment provided. The type and extent of the cleft was not usually known at this stage and so counseling was kept at a general level.
5.5.3 Early counselling

Counseling was provided within forty-eight hours of the infant’s birth in the maternity unit by one of the Clinical Nurse Specialists and the Surgeon or the Cleft Fellow. This counseling included discussion about the plan of management and surgery involved. Photographs of infants pre and post surgery, and throughout childhood were shown and literature provided.

5.5.4 Feeding advice

The Clinical Nurse Specialist provided feeding advice within forty eight hours of birth. This included education about the nature of feeding problems in infants with CL and/or P including inefficient sucking, extended feed times, fatigue and ingestion of air with need for frequent burping. It is standard practice for the NTRCU to recommend the use of adaptive bottles (Mead Johnson and/or Soft Plas) in conjunction with vented NUK orthodontic teats. An appropriately sized teat was recommended depending on the child’s weight and the size of the cleft. Parents were provided with at least two bottles and teats. In some cases modification of the teat, by enlarging or creating additional holes, was required. Information regarding purchase of this equipment was provided. Further useful strategies to facilitate feeding, such as positioning and frequent burping, were discussed and demonstrated. Although breastfeeding infants with clefts is often unsuccessful (Arvedson, 1998; Glass and Wolf, 1999) this practice was not discouraged. When a mother was keen to trial breastfeeding, advice was provided with particular reference to ensuring the infant was thriving. Given the possible influence of the advice on feeding and growth, a checklist was developed to ensure there was consistency (Appendix 9).

Some infants experienced feeding difficulties that required further advice and management. This was routinely provided by the Clinical Nurse Specialist and on occasion the researcher. The documentation used to record this is given in Appendix 10.
Occasionally, if an infant had more complex feeding problems requiring ongoing specialist intervention, they were referred to the Dysphagia Team Senior Speech and Language Therapist specialising in paediatric dysphagia at GOSH. Any intervention was documented (Appendix 10).

5.5.5 Intra-oral impressions

Intra-oral impressions were taken by one of the named consultant orthodontists, neonatally and again pre-surgery. The impression procedure involved placing a custom-made light cure acyclic impression tray, filled with Optosil (silicon based impression material) (Figure 46) into the infant’s mouth. The loaded impression tray was gently squeezed into place, allowing the impression material to flow into the affected cleft area. This was held in place until the Optosil set, which usually took about one minute. The impression was then removed from the infant’s mouth, disinfected and sent to the dental laboratory, where a dental cast was poured (Figure 47). These casts were used for fabricating the PSO. An additional dental study model was poured and archived for future measurements and research.
Figure 46: Impression tray and Optosil

Figure 47: Dental cast
5.5.6 Surgery

For all infants recruited to the project, the Consultant Plastic Surgeon carried out surgery using a standardised technique. For UCLP patients lip repair was performed as close to 12 weeks as possible. During this surgery vomerine flaps were raised. Palate repair was performed as close to 6 months as possible and was standardised with a "no flap" repair wherever possible. If necessary, von Langenbeck flaps were used. Radical velar muscle dissection (intravelar veloplasty) was undertaken in the repair of the soft palate (Sommerlad, 2000). The timing of repair and any variation in technique were documented.

5.5.7 Cleft Lip and Palate Clinic appointments (CLPC)

All infants attended the CLPC for a standardised number of appointments in the first year of life. These appointments were adapted to the different conditions of ICP and UCLP, and are summarised in Figures 48 and 49.

It was decided to co-ordinate research assessment/data collection points with CLPC appointments. These are summarised on Figures 48 and 49.
Figure 48: Timetable for CLPC appointments for infants with UCLP, in relation to surgery
Figure 49: Timetable for CLPC appointments for infants with ICP, in relation to surgery
5.5.8 Multidisciplinary team

Many staff were involved in the care of the infants. GOSH clinics were all undertaken at that hospital. St Andrews staff were based at Broomfield Hospital but clinics were undertaken at several sites including the Royal London Hospital NHS Trust, Colchester General Hospital, Southend Hospital and Basildon Hospital.

5.5.9 Orthodontic appointments

All infants were seen by one of the five named orthodontists for an initial intra-oral impression within two weeks of birth either as an inpatient in the maternity unit or as an outpatient.

All infants with UCLP had intra-oral impressions (Section 5.5.5) taken within the week prior to lip repair and again just prior to palate repair. Infants with ICP had impressions taken prior to palate repair at 6 months of age. For patients recruited at the GOSH site, repeat impressions were carried out in the Maxillofacial Dental Department. Where there were concerns about the infant's airway status (e.g. when ICP was associated with Pierre Robin Sequence), a Respiratory Physician was present. At St Andrews the orthodontists undertook repeat impressions in theatre without anaesthetic.

If the infant was randomised to the PSO group, the fitting of the plate took place before 2 weeks of age. The plate was adjusted as necessary at the routine appointments. Infants in the UCLP group were fitted with a new plate during their admission.

Regardless of which group they were randomised to, all UCLP infants were seen by the orthodontist 5-6 times within the first 3 months and a further 3 times before palate repair. All infants with ICP were seen 4 times within the first 6 months. The review appointments involved an interview about how the parents were managing the appliance, whether they were concerned about any aspects of the appliance (for example rubbing) and how feeding was
progressing. In addition the infant's mouth was examined for any signs of rubbing, ulceration, infection or neonatal teeth erupting, which might alter fitting of the appliance. Guidelines and documentation for review orthodontic appointments were developed (Appendix 11).

5.6 Assessment tools

Given the developmental changes that occur during the first year of life, the tools used to measure the outcomes varied according to age. The following section describes the tools used.

5.6.1 Anthropometry

Anthropometric data are widely used to monitor growth and provide information about nutritional status in infants and children (Gibson, 1990). Weight, length and head circumference data were selected as anthropometric measures for this study, because they could be made easily, quickly and accurately. Body mass index scores were calculated using weight and length measurements. The data was collected by the researcher, using a standardised technique and following training by a paediatrician.

Seca Baby Scales were used for weight measurements, with the infants unclothed, lying or sitting.

Recumbent length measurements were taken using Rollemetre and according to the technique described by Gibson and Lohman et al (1990). The infant was placed face upward, with his/her head towards the fixed end (head board) of the Roller Metre and the body along the mat as shown in Figure 50. The infant's head was gently held by a parent at the point at which contact was made with the headboard. The researcher, whilst holding the infant's feet (with toes pointing upwards), keeping the knees straight and ensuring the infant's shoulder blades were in contact with the mat, brought the movable
footboard to rest against the infant’s heels and noted the measurement shown to the nearest millimetre.

Head circumference was measured using a tape measure. The infants were held in a sitting position by a parent or carer with the infants looking straight ahead and the head in a horizontal position as shown in Figure 51. The researcher placed the tape measure just above the supra-orbital ridges, covering the most prominent part of the frontal bulge and over the part of the occiput, which gave the maximum circumference (Weiner and Lourie, 1969). Care was taken to ensure that the tape measure was level on both
sides of the head and that the tape measure was pulled tightly to compress the hair. Measurements were taken to the nearest millimetre.

Figure 51: Diagrammatic representation of method of measuring head circumference (Gibson, 1990)

All growth measurements were converted to z scores using the SDSGAIN macro (Freeman et al., 1995; Cole et al., 1995; Cole et al., 1998; Child Growth Foundation, 1999; Wright et al., 2002) allowing comparison of infants without concerns about exact ages at the data collection points. (World Health Organisation, 1983; World Health Organisation, 1986; Gibson, 1990; Wright et al., 2002).

5.6.2 Oral motor skills

Neonatal oral motor skills (at neonatal and 3 months of age assessments) were measured using the “Neonatal Oral-Motor Assessment Scale” or NOMAS developed by Braun and Meyer Palmer (1985, 1990). The revised 1990 version of the NOMAS was used (Appendix 12). According to the
authors' instructions the assessment was rated on the first two minutes of feeding taken from the video obtained during the GOSMIF assessment, and consequently was based on infants taking glucose solution rather than milk. In addition a second rating was made of the infants taking a routine milk feed with their own adaptive feeding equipment.

Oral motor skills during infancy (at 6 and 12 months of age) were measured using the “Schedule for Oral Motor Assessment” or SOMA developed by Skuse et al, 1995 (Appendix 13). Infants were given a variety of developmentally appropriate food textures in the prescribed manner. At 6 months of age, the infants were assessed with two textures; liquid from a bottle and/or beaker, and puree. At 12 months of age, up to six textures were assessed including liquid from a bottle and/or training beaker, puree, semi-solid, cracker and biscuit. If infants had not previously been offered any of these textures, these were omitted.

5.6.3 Physiological measures of bottle feeding

The physiological measures of bottle-feeding were obtained using the “Great Ormond Street Measurement of Infant Feeding” (GOSMIF). Assessment with the GOSMIF was carried out according to the protocol described in Chapter 4.

5.6.4 Feeding questionnaires

Structured questionnaires were designed to collect information about aspects of feeding such as methods used, routines, use of adaptive equipment, reported duration of feeds and clinical symptoms. These questionnaires were administered at all data collection points. Although they are similar in their design they were modified to reflect developmental changes (Appendices 18, 21, 23, 24 and 25).
5.6.5 Videofluoroscopy

Initially it was planned to use this assessment only in cases where there were clinical symptoms suggestive of a possible pharyngeal component to the infant's feeding difficulties. The majority of infants were reported and observed to show coughing/choking or gurgly breath sounds (clinical suggestions of a pharyngeal component to swallowing) during assessment feeds. One such infant was reported to demonstrate these clinical signs on a regular basis causing the mother concern and therefore underwent videofluoroscopy. The extent of pharyngeal involvement in this infant contradicted reports in the literature that infants with CL and/or P show only oral stage difficulties. In order to ensure that this was not a phenomenon specific to this infant a decision was made to assess a consecutive series of infants. Since videofluoroscopy is an invasive procedure and exposes children to radiation it was decided to assess a limited number of infants. The aim of carrying out this procedure was firstly to confirm the presence/absence of pharyngeal stage difficulties, and secondly to evaluate the pharyngeal stage of the swallow. This would identify any major differences in components of the swallow, specifically where the swallow was triggered and amounts of pharyngeal residue, in infants fitted with PSO and those without. As a result, the Ethics Committee of GOSH and the Institute of Child Health, gave permission to carry out videofluoroscopy on 20 consecutive infants at GOSH (whether PSO or no PSO). This assessment was carried out at 3 months of age, prior to any surgical intervention ensuring that surgery had not influenced the feeding patterns. Additionally feeding remains essentially reflexive at this developmental stage (as discussed in Chapter 2) and infants were co-operative for the assessment. In order to minimise radiation levels and ensure that the infants fed in a pattern as similar as possible to normal and reflecting their “best” feeding, the protocol for the videofluoroscopic assessment was modified slightly from that suggested by Arvedson and others in terms of the positioning of the infant and the feeding equipment used (Ardran et al., 1958a; Ardran et al., 1958b; Ekberg et al., 1988; Kramer, 1989;
Griggs et al., 1989; Dodds et al., 1990; Zerrili et al., 1990; Newman et al., 1991; Geyer and McGowan, 1995; Arvedson and Lefton-Greif, 1998; Dick, 1998; O'Donoghue and Bagnall, 1999). The infants were positioned in a Tumbleform seat, in an upright position as similar to their usual position as possible. They were fed liquid barium by a parent or carer, from their routine feeding bottle and teat. Ten consecutive swallows were recorded and analysed using a specifically designed rating scale adapted from Arvedson and Lefton-Greif (1998) (Appendix 14).

5.7 Potential confounders/additional information

In addition to the primary and secondary outcome measures, background information that might have an effect on feeding was collected including family illness and infant medical or developmental problems.

5.7.1 Structured interviewer led questionnaires

Questionnaires were designed to allow collection of information about factors that have been reported to affect feeding.

The first of these structured questionnaires (administered at the neonatal assessment) included collection of demographic data. Information was also collected about medical and social risk factors for developmental delay and feeding problems (Section 2.5) (Appendix 17).

Follow-up information regarding these aspects was collected at all subsequent assessments. This included details of medical problems and interventions, and social information such as family structure and employment (Appendix 20).

5.7.2 Denver II

Given the implications of developmental delay on feeding, oral motor skills and growth, a developmental screen was included. The Denver II is a general developmental screening test that evaluates personal-social ability, fine motor-
adaptive skills, language and gross motor skills in pre-school children (Appendix 15). It identifies children whose development is delayed in comparison with other children of the same age. It can be repeated as required. The Denver II was initially standardised on a cross section of the American population but has been further standardised in other countries including the UK (Wade, 1992; Sperhac and Salzer, 1991; Franken burg, 1969; Bryant et al., 1974; Franken burg, 1988).

Age appropriate items were assessed as prescribed in the administration manual (Frankenburg et al., 1990a). These test items were administered by the researcher either directly evaluating the child's performance or by parent report. Individual items were scored as advanced, normal, caution, delayed or no opportunity. An overall interpretation of normal, suspect or untestable was then made based on the number of normal, caution, delayed scores for individual items (according to the detailed instructions in the Denver II manual).

All infants identified as having suspect scores were referred to the team's paediatrician for further assessment.

5.8 Data collection routine

All infants underwent feeding assessment on 4 occasions during the first year of life. These assessments occurred neonatally (before 3 weeks of age), at 3, 6 and 12 months of age, and were carried out in the infant's home or at a routine clinic appointment. All assessments took place at an anticipated/routine feed time and took approximately 90 minutes to complete. An additional interview was carried out when the infant was approximately 9 months of age. It was carried out as a telephone interview or was co-ordinated with a routine Cleft Lip and Palate Clinic appointment. All assessments were administered by the researcher.
Figure 52: Timetable of data collection points and assessment tools used in relation to surgical management.

* The different colours on this table correspond to the different assessment tools discussed below and included in the appendices.
5.8.1 Neonatal assessment (Data collection point 1)

If the infant was allocated to the PSO group, this assessment took place more than 24 hours after fitting of the PSO.

Reinforcement and/or modifications of the feeding advice previously given, also took place at this stage. Revision or clarification of the use of adaptive equipment was given. If it was felt that feeding efficiency or safety might be improved, adjustments to the mother's feeding technique or equipment were made. To ensure consistency, advice given was documented. The forms used to record this are shown in Appendix 16.

There were 3 components to this assessment.

Feeding assessment. The infant's feeding was assessed using the GOSMIF according to the protocol (Section 4.5). This assessment was also recorded on videotape. Following the GOSMIF assessment the infant was fed a routine feed by a parent or carer, which was also video recorded.

A standardised history was taken (Appendix 17 and 18).

Anthropometric data was collected, as described in 5.6.1 (Appendix 19).

The order in which the first two components were carried out varied as appropriate for each infant. For example, if the infant was sleeping when the researcher arrived the feeding history was carried out prior to the feeding assessment. However anthropometric data was always collected after the feeding assessment.

5.8.2 3 month assessment (Data collection point 2)

The second assessment was carried out at 12 weeks of age (range 10-14 weeks) and prior to the lip repair of infants with UCLP.
There were 4 components to this assessment.

*Feeding assessment.* The infant's feeding was assessed using the GOSMIF according to the protocol (Section 4.5). This assessment was also recorded on videotape. Following the GOSMIF assessment the infant was fed a routine feed by a parent or carer, which was also video recorded.

*A standardised history update* was taken (Appendices 20 and 21).

*Anthropometric data* was collected, as described in 5.6.1 (Appendix 22).

A *Developmental screen* using the Denver II was administered (Section 5.7.2) (Appendix 15).

The order in which the first three components were carried out varied as appropriate for each infant. However anthropometric data was always collected after the feeding assessment.

**5.8.3 6 month assessment (Data collection point 3)**

The third assessment was carried out at approximately 6 months of age (range 22-26 weeks) and prior to palate repair.

There were 4 components to this assessment.

*Feeding assessment.* The infant's feeding was assessed using the GOSMIF according to the protocol (Section 4). This assessment was also recorded on videotape. Following the GOSMIF assessment the infant was fed a routine feed by a parent or carer. Solids (smooth puree) and milk feeds were given. This was also video recorded.

*A standardised history update* was taken (Appendices 20 and 23).

*Anthropometric data* was collected, as described in (Section 5.6.1) (Appendix 22).
A Developmental screen using the Denver II was administered (Section 5.7.2) (Appendix 15).

The order in which the first, second and fourth components were carried out varied as appropriate for each infant. However anthropometric data was always collected after the feeding assessment.

5.8.4 9 month assessment (Data collection point 4)

Assessment 4 was carried out when the infant was 9 months of age (+/- 2 weeks) and consisted of a parental interview (Appendices 20 and 24). It was carried out either as a telephone interview or at a routine cleft lip and palate clinic appointment.

5.8.5 12 month assessment (Data collection point 5)

The final assessment was carried out at 12 months of age (+/- 2 weeks.)

There were 4 components to the assessment.

Feeding assessment. The infant was fed a variety of food textures by a parent or carer as specified in the SOMA (Section 5.6.2). In some cases the infant self fed.

A standardised history update was taken (Appendices 20 and 25).

Anthropometric data was collected (Section 5.6.1) (Appendix 22)

A developmental screen was administered using the Denver II (Section 5.7.2) (Appendix 15).

The order in which the components were carried out varied as appropriate for each infant. However anthropometric data was always collected after the feeding assessment.
5.9 Blinding

The researcher carried out all interviews during the trial. As she was fully aware of whether the infants had been randomised to be managed with or without PSO this component of the assessment was not blinded.

Both oral motor skills assessments were administered by the researcher according to prescribed protocols. In order to ensure there was no bias in the rating of these assessments, they were video-recorded and rated by specialist dysphagia trained speech and language therapists who were not involved in the study and were therefore blinded as to whether the infants had been randomised to the No PSO or PSO groups.

All NOMAS studies were rated by an experienced dysphagia trained speech and language therapist, who had attended a NOMAS training course (during the first year of the trial) and achieved 90 percent inter-rater reliability on reliability assessment. She was blinded as to whether infants had been randomised to have PSO or no PSO.

All the SOMA studies were rated by a different experienced dysphagia trained speech and language therapist, who was trained to rate SOMA studies. She was blinded as to whether infants had been randomised to have PSO or no PSO.

The videofluoroscopy assessments were independently rated by the researcher and the paediatric feeding advisor to the study. PSO are clearly visible on videofluoroscopy and it was therefore not possible to rate this assessment blindly.

The GOSMIF assessments were administered by the researcher who was not blinded to whether the infants were randomised to No PSO or PSO.
The Denver II assessments were administered and rated by the researcher following completion of the recommended training (Frankenburg et al., 1990b). Inter-rater reliability of the Denver II has been reported to be excellent, with 99.7% agreement and a Kappa coefficient of >0.75 (Sperhac and Salzer, 1991; Wade, 1992; Frankenburg et al., 1990a; Frankenburg et al., 1990b). As the assessments were carried out by the researcher, blinding was not possible. However if infants were referred to the paediatrician for further assessment he was blinded to whether the infant had been managed with or without PSO. Given the reported reliability of the Denver II, the time required from other staff to carry out reliability checks and the impracticality of repeating assessments no reliability checks were carried out. It is acknowledged that this may have allowed room for rater bias.

Similarly anthropometric data was collected by the researcher, who knew whether the infants had been randomised to be managed with or without PSO, and was therefore not blinded.

5.10 Statistical analysis

PSO and no PSO groups were compared for UCLP and ICP with respect to the potentially confounding background factors (general health questionnaire and Denver II) at each time point. Differences were presented. The extent to which a background factor may confound any results was not determined solely on the basis of significance (Altman, 1985).

Comparisons were initially made between PSO and no PSO groups for UCLP and ICP separately at each time point. Throughout, t-tests were used for comparison of continuous numeric outcomes where normality could be assumed. For continuous outcomes that did not appear to be normally distributed, or where numbers were too small to establish normality, comparisons were made using Mann-Whitney U-tests. Categoric outcomes were compared using exact tests with StatXact v4.0.1. All results were
presented with 95% confidence intervals for the differences in mean, median or percentages as appropriate.

Primary comparisons were made between SOMA and anthropometry z-scores at 12 months of age. The secondary outcomes were investigated via comparison at each data collection point and then by consideration of trends over time for the continuous outcomes that were serially measured (anthropometry z-scores, GOSMIF measures, length of feeds). The serial measurements were charted over time and the trajectories compared. All data were adjusted for confounding factors prior to comparison as necessary. The percent agreement between NOMAS results during the GOSMIF and the routine feed were calculated, and Kappa coefficients used to quantify agreement corrected for chance.

5.11 Sample size

Figure 53(a) shows the sample sizes required to detect shifts of between 0.5 and 2.5 standard deviations in the continuous numeric outcomes with 80, 90 and 95% power at the 5% significance level. A shift of 0.8 SD in any of the anthropometry z-scores was felt to be clinically important and this could be detected with 80% power (5% significance) using 2 groups of 25 infants. It was therefore planned to allocate 50 infants with ICP and 50 infants with UCLP to either No PSO or PSO (25 UCLP No PSO, 25 UCLP PSO, 25 ICP No PSO, 25 ICP PSO).

Figure 53(b) shows the percentages in the reduction of infants rated abnormal using SOMA at 12 months of age when using PSO that could be detected with 80, 90 or 95% power at the 5% significance level if 50 infants are evenly randomised to 2 groups. There is currently no information on the percentage with abnormalities expected in the control (no PSO) group. Figure 53 shows that if an abnormality rate of 70% falls by 28% to 42%, it will be detected
with 80% power, a fall of 37% (to 33%) would be detected with 90% power and a fall of 46% (to 24%) would be detected with 95% power.

Figure 53(a): Graph showing the standardised difference in means that can be detected with 80, 90 and 95% power at 5% significance with specified numbers per group.
Control percentage abnormal

Figure 53(b): Graph showing percentage reduction detected with 80, 90, 95% power at 5% significance level for different control percentages and two groups of 25 infants.

5.12 Availability of patients

The initial applications for funding for this study were submitted in 1998. The average annual referral rate of infants with non-syndromic CL &/or P for the 2 years prior to applications for research funding (1996 and 1997) was 36 at GOSH and 28 at St Andrews. The proportion of infants with ICP was 66% at GOSH and 60% at St Andrews. Therefore it was expected that there would be approximately 40 infants born with ICP over a 12-month period. However the classification of ICP included all isolated clefts including smaller
clefts such as those of the soft palate only and it was therefore expected that there would be fewer infants than 40 meeting the trials inclusion criteria. It was expected that 24 infants with UCLP would be recruited over a 12-month period. With recruitment due to start in July/August 1999, it was therefore anticipated that inadequate numbers of infants would be recruited over the 2-year recruitment period. However, in recent years in the UK, there had been a major evaluation of the process and outcomes of services to infants, children and adults with CL and/or P, with the consequence that reorganisation of services was recommended (CSAG Report, 1998; Bearn et al., 2001; Sandy et al., 2001; Sell et al., 2001; Williams et al., 2001). This re-organisation was scheduled for implementation in April 2000 and hence it was envisaged that the referral rate would increase significantly at this time, boosting recruitment to well over the 100 required for our study. However, implementation of the CSAG recommendations was significantly delayed and recruitment fell behind schedule. An application for additional funding to enable extension of the recruitment period was made in March 2001. At this time 33 infants had been enrolled, 13 ICP and 20 UCLP. Based on the recruitment pattern during the trial and on the assumption that the CSAG recommended reorganisation would now occur in October 2002, it was anticipated that the additional year requested would allow analysis of feeding data of 44 infants at 12 months of age, 62 infants at 6 months of age and 70 infants at 3 months of age.
Chapter 6

RESULTS

This chapter presents the results of the trial. Baseline characteristics are presented (Sections 6.4 and 6.5) and any subsequent changes in the demographic and medical status of the infants that occur over the data collection period are reported (Section 6.6). Adherence to the standardised care protocol (nursing advice and orthodontic appointments) is described (Section 6.8). Protocol variation in the timing of data collection points are presented and reasons for this are given (Section 6.3). Comparisons of the primary and secondary outcome measures for each cleft type with and without PSO, as detailed in Chapter 5 are reported (Sections 6.10). Compliance with the treatment (PSO) is reported (Section 6.9) and finally per protocol analyses for primary outcome measures are reported (Section 6.11).

6.1 Recruitment

It was anticipated that planned CSAG re-organisation, initially proposed to start April 2000, would result in an increase in the numbers of infants referred to the NTRCU. However, implementation of the new service model did not take place until April 2002 well into the data collection period, which commenced August 1999. Following this, the numbers of infants referred to the NTRCU did increase but unfortunately many infants did not meet the inclusion criteria (cleft type and extent) for the trial and the pattern of recruitment did not change as anticipated.

The approximate numbers of infants referred each year (over the data collection period) to GOSH was 70 to 75, and to St Andrews approximately 50. However many of the infants referred to GOSH had syndromic clefts and
were medically unwell therefore not meeting the inclusion criteria. Of the 34 recruited infants with UCLP 13 were recruited from GOSH and 21 from St Andrews. The 16 infants recruited with isolated cleft palate were distributed evenly across the sites (Table 6).

The majority of infants meeting the inclusion criteria were recruited. However, there were eight infants whose parents did not consent to participate in the trial. Parents of four infants refused to participate due to the increased number of appointments together with concerns about managing the PSO if they were randomised to that group. All of these infants were under the care of GOSH and were therefore managed without PSO. Parents of another two infants had chosen to attend GOSH for management of their infants but lived well outside the immediate area of either St Andrews or GOSH, making travel for orthodontic appointments impractical and they therefore declined to join the trial. Parents of the further two infants refused as they wished their infants to be fitted with plates. One of these infants was managed at St Andrews and therefore received PSO. The other was from GOSH and while they would not routinely have received PSO, they did so at the parents request. There were two infants whom the consultant plastic surgeon judged inappropriate for recruitment. In the first case there were significant social problems. In the second both parents had drug addictions and the infant was born with an addiction requiring weaning with morphine.

In total 50 infants with CL and/or P were recruited, 34 with complete unilateral cleft lip and palate (UCLP) and 16 with isolated clefts of the soft and at least 2/3rds of the hard palate (ICP). One infant (UCLP) was withdrawn from the trial as a result of complex medical problems before intervention was begun.
6.2 Numbers of patients assessed and analysed at different data collection points

It was initially planned to complete recruitment twelve months prior to the end of the trial, allowing follow-up of all infants at 12 months of age. However given the recruitment difficulties, the recruitment period was extended resulting in some infants not having been followed to 12 months at the time of analysis. Forty-nine infants underwent the neonatal assessment. Forty-eight infants had reached 3 months of age during the data collection period, however one infant was not assessed as the mother failed to attend. All infants who reached 6 months of age (n=43) and 12 months of age (n=34) during the data collection period were assessed (Table 4).

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO</td>
<td>PSO</td>
</tr>
<tr>
<td>Neonatal assessment</td>
<td>16</td>
<td>17</td>
</tr>
<tr>
<td>3 month assessment</td>
<td>15</td>
<td>17</td>
</tr>
<tr>
<td>6 month assessment</td>
<td>14</td>
<td>15</td>
</tr>
<tr>
<td>12 month assessment</td>
<td>8</td>
<td>13</td>
</tr>
</tbody>
</table>

*Table 4: Number of infants assessed at each data collection point according to cleft type and PSO status*

6.3 Adherence to protocol assessment times

All neonatal assessments were within the time frame specified in the protocol (i.e. under 1 month of age) as were the majority of assessments at 3 and 6 months of age (Table 5). The exceptions were:
1. One infant with (UCLP No PSO) had early lip and palate repair, allowing the family to travel to Bangladesh for a family wedding (3 month assessment at 2.4 months of age and 6 month assessment at 4.4 months of age).

2. One infant (ICP PSO) was hospitalised for airway management. The 3 month assessment therefore occurred late (4.8 months of age).

3. One infant whose palate repair and hence 6 month data collection was delayed because of abnormal blood results (UCLP No PSO) (7.56 months of age).

4. One infant (UCLP PSO) who was assessed early at the 6 month assessment point (4.92 months of age) allowing the researcher to take leave.

5. One infant (ICP No PSO) was seen slightly early at the 6 months assessment (5.28 months of age) to coincide with surgery.

By the time the infants were 12 months of age, it was more difficult to schedule assessments within the specified period due to commitments such as nursery placements, mothers returning to work and family holidays. While all assessments for the ICP groups were “on time”, several of the UCLP group assessments were up to a month “late”. This was similar for both the UCLP No PSO and UCLP PSO groups.
<table>
<thead>
<tr>
<th>Assessment</th>
<th>Cleft Type</th>
<th></th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td>UCLP</td>
<td>ICP</td>
<td>No PSO</td>
<td>PSO</td>
</tr>
<tr>
<td></td>
<td></td>
<td>No PSO</td>
<td>PSO</td>
<td>No PSO</td>
<td>PSO</td>
</tr>
<tr>
<td>Neonatal assessment</td>
<td>mean</td>
<td>0.49</td>
<td>0.56</td>
<td>0.51</td>
<td>0.57</td>
</tr>
<tr>
<td>(under 1 month of age)</td>
<td>range</td>
<td>.24 - .84</td>
<td>.12 - 1</td>
<td>.12 - .96</td>
<td>.24 - .96</td>
</tr>
<tr>
<td></td>
<td>SD</td>
<td>0.21</td>
<td>0.24</td>
<td>0.29</td>
<td>0.27</td>
</tr>
<tr>
<td>3 month assessment</td>
<td>mean</td>
<td>2.85</td>
<td>2.81</td>
<td>3.31</td>
<td>3.43</td>
</tr>
<tr>
<td>(2.5 to 3.5 months of age)</td>
<td>range</td>
<td>2.4 - 3.72</td>
<td>2.46 - 3.48</td>
<td>3.12-3.48</td>
<td>2.52 - 4.80</td>
</tr>
<tr>
<td></td>
<td>SD</td>
<td>0.31</td>
<td>0.27</td>
<td>0.26</td>
<td>0.75</td>
</tr>
<tr>
<td>6 month assessment</td>
<td>mean</td>
<td>5.96</td>
<td>5.82</td>
<td>5.64</td>
<td>5.88</td>
</tr>
<tr>
<td>(5.5 to 6.5 months of age)</td>
<td>range</td>
<td>4.44 - 7.56</td>
<td>4.92 - 6.24</td>
<td>5.28 - 6</td>
<td>5.76 - 6</td>
</tr>
<tr>
<td></td>
<td>SD</td>
<td>0.66</td>
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<td>12 month assessment</td>
<td>mean</td>
<td>12.81</td>
<td>12.43</td>
<td>12.32</td>
<td>12.43</td>
</tr>
<tr>
<td>(12 to 13 months)</td>
<td>range</td>
<td>11.88 - 14.04</td>
<td>11.64 - 13.92</td>
<td>12 - 12.72</td>
<td>12 - 12.96</td>
</tr>
<tr>
<td></td>
<td>SD</td>
<td>0.71</td>
<td>0.62</td>
<td>0.28</td>
<td>0.38</td>
</tr>
</tbody>
</table>

*Table 5: Table of ages of infants (months) at data collection points*

The following figures (Figure 54a and 54b), based on the template recommended by the CONSORT group (Altman et al., 2001), summarises the flow of infants through each stage of the trial, from recruitment to analysis.
Unilateral Cleft Lip and Palate
Assessed for eligibility (n=40)

Excluded (n=6)
* Did not meet inclusion criteria (n=0)
* Refused to participate (n=5)
* Other reasons (n=1)

Randomised

Allocated to control group; No PSO (n=16)
* Received allocated intervention (n=16)
* Did not receive allocated intervention (n=0)

Allocated to intervention group; PSO (n=18)
* Received allocated intervention (n=17)
* Did not receive allocated intervention (n=1); infant was withdrawn with complex medical problems

Lost to follow-up (n=0)

Lost to follow-up (n=0)

Analysed (n=16)
(detailed in section 6.2)
Excluded from analysis (n=0)

Analysed (n=17)
(detailed in section 6.2)
Excluded from analysis (n=0)

Figure 54a: Diagram showing the flow of participants with UCLP through each stage of the trial; based on template recommended by Altman et al (2001)
Assessed for eligibility (n=20)

Excluded (n=4)
* Did not meet inclusion criteria (n=0)
* Refused to participate (n=3)
* Other reasons (n=1)

Randomised

Allocated to control group; No PSO (n=8)
  * Received allocated intervention (n=8)
  * Did not receive allocated intervention (n=0)

Allocated to intervention group; PSO (n=8)
  * Received allocated intervention (n=8)
  * Did not receive allocated intervention (n=0)

Lost to follow-up (n=0)

Analysed (n=8)
(detailed in section 6.2)
Excluded from analysis (n=0)

Analysed (n=8)
(detailed in section 6.2)
Excluded from analysis (n=0)

Figure 54b: Diagram showing the flow of participants with ICP through each stage of the trial; based on template recommended by Altman et al (2001)
6.4 Baseline data

Approximately 2/3rds of the infants recruited to the study were male (UCLP 21, 63.41% and ICP 9, 61.88%).

Since minimisation was used to allocate infants, the No PSO and PSO groups were balanced for sex and birth order. Mean and median gestational age for the groups were also similar (UCLP: p=0.70 with 95% ci for the difference in means –0.29 to 0.38; ICP: p=0.07 with ci –2.31 to 0.88 ). Birth weights were on average slightly higher amongst the groups allocated to PSO (average 50 grams higher for UCLP and 10 grams higher for ICP) (UCLP; p= 0.77 with ci –290 to 380 ; ICP p= 0.98 with ci -570 to 560). Representation from ethnic groups was balanced in the ICP groups but there was a slightly higher proportion of infants of Asian descent (including Indian, Bangladeshi and “other” Asian) in the UCLP PSO group (p=0.20). Father’s occupational classification (as an indication of social class) was similar in UCLP No PSO and PSO groups. There was however a slightly higher but not significant (p=0.36) proportion of professional occupations in the ICP PSO group as compared to the ICP No PSO group.

These results are summarised in Table 6.
<table>
<thead>
<tr>
<th></th>
<th>Cleft Type</th>
<th></th>
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<tbody>
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<td>ICP (n=16)</td>
<td></td>
<td></td>
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<tr>
<td></td>
<td>No PSO</td>
<td>PSO</td>
<td>No PSO</td>
<td>PSO</td>
</tr>
<tr>
<td></td>
<td>n=16</td>
<td>n=17</td>
<td>n=8</td>
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<tr>
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<td></td>
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<td>Male</td>
<td>10</td>
<td>11</td>
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<td>5</td>
</tr>
<tr>
<td>Female</td>
<td>6</td>
<td>6</td>
<td>4</td>
<td>3</td>
</tr>
<tr>
<td>Referral site</td>
<td></td>
<td></td>
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<tr>
<td>GOSH</td>
<td>7</td>
<td>6</td>
<td>4</td>
<td>4</td>
</tr>
<tr>
<td>St Andrews</td>
<td>9</td>
<td>11</td>
<td>4</td>
<td>4</td>
</tr>
<tr>
<td>Gestational age (weeks)</td>
<td></td>
<td></td>
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<td></td>
</tr>
<tr>
<td>Mean</td>
<td>39.61</td>
<td>39.75</td>
<td>40.29</td>
<td>39.21</td>
</tr>
<tr>
<td>Std Deviation</td>
<td>1.31</td>
<td>1.38</td>
<td>1.16</td>
<td>1.1</td>
</tr>
<tr>
<td>Minimum</td>
<td>37.2</td>
<td>36</td>
<td>38</td>
<td>38</td>
</tr>
<tr>
<td>Maximum</td>
<td>42</td>
<td>42</td>
<td>42</td>
<td>40.71</td>
</tr>
<tr>
<td>Birthweight (kg)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mean</td>
<td>3.36</td>
<td>3.41</td>
<td>3.47</td>
<td>3.46</td>
</tr>
<tr>
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<td>0.5</td>
<td>0.66</td>
<td>0.37</td>
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<td>Minimum</td>
<td>2.84</td>
<td>2.33</td>
<td>2.43</td>
<td>2.96</td>
</tr>
<tr>
<td>Maximum</td>
<td>4.1</td>
<td>4.03</td>
<td>4.2</td>
<td>3.85</td>
</tr>
<tr>
<td>Birth order</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mean</td>
<td>2</td>
<td>1</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Mode</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Std Deviation</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Minimum</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Maximum</td>
<td>5</td>
<td>4</td>
<td>3</td>
<td>3</td>
</tr>
</tbody>
</table>

Table 6: Summary of infants recruited, distribution across groups; sex, referral site, gestational ages, birth-weights and birth order
<table>
<thead>
<tr>
<th>Ethnic origin</th>
<th>Indian</th>
<th>Bangladeshi</th>
<th>Other Asian</th>
<th>White/UK</th>
<th>White/European</th>
<th>Turkish</th>
<th>Black African</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>11</td>
<td>3</td>
<td>1</td>
<td>0</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Fathers' Occupational classification</th>
<th>1 (Managers and Senior Officials)</th>
<th>2 (Professional Occupations)</th>
<th>3 (Associate Professional and Technical Occupations)</th>
<th>5 (Skilled Trade Occupations)</th>
<th>8 (Process, Plant and Machine Operatives)</th>
<th>9 (Elementary Occupations)</th>
<th>unemployed or student</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>0</td>
<td>5</td>
<td>2</td>
<td>2</td>
<td>2</td>
<td>1</td>
<td>3</td>
</tr>
</tbody>
</table>

Table 6 continued: Summary of demographics of infants recruited; distribution across groups, ethnicity and occupational classification

6.5 Baseline risk factors for developmental delay and feeding difficulties

In order to ensure the infants within the No PSO and PSO groups were comparable, baseline risk factors for developmental delay and feeding difficulties were recorded.
6.5.1 Prenatal factors

*Mothers' health during the pregnancy*

Thirty-five of the 49 mothers reported being completely well or only slightly unwell with expected pregnancy difficulties such as nausea. Fourteen women reported being unwell with more complex pregnancy related problems, of whom 7 required specialist intervention and/or hospitalisation. Medical problems included bleeding (n= 8), infections (n=6), thyroid disease (n=1) and polyhydramnios (n=1). These problems were more common in the No PSO groups where mothers were more likely to report bleeding and infections during the pregnancy, but this difference was not statistically significant (Table 7).

*Other factors including medications, alcohol consumption, smoking and medications*

Only 5 mothers drank alcohol during their pregnancy; 3 drank occasionally (less than 3 units a week) and another 2 drank more than 14 units a week (Table 7).

Five mothers smoked during pregnancy. One mother reported that she smoked 15 cigarettes per week and the other 4 reported smoking 70 or 75 cigarettes per week (Table 7).

Thirteen mothers took some form of medication during their pregnancy. In all cases the medications were iron supplements and/or antibiotics.

There were no significant differences between the groups for any of these factors (Table 7).
6.5.2 Perinatal factors

Pain relief

Forty five mothers had pain relief during delivery. There was no difference in the distribution across groups (Table 7). The majority had gas and air (n=16) or epidural (n=16). Nine mothers had pethadine. Two mothers had general anaesthetics and 2 had spinal blocks (all associated with caesarean sections).

Delivery complications

Thirty mothers had uncomplicated deliveries requiring no assistance. In 9 cases assistance (vontous or forceps) were used. Ten mothers had emergency caesarean sections (Table 7).

Percentages within groups were similar and there were no significant differences between the groups for any of these factors although confidence intervals were wide (Table 7).

Immediate post natal period

Immediate postnatal foetal distress was reported in 11 cases, however all infants had APGAR scores of 7 or above at 5 and 10 minutes. Twenty-one infants were admitted to special baby care units or neonatal units. In all cases this was to manage feeding difficulties rather than medical problems.
<table>
<thead>
<tr>
<th>Mothers' health</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td>generally well</td>
<td>6</td>
<td>5</td>
</tr>
<tr>
<td>unwell with expected pregnancy</td>
<td>3</td>
<td>2</td>
</tr>
<tr>
<td>difficulties</td>
<td>2</td>
<td>1</td>
</tr>
<tr>
<td>unwell with more than expected</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>pregnancy difficulties</td>
<td></td>
<td></td>
</tr>
<tr>
<td>specialist management required</td>
<td>4</td>
<td>0</td>
</tr>
<tr>
<td>hospitalisation required</td>
<td></td>
<td>1</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Mothers' medical problems</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td>bleeding during pregnancy?</td>
<td>4</td>
<td>3</td>
</tr>
<tr>
<td>infections during pregnancy?</td>
<td>3</td>
<td>1</td>
</tr>
<tr>
<td>thyroid disease?</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>polyhydrammos?</td>
<td>1</td>
<td>0</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Other factors</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td>medications during pregnancy?</td>
<td>6</td>
<td>0</td>
</tr>
<tr>
<td>alcohol / week during pregnancy?</td>
<td></td>
<td>3</td>
</tr>
<tr>
<td>occasional (3 units or less per week)</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>14 units per week</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>cigarettes per week</td>
<td>15</td>
<td>0</td>
</tr>
<tr>
<td>70-75</td>
<td>2</td>
<td>1</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Delivery</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td>pain relief?</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>nothing</td>
<td></td>
<td>2</td>
</tr>
<tr>
<td>gas and air</td>
<td>3</td>
<td>6</td>
</tr>
<tr>
<td>pethadine</td>
<td>6</td>
<td>2</td>
</tr>
<tr>
<td>epidural</td>
<td>4</td>
<td>2</td>
</tr>
<tr>
<td>general anaesthetics</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td>spinal block</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>assistance required?</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>caesarean required?</td>
<td>4</td>
<td>2</td>
</tr>
<tr>
<td>foetal distress?</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>admission to neonatal unit?</td>
<td>8</td>
<td>6</td>
</tr>
</tbody>
</table>

* All confidence intervals (ci) are for differences in the percentages (PSO - No PSO)

Table 7: Risk factors for developmental delay and/or feeding problems

-199-
6.5.3 Later postnatal factors

*Infants' medical problems identified during the neonatal period*

Twenty-one infants were completely well at birth. Eighteen had mild jaundice, but none required intervention other than having their cot by the window in sunlight. One infant was diagnosed with a cardiac arrhythmia but no medical or surgical intervention was required. Ten infants had some respiratory difficulties. Most of these infants had ICP (n=8). There was a tendency for the number of infants in the ICP PSO group to be greater than in the No PSO group, although this was not significant. At the first data collection point (within 1 month of birth), these respiratory difficulties were being managed with positioning.

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP (n=33)</th>
<th>ICP (n=16)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO n=16</td>
<td>PSO n=17</td>
</tr>
<tr>
<td>No medical problems</td>
<td>7</td>
<td>10</td>
</tr>
<tr>
<td>Number of medical problems</td>
<td>9</td>
<td>7</td>
</tr>
<tr>
<td>Nature of medical problems</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Jaundice?</td>
<td>7</td>
<td>6</td>
</tr>
<tr>
<td>cardiac problems?</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>respiratory problems?</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td>gastrointestinal problems?</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>neurological problems?</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>other</td>
<td>0</td>
<td>0</td>
</tr>
</tbody>
</table>

* All confidence intervals are for differences in the percentages (PSO – No PSO)

<table>
<thead>
<tr>
<th></th>
<th>No PSO</th>
<th>PSO</th>
<th>p values and ci</th>
<th>No PSO</th>
<th>PSO</th>
<th>p values and ci</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>7</td>
<td>10</td>
<td>p=0.45</td>
<td>3</td>
<td>1</td>
<td>p=0.46</td>
</tr>
<tr>
<td></td>
<td>9</td>
<td>7</td>
<td>p=0.45</td>
<td>4</td>
<td>6</td>
<td>p=0.46</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>0</td>
<td>p=0.4</td>
<td>1</td>
<td>1</td>
<td>p=0.4</td>
</tr>
</tbody>
</table>

Table 8: Infant medical problems
6.6 Subsequent risk factors for developmental delay and feeding difficulties

6.6.1 Family stress

3 month assessment

By the time the infants were 3 months of age there had been few reported changes in the family environment. One mother had returned to work on a full time basis (UCLP No PSO group), 1 family had moved home (UCLP No PSO group), social services had been introduced in one family to help with a sibling who had developmental and behavioural problems (UCLP PSO group) and one infant (ICP PSO group) had become unwell requiring frequent hospital admissions. Consequently the father was required to make frequent hospital visits, care for siblings and lost his job.

6 month assessment

By the time the infants were 6 months of age more family changes were reported. Within the UCLP No PSO group 1 family had moved home, 1 infant had been subject to child abuse and was removed from the parents care by social services, 2 mothers had returned to work on a full time basis and another on a part time basis. Within the UCLP PSO group, 1 mother had returned to work on a full time basis and a further family moved into another family member’s home. There were no changes reported in the ICP No PSO group. In the ICP PSO group, 1 mother had returned to work on a full time basis, and 2 had moved home.

12 month assessment

At the 12 month assessment point, the majority of changes within the family structure were related to mothers’ returning to work on a full time or part time basis. One family moved home, another father had become unemployed.
and 1 family had split up with the father leaving. As at previous assessment points, the distribution of these changes was fairly evenly spread across the four groups, with 4 parents in the UCLP No PSO group, 5 parents in the UCLP PSO group, and 2 parents in the ICP No PSO group reporting changes. No changes were reported in the ICP PSO group.

6.6.2 Infants' medical problems

3 month assessment

The majority of infants were well at 3 months of age. The majority of infants with ICP had been given a diagnosis of Pierre Robin Sequence (Masarei et al., 1999) (n=5 in ICP No PSO group; n=6 in ICP PSO group). Two of the infants with ICP, who had been diagnosed earlier with respiratory problems (which had been managed with positioning) had worsened and by 3 months of age and had been referred to Respiratory Medicine with subsequent placement of nasopharyngeal airways.

The reported respiratory difficulties at birth for the infants with UCLP had resolved by 3 months of age. However another 2 infants (1 UCLP No PSO group and 1 PSO group) were reported to have developed respiratory problems characterised by frequent coughs and colds requiring antibiotic treatment.

Gastro-oesophageal reflux requiring medical intervention (medication) was diagnosed in a further 9 cases (n=4 in UCLP No PSO group; n=2 in UCLP PSO group; n=1 in ICP No PSO group; n=2 in ICP PSO group).

One infant (UCLP No PSO group) had been diagnosed with biliary atresia and underwent surgical management of this at 3 months of age.
6 month assessment

There were few further medical problems diagnosed by the time the infants were 6 months of age. The 2 infants with nasopharyngeal airways were now managing without these in situ. The 2 cases of respiratory difficulties in the UCLP groups persisted and an additional infant (UCLP No PSO) was reported to have similar problems. The infant with biliary atresia (UCLP No PSO) had undergone surgical repair and was recovering, but had been admitted to hospital with a head injury and fractures reportedly due to child abuse. This child was consequently in the care of a foster mother and social worker. Three infants had had chickenpox in the 3 months since the previous assessment. The frequency of gastro-oesophageal reflux requiring medication for management had changed slightly (n=3, UCLP No PSO; n=4, UCLP PSO; n=1 ICP No PSO; n=0 ICP PSO).

12 months of age

By 12 months of age, the nature of reported medical problems had changed a little. Most of the problems were related to childhood illness and coughs and colds (n=4 UCLP No PSO; n=5 UCLP PSO; n=3 ICP No PSO and n=1 ICP PSO). Gastro-oesophageal reflux requiring medical intervention had resolved in all but 3 cases (UCLP PSO group).

6.6.3 General development

Only one infant (ICP PSO) had delayed development at 3, 6 or 12 months of age.

At 3 months of age 40 infants (n=13 UCLP No PSO; n=15 UCLP PSO; n=6 ICP No PSO; n=6 ICP PSO) were rated as normal on the Denver II. Seven were rated as suspect (n=2 UCLP No PSO and n=2 UCLP PSO; n=1 ICP No PSO; n=2 ICP PSO) but on examination by a paediatrician were found to have normal development. One infant only was diagnosed as developmentally delayed (ICP PSO).
A similar picture was evident at 6 and 12 months of age.

6.7 Feeding advice

All infants received feeding advice from the clinical nurse specialist and speech and language therapist as detailed in Chapter 5.

6.8 Orthodontics

There was a tendency for infants randomised to the PSO groups to have more orthodontic appointments than those managed without, as can be seen in Table 9. This difference was significant (p=0.008) between the UCLP groups (ci 0.73 to 4.63). There was no significant difference (p=0.1) between the ICP groups (ci -0.39 to 4.73) although the confidence intervals were wide. In both the UCLP and ICP groups the number of extra appointments was between 2 and 3 on average. According to the protocol the PSO and No PSO groups would be expected to have had the same number of orthodontic visits. However as can be seen parents of infants not managed with PSO did not attend orthodontic review appointments as regularly.

There were few problems related to the PSO. In 9 cases orthodontists reported loose fitting plates. These problems were resolved with slight modifications or in 2 cases new plates were made. Orthodontic problems reported included oral thrush (managed in all cases with Daktarin gel), minor ulceration requiring no intervention, and neonatal teeth, which were removed as required (Table 9).

The incidence of intra-oral problems such as oral thrush and ulceration is shown in Table 9.
<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td>No PSO n=16</td>
<td>PSO n=17</td>
<td>p values and ci</td>
</tr>
<tr>
<td>number of orthodontic visits</td>
<td></td>
<td></td>
</tr>
<tr>
<td>mean</td>
<td>6.2</td>
<td>8.88</td>
</tr>
<tr>
<td>range</td>
<td>2-10</td>
<td>3-13</td>
</tr>
<tr>
<td>SD</td>
<td>2.45</td>
<td>2.89</td>
</tr>
<tr>
<td>orthodontic problems</td>
<td></td>
<td></td>
</tr>
<tr>
<td>poorly fitting plate</td>
<td>0</td>
<td>7</td>
</tr>
<tr>
<td>ulceration</td>
<td>4</td>
<td>2</td>
</tr>
<tr>
<td>oral thrush</td>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>neonatal teeth</td>
<td>1</td>
<td>3</td>
</tr>
</tbody>
</table>

*Confidence intervals are for differences in the mean number of orthodontic visits (PSO vs No PSO)

Table 9: Orthodontic visits and problems

6.9 Compliance with PSO

At the neonatal assessment all but 2 infants (n=25) were wearing their PSO all day except for cleaning. Both infants who were not wearing their PSO consistently wore them only for feeding. In one case the parents were unhappy to sleep their infant on his side or front, and felt concerned about the plate “dropping” when the infant was sleeping on his back. By 3 months this family had abandoned using the PSO completely. In the other case, the parents reported that the presence of a neonatal tooth meant that the plate did not fit well and caused their infant discomfort. By three months however this infant was tolerating his PSO well. In addition another family had at 3 months, abandoned use of the PSO reporting that there were problems with
it fitting securely and the infant not tolerating it. By 6 months, there was significantly poorer compliance. Only 14 of the 23 infants allocated to PSO, who had reached 6 months of age during the study, were still wearing their PSOs all day. Another infant wore the plate for 12 hours a day. The remaining 8 infants were not wearing their PSO at all. Parents reported that they had abandoned use of the PSO because they were not tolerated by their infant or because “the PSO didn’t seem to help feeding and was a bother”.

6.10 Intention to treat analyses

In this section, the results of the assessments are presented. All comparisons are based on intention to treat. This section begins with the results of the information obtained by parent report, providing an overview of the infants’ feeding methods, techniques and equipment used. Parent reported feeding symptoms, including nasal regurgitation, and risk factors for aspiration are then outlined. This is followed by the results of assessments administered by the researcher including oral motor skills, physiological measures of bottle-feeding, assessment of the pharyngeal stage of swallowing (videofluoroscopic assessment) and anthropometry.

While the primary outcome measures are collected at 12 months of age and secondary outcome measures at 3 and 6 months of age, in order to aid the reader the results are presented in chronological order.

6.10.1 Feeding methods

Neonatal assessment

At the first assessment (within 1 month) the majority of infants (n=38) were being fed by bottle only. However, 11 infants were both breast and bottle-fed or, in one case breast-fed with supplements given from a “scoop bottle”. Interestingly, 22 mothers had tried breast-feeding but had stopped prior to data collection point 1. The majority of mothers reported the reason for
stopping breast-feeding was that the infant was unable to latch on successfully. As seen in Table 10, the numbers for these feeding methods used were remarkably similar. There were no significant differences between the No PSO and PSO groups for either cleft type for method of feeding used.

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP (n=33)</th>
<th>ICP (n=16)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO N=16</td>
<td>PSO n=17</td>
</tr>
<tr>
<td>Breast-feeding</td>
<td>4</td>
<td>6</td>
</tr>
<tr>
<td></td>
<td>p=0.62 95% CI 22 to 47</td>
<td>p=0.93 95% CI 16 to 29</td>
</tr>
<tr>
<td>Bottle-feeding</td>
<td>16</td>
<td>16</td>
</tr>
<tr>
<td></td>
<td>p=0.98 95% CI 11 to 22</td>
<td>p=1 95% CI 4 to 50</td>
</tr>
<tr>
<td>Other feeding method</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td></td>
<td>p=1 95% CI 71 to 87</td>
<td>p=1 95% CI 71 to 87</td>
</tr>
<tr>
<td>Tube feeding</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Number of feeding methods used</td>
<td>12</td>
<td>11</td>
</tr>
<tr>
<td></td>
<td>p=0.56 95% CI 37 to 75</td>
<td>p=0.58 95% CI 49 to 63</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>6</td>
</tr>
<tr>
<td></td>
<td>p=0.28 95% CI 22 to 49</td>
<td>p=0.30 95% CI 38 to 65</td>
</tr>
</tbody>
</table>

"All confidence intervals are for differences in the percentages (PSO-No PSO)

*Table 10: Summary of feeding methods used at neonatal assessment*

Closer investigation revealed that 11 mothers were using 2 different types of bottles when feeding their infants. In all but one case, the 2 bottles used were soft and squeezable bottles; the Mead Johnson and Soft Plas bottles. In the remaining case, the infant was being fed with the Mead Johnson bottle and a standard rigid bottle.

In total, 47 infants were fed with soft, squeezable bottles (either the Mead Johnson and/or the Soft Plas). There were 2 infants who were fed with “scoop bottles”. This method was used in one case as the mother was
persisting with breast-feeding and had been advised by the midwife that the infant should not be given a teat, to avoid the possibility of nipple confusion. In the other case the scoop bottle was being used as the infant was unable to use a soft, squeezable bottle.

Thirty-nine parents chose to restrict their use of teats to one kind, however in 7 cases 2 different teats were used.

Adaptation of teats, is standard practice in the establishment of feeding in infants with CL and/or P. At this assessment parents adapted teats in 29 cases. These adaptations consisted of enlarging pre-existing holes in the teats (6 cases), creating extra holes in the teat (20 cases) or using a fast flow version of the teat (3 cases). There were no significant differences in the number of adaptations made to teats between the No PSO and PSO groups, for either cleft group, although confidence intervals were wide because of the small numbers.

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP (n=33)</th>
<th>ICP (n=16)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO : PSO</td>
<td>No PSO : PSO</td>
</tr>
<tr>
<td></td>
<td>n=16 : n=17</td>
<td>n=8 : n=8</td>
</tr>
<tr>
<td>Enlarging holes</td>
<td></td>
<td></td>
</tr>
<tr>
<td>1:1</td>
<td>2:2</td>
<td></td>
</tr>
<tr>
<td>Creating new hole(s)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>7:8</td>
<td>1:4</td>
<td></td>
</tr>
<tr>
<td>Using &quot;fast flow&quot; version of teat</td>
<td></td>
<td></td>
</tr>
<tr>
<td>1:1</td>
<td>0:1</td>
<td></td>
</tr>
</tbody>
</table>

*All confidence intervals are for differences in the percentages (PSO – No PSO)*

Table 11: Summary of modifications to teats at neonatal assessment
3 month assessment

At 3 months of age a greater proportion of infants were being fed by bottle only. Only 2 mothers had continued breast-feeding and this was supplemented with bottle feeds. There were also 3 infants who were not being fed orally but were being fed by a nasogastric tube (Table 12), as they had been found to be at risk of aspiration. Two infants (n=1, ICP No PSO and n=1 ICP PSO) had nasopharyngeal airways in situ for airway management and 1 infant was aspirating silently (as seen on videofluoroscopy) and failing to thrive.

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th></th>
<th>ICP</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO</td>
<td>PSO</td>
<td>p values and ci</td>
<td>No PSO</td>
</tr>
<tr>
<td>Breast feeding</td>
<td>0</td>
<td>1</td>
<td>p=0.75 0.24 to 0.45</td>
<td>1</td>
</tr>
<tr>
<td>Bottle feeding</td>
<td>14</td>
<td>17</td>
<td>p=0.75 0.44 to 0.24</td>
<td>6</td>
</tr>
<tr>
<td>Tube feeding</td>
<td>1</td>
<td>0</td>
<td>p=0.75 0.44 to 0.24</td>
<td>1</td>
</tr>
<tr>
<td>Number of feeding methods used</td>
<td>1</td>
<td>15</td>
<td>16</td>
<td>p=0.75 0.43 to 0.24</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>0</td>
<td>1</td>
<td>p=0.75 0.43 to 0.24</td>
</tr>
</tbody>
</table>

NB: 2 infants omitted; 1 failed to attend assessment and 1 was not 3 months of age

* All confidence intervals are for differences in the percentages (PSO – No PSO)

Table 12: Feeding methods used at 3 months of age
As at the previous assessment, the majority of infants who were feeding orally were being fed with soft, squeezable bottles (Mead Johnson or Soft Plas) (Table 13). One infant continued to be fed with a “scoop bottle” as he persistently failed with a standard teat. It is of interest that 2 infants (UCLP) were managing feeding with a standard rigid bottle. Both were from the UCLP PSO group. As at the neonatal assessment, there was no significant difference in feeding methods used, between the No PSO and PSO groups for either cleft type. Confidence intervals were however wide.

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO</td>
<td>PSO</td>
</tr>
<tr>
<td>Using Mead Johnson bottle</td>
<td>9</td>
<td>7</td>
</tr>
<tr>
<td>Using Soft Plas bottle</td>
<td>11</td>
<td>13</td>
</tr>
<tr>
<td>Using standard bottle</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>Using scoop bottle</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>Number of bottles being used</td>
<td>1</td>
<td>8</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>6</td>
</tr>
</tbody>
</table>

NB: 3 infants omitted from this table as they were not feeding orally (being fed through NG tubes)
* All confidence intervals are for differences in the percentages (PSO – No PSO)

Table 13: Bottles used at 3 months of age
The majority of mothers continued to use NUK size 1 and/or 2 teats, in conjunction with soft, squeezable bottles (n=33). A minority (n=11) changed to standard teats or cross-cut teats.

By 3 months of age, there had been an increase in the number of adaptations to teats, although the majority of mothers had made only one adaptation (Table 14). As previously the adaptations generally involved enlarging the pre-existing hole in the teat, creating additional holes or using a fast flow version of the teat.

As can be seen in Table 14, there were no significant differences between the No PSO and PSO groups for either cleft type.

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO</td>
<td>PSO</td>
</tr>
<tr>
<td>Number of adaptations</td>
<td>12</td>
<td>10</td>
</tr>
<tr>
<td></td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>Using teat upside down</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Enlarging holes</td>
<td>2</td>
<td>1</td>
</tr>
<tr>
<td>Creating new hole(s)</td>
<td>8</td>
<td>8</td>
</tr>
<tr>
<td>Using &quot;fast flow&quot; version of teat</td>
<td>3</td>
<td>1</td>
</tr>
</tbody>
</table>

NB: 5 infants are omitted from this table; 1 infant failed to attend the assessment, 3 were not feeding orally and 1 infant was feeding with a scoop bottle.

* All confidence intervals are for differences in the percentages (PSO – No PSO)

Table 14: Summary of modifications to teats at 3 months
6 month assessment

By 6 months of age, only 1 mother continued to breast-feed. With the exception of 1 infant who continued to be fed non-orally by nasogastric tube (ICP No PSO), all infants were being fed with bottles (Table 15). There were no significant differences in methods of feeding used between the No PSO and PSO groups for either cleft type (Table 15).

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO</td>
<td>PSO</td>
</tr>
<tr>
<td>Breast-feeding</td>
<td>n=14</td>
<td>n=15</td>
</tr>
<tr>
<td>Bottle-feeding</td>
<td>14</td>
<td>15</td>
</tr>
<tr>
<td>Tube-feeding</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Number of feeding methods used</td>
<td>1</td>
<td>13</td>
</tr>
<tr>
<td>2</td>
<td>1</td>
<td>0</td>
</tr>
</tbody>
</table>

* All confidence intervals are for differences in the percentages (PSO - No PSO)

Table 15: Feeding methods used at 6 months of age
The majority of infants continued to be fed with soft, squeezable bottles, however there were 5 infants (n=1 UCLP No PSO; n=4 UCLP PSO) who were being fed with standard rigid bottles at least some of the time (Table 16). As one might expect at this developmental stage, 3 infants were also being given fluids from a standard beaker, training beaker or child sports bottle (Table 16). There was no significant difference in the bottles used, between the No PSO and PSO groups for either cleft type.

<table>
<thead>
<tr>
<th></th>
<th>Cleft Type</th>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>UCLP</td>
<td>No PSO</td>
<td>PSO</td>
<td></td>
<td></td>
<td>ICP</td>
<td>No PSO</td>
<td>PSO</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>n=14</td>
<td>n=15</td>
<td>p values and ci</td>
<td>n=7</td>
<td>p values and ci</td>
<td>n=7</td>
<td>p values and ci</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Using Mead Johnson bottle</td>
<td>5</td>
<td>1</td>
<td>1</td>
<td>(p=0.1)</td>
<td>4.4 to 9.9</td>
<td>1</td>
<td>1</td>
<td>(p=1)</td>
<td>54 to 31</td>
<td></td>
</tr>
<tr>
<td>Using Soft Plas bottle</td>
<td>11</td>
<td>10</td>
<td>5</td>
<td>(p=0.054)</td>
<td>1.3 to 23</td>
<td>6</td>
<td>(p=0.73)</td>
<td>43 to 68</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Using standard bottle</td>
<td>1</td>
<td>4</td>
<td>0</td>
<td>(p=0.27)</td>
<td>0.0 to 30</td>
<td>0</td>
<td>0</td>
<td>(p=1)</td>
<td>54 to 31</td>
<td></td>
</tr>
<tr>
<td>Using scoop bottle</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>(p=0.75)</td>
<td>0.0 to 48</td>
<td>0</td>
<td>0</td>
<td>(p=1)</td>
<td>34 to 74</td>
<td></td>
</tr>
<tr>
<td>Using training beaker</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>(p=1)</td>
<td>41 to 31</td>
<td>1</td>
<td>(p=0.75)</td>
<td>42 to 76</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Using beaker</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>(p=0.75)</td>
<td>24 to 47</td>
<td>0</td>
<td>0</td>
<td>(p=1)</td>
<td>34 to 74</td>
<td></td>
</tr>
<tr>
<td>Using other method</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>(p=1)</td>
<td>41 to 31</td>
<td>0</td>
<td>0</td>
<td>(p=0.75)</td>
<td>63 to 42</td>
<td></td>
</tr>
</tbody>
</table>

* All confidence intervals are for differences in the percentages (PSO – No PSO)

Table 16: Bottles used at 6 months of age
The majority of mothers continued to modify their infants’ teats at 6 months (n=32) with a slight increase in the number of modifications within the ICP groups (n=4 ICP No PSO; n=5 ICP PSO). There was no significant difference in the number of modifications to teats, between the No PSO and PSO groups for either cleft type (Table 17).

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO</td>
<td>PSO</td>
</tr>
<tr>
<td>Number of adaptations</td>
<td>1</td>
<td>12</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td>Using teat upside down</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Enlarging holes</td>
<td>1</td>
<td>4</td>
</tr>
<tr>
<td>Creating new hole(s)</td>
<td>6</td>
<td>5</td>
</tr>
<tr>
<td>Cross cutting teat</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>Using &quot;fast flow&quot; version of teat</td>
<td>5</td>
<td>2</td>
</tr>
</tbody>
</table>

NB: 2 infants are omitted; 1 infant was being fed with scoop bottle and therefore no teat, and 1 infant was receiving fluids via NG.

* All confidence intervals are for differences in the percentages (PSO – No PSO)

Table 17: Adaptations made to teat at 6 months of age
Until recently it was standard practice to introduce solids before 6 months of age (World Health Organisation, 2001a; World Health Organisation, 2001b). With the exception of 1 infant who was being fed via nasogastric tube (ICP No PSO) all infants were reported to be taking developmentally appropriate solids (runny puree or thicker puree). Several infants were taking additional or “advanced” (7 month puree, lumpy puree, bite and dissolve foods and chocolate) textures for their age (Table 18). Thick puree was more likely to be taken by infants managed with PSO (both UCLP and ICP groups). This difference was highly significant with 58 % more infants in the UCLP PSO group taking thick puree (p=0.0042, ci 20 to 87%). There was a non significant trend towards an increased acceptance of 7 month and lumpy purees by infants in the PSO groups.

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP No PSO</th>
<th>UCLP PSO</th>
<th>ICP No PSO</th>
<th>ICP PSO</th>
<th>p values and ci</th>
<th>p values and ci</th>
</tr>
</thead>
<tbody>
<tr>
<td>Fluids</td>
<td>N=14</td>
<td>n=15</td>
<td>n=7</td>
<td>n=7</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Runny puree</td>
<td>14</td>
<td>15</td>
<td>7</td>
<td>6</td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Thick puree</strong></td>
<td><strong>12</strong></td>
<td><strong>15</strong></td>
<td><strong>7</strong></td>
<td><strong>6</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>“7 month” puree</td>
<td>4</td>
<td>13</td>
<td>3</td>
<td>6</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Lumpy puree</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>2</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Bite and dissolve foods</td>
<td>1</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>other solids (chocolate buttons)</strong></td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Table 18: Textures accepted at 6 months of age

* All confidence intervals are for differences in the percentages (PSO – No PSO)
**12 month assessment**

By 12 months of age no infants in the cohort were being breast-fed. All of the infants were taking fluids from a standard bottle and/or training beaker. One infant (ICP No PSO) remained on supplementary nasogastric feeds (Table 19).

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO</td>
<td>PSO</td>
</tr>
<tr>
<td>Breast feeding</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Bottle/beaker feeding</td>
<td>8</td>
<td>13</td>
</tr>
<tr>
<td>Tube feeding</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Number of feeding methods used</td>
<td>1 8 13</td>
<td>p = 1, 53% to 3%</td>
</tr>
<tr>
<td></td>
<td>2 0</td>
<td>0</td>
</tr>
</tbody>
</table>

* All confidence intervals are for differences in the percentages (PSO – No PSO)

**Table 19: Feeding methods used at 12 months of age**
It might have been expected that following repair of the palate the majority of infants would have progressed to taking fluids from a standard bottle. This was not, however, the case with 7 infants still being fed with soft and squeezable bottles (n=3 UCLP No PSO; n=2 UCLP PSO; n=0 ICP No PSO; n=2 ICP PSO). Fourteen infants took fluids from a beaker or training beaker. The beakers used were in all but one case unvalved (i.e. not “anyway up beakers”). All mothers had tried valved beakers but had abandoned them or removed the valving system, as their infants had not been able to manage them. There was no significant difference in terms of method used for fluids, between the No PSO and PSO groups for either cleft type.

<table>
<thead>
<tr>
<th>Number of bottles/beakers used</th>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO</td>
<td>PSO</td>
<td>p values</td>
</tr>
<tr>
<td>1</td>
<td>n=8</td>
<td>n=13</td>
<td>86.7%</td>
</tr>
<tr>
<td>2</td>
<td>0%</td>
<td>10%</td>
<td>53.1%</td>
</tr>
<tr>
<td>3</td>
<td>1%</td>
<td>1%</td>
<td>58.6%</td>
</tr>
<tr>
<td>Using Mead Johnson bottle</td>
<td>0%</td>
<td>0%</td>
<td>53.1%</td>
</tr>
<tr>
<td>Using Soft Plas bottle</td>
<td>3%</td>
<td>2%</td>
<td>69.2%</td>
</tr>
<tr>
<td>Using standard bottle</td>
<td>5%</td>
<td>9%</td>
<td>36.8%</td>
</tr>
<tr>
<td>Using Scoop bottle</td>
<td>0%</td>
<td>1%</td>
<td>36.8%</td>
</tr>
<tr>
<td>Using training beaker</td>
<td>1%</td>
<td>2%</td>
<td>40.9%</td>
</tr>
<tr>
<td>Using beaker</td>
<td>0%</td>
<td>3%</td>
<td>22.6%</td>
</tr>
<tr>
<td>Using other method</td>
<td>1%</td>
<td>0%</td>
<td>54.1%</td>
</tr>
</tbody>
</table>

* All confidence intervals are for differences in the percentages (PSO – No PSO).

Table 20: Bottles/beakers used at 12 months of age
At 12 months of age the majority of infants managed to feed with unadapted teats (n= 25). Modifications were used in 9 cases only (n=3 UCLP No PSO; n=3 UCLP PSO; n=1 ICP No PSO; n=2 ICP PSO) (Table 21).

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>No PSO</td>
<td>2</td>
<td>1</td>
</tr>
<tr>
<td>PSO</td>
<td>3</td>
<td>2</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>No PSO</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>PSO</td>
<td>1</td>
<td>0</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number of adaptations</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>2</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td>Using teat upside down</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td></td>
<td>0</td>
<td>0</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td>Enlarging holes</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td></td>
<td>1</td>
<td>1</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td>Creating new hole(s)</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td></td>
<td>0</td>
<td>1</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cross cutting teat</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td></td>
<td>0</td>
<td>0</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td>Using &quot;fast flow&quot; version of teat</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td></td>
<td>1</td>
<td>1</td>
</tr>
</tbody>
</table>

* All confidence intervals are for differences in the percentages (PSO - No PSO)

Table 21: Summary of modifications to teats at 12 months (for 9 infants requiring adaptations)
Acceptance of solids for the majority of infants was age appropriate. Several infants had not been offered textures such as mashed or chopped table foods as mothers either did not think it age appropriate or preferred to puree all foods for “ease and speed” of feeding. There was no difference between the No PSO and PSO groups of either cleft type with regards to the range of solids infants were taking (Table 22). In the UCLP group there was a trend towards more infants with PSO accepting mashed table foods than those managed without PSO. There was a slightly greater proportion of infants in the UCLP PSO group taking chopped table foods and other solids although the differences were not significant.

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>Fluids</th>
<th>Runny puree</th>
<th>Thick puree</th>
<th>7 month puree</th>
<th>Lumpy puree</th>
<th>Bite and dissolve foods</th>
<th>Mashed table foods</th>
<th>Soft solids</th>
<th>Chopped table foods</th>
<th>Other solids</th>
</tr>
</thead>
<tbody>
<tr>
<td>UCLP No PSO n=8</td>
<td>8</td>
<td>13</td>
<td>8</td>
<td>8</td>
<td>8</td>
<td>7</td>
<td>4</td>
<td>7</td>
<td>4</td>
<td>0</td>
</tr>
<tr>
<td>UCLP PSO n=13</td>
<td></td>
<td></td>
<td>13</td>
<td>13</td>
<td>12</td>
<td>12</td>
<td>12</td>
<td>12</td>
<td>12</td>
<td>1</td>
</tr>
<tr>
<td>p values</td>
<td></td>
<td>p=0.76</td>
<td>p=0.90</td>
<td>p=0.88</td>
<td>p=1</td>
<td>p=0.56</td>
<td>p=1</td>
<td>p=0.62</td>
<td>p=1</td>
<td>p=0.70</td>
</tr>
<tr>
<td>CI</td>
<td></td>
<td>0.33 to 0.5</td>
<td>0.59 to 0.9</td>
<td>0.59 to 0.9</td>
<td>0.01 to 0.62</td>
<td>0.56 to 0.76</td>
<td>0.01 to 0.62</td>
<td>0.01 to 0.62</td>
<td>0.01 to 0.76</td>
<td>0.01 to 0.85</td>
</tr>
<tr>
<td>ICP No PSO n=6</td>
<td>6</td>
<td>7</td>
<td>5</td>
<td>4</td>
<td>4</td>
<td>5</td>
<td>4</td>
<td>5</td>
<td>4</td>
<td>1</td>
</tr>
<tr>
<td>ICP PSO n=7</td>
<td></td>
<td></td>
<td>5</td>
<td>6</td>
<td>6</td>
<td>7</td>
<td>7</td>
<td>6</td>
<td>7</td>
<td>0</td>
</tr>
<tr>
<td>p values</td>
<td></td>
<td>p=1</td>
<td>p=1</td>
<td>p=1</td>
<td>p=1</td>
<td>p=1</td>
<td>p=1</td>
<td>p=1</td>
<td>p=1</td>
<td>p=1</td>
</tr>
<tr>
<td>CI</td>
<td></td>
<td>0.90 to 0.92</td>
<td>0.90 to 0.92</td>
<td>0.90 to 0.92</td>
<td>0.90 to 0.92</td>
<td>0.90 to 0.92</td>
<td>0.90 to 0.92</td>
<td>0.90 to 0.92</td>
<td>0.90 to 0.92</td>
<td>0.90 to 0.92</td>
</tr>
</tbody>
</table>

* All confidence intervals are for differences in the percentages (PSO – No PSO)

Table 22: Textures accepted at 12 months
6.10.2 Parental report of feeding characteristics

Length of feed

*Neonatal assessment*

The time taken for an infant to take the required amount of feed is often used as a guide to feeding efficiency (Section 3.2). Parents were asked to estimate the average time taken for their infant to complete a feed. There was some variability in this measure. In the UCLP No PSO group the mean length of feed was 37.5 minutes (mode 30, range 10-60) in contrast to 30 minutes (mode 30, range 15-60) for the UCLP PSO group. This was not statistically different (p= 0.12, ci –1.96 to 16.97). For the ICP groups, the difference was in the other direction with the No PSO group having a mean time of 38.13 minutes (mode 20, range 15-90 minutes), 4.97 minutes less on average (p= 0.62, ci –26.44 to 16.44) than the PSO group (mode 30, range 30-60 minutes).

*3 month assessment*

At 3 months of age, there was a non-significant trend for infants managed with PSO in both UCLP and ICP groups to feed in a shorter time than those without PSO. For the UCLP No PSO group the mean length of a feed was 29.29 minutes, slightly longer than that of the UCLP PSO group, which was 25.59 minutes (p=0.46, ci –6.32 to 13.71). Similarly within the ICP groups, there was no significant difference in the length of feeds (ICP No PSO mean 34.17; ICP PSO mean 29.29; p=0.59, ci –14.45 to 24.15).

*6 month assessment*

Similar results were obtained at 6 months of age, where there appeared to be a trend for shorter feeds associated with PSO in both the UCLP (UCLP No PSO mean 24.57; UCLP PSO mean 17.33) and ICP (ICP No PSO mean
26.67; ICP PSO mean 17.86) groups. The differences were however not significant (UCLP p=0.24, ci -5.31 to 19.80; ICP p=0.33, ci -10.29 to 27.91).

**12 month assessment**

Within the UCLP groups a similar non-significant trend was evident at 12 months, with the UCLP No PSO group having a mean feed time of 15.62 minutes and the UCLP PSO group a mean time of 12.31 minutes (p=0.33, d -3.34 to 10.32). However within the ICP groups the difference was more marked with infants managed with PSO feeding in a mean shorter period of time (ICP No PSO mean 22 minutes, ICP PSO mean 6.71 minutes; p=0.01, ci 3.66 to 26.91).

The changes in each infant's reported length of feed can be traced over time in Figures 55 to 58. In these graphs, each infant is represented by a different colour within their allocated group i.e. UCLP No PSO, UCLP PSO, ICP No PSO or ICP PSO.

The analysis at the different assessment times showed a fairly consistent, although for the most part statistically non-significant reduction in feed lengths amongst the infants allocated to PSO. Multilevel modelling of feed lengths across assessments was performed so that the measurements could be investigated over time taking into account the exact ages at which the assessments were made. The changes in feed length over time showed an exponential decline with age and hence feed lengths were logged when fitting the models. The best fitting model indicated that feed length amongst the PSO group were on average 97% of those in the No PSO groups (95% ci 76 to 123%, p=0.78) at the neonatal assessment and that the length of feeding fell significantly faster as the infants grew older. Overall feed lengths declined by 7% (95% ci 4 to 9%, p=0.00000017) for each extra month of age and this fall was increased by 4% (95% ci 1 to 7%, p=0.0186) amongst the infants allocated to PSO. There was no significant difference in either initial feed lengths or the later decline in lengths between the UCLP and ICP groups.
Figure 57: Length of feeding over time for ICP NO PSO group

Figure 58: Length of feeding over time for ICP PSO group
**Time taken to finish solid meals**

**6 month assessment**

At the 6 month assessment parents were asked how long it took their infant to complete a solid feed. This was difficult in some cases, as behavioural aspects of feeding such as “playing with food” or attempts at “self feeding” were reported to affect the time taken. There was however, no significant difference in the reported time infants took to complete solid feeds in either the UCLP or ICP groups at 6 months of age (UCLP p=0.41, ci -5.70 to 2.41; ICP p= 1, ci -8.71 to 8.71) (Table 23).

**12 month assessment**

At this assessment point, behavioural aspects of feeding, in particular self-feeding, made it even more difficult than at previous assessment points for some parents to report the length of time taken to complete solid feeds. In contrast to previous results, at 12 months of age the UCLP PSO group took slightly longer than those managed without PSO, although the difference was not significant (Table 23). The reverse was apparent in the ICP groups where the mean length of time for those infants managed without PSO was longer than those managed with PSO (p= 0.43, ci -11.12 to 24.22).
### Feeding characteristics

Other feeding characteristics reported to be present in infants with cleft lip and palate include nasal regurgitation, coughing and choking during feeds (Section 2.1).

### Neonatal assessment

Nasal regurgitation during feeds was reported in only 13 cases. The frequency was reported as rare (less than once a day) in 7 cases, and occurring once or twice a day in 6 cases. The amount of nasal regurgitation during feeds was reported as trace in all cases.

---

**Table 23: Reported length of solid feeds**

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO</td>
<td>PSO</td>
</tr>
<tr>
<td></td>
<td>n=8</td>
<td>n=13</td>
</tr>
<tr>
<td><strong>Length of feed</strong></td>
<td><strong>Mean</strong></td>
<td>10.36</td>
</tr>
<tr>
<td>(6 months)</td>
<td><strong>Minimum</strong></td>
<td>0</td>
</tr>
<tr>
<td></td>
<td><strong>Maximum</strong></td>
<td>20</td>
</tr>
<tr>
<td><strong>Length of feed</strong></td>
<td><strong>Mean</strong></td>
<td>13.75</td>
</tr>
<tr>
<td>(12 months)</td>
<td><strong>Minimum</strong></td>
<td>10</td>
</tr>
<tr>
<td></td>
<td><strong>Maximum</strong></td>
<td>20</td>
</tr>
</tbody>
</table>
Coughing and choking during or after feeds was reported in 33 cases. Similarly in 33 cases gurgley breathing during or after feeding (specific to the laryngeal region rather than the nasopharyngeal region) was reported.

Infants were reported to show signs of discomfort such as arching, crying or pulling away from the bottle during feeding in 16 cases. Vomiting after feeds was reported frequently (n=8, UCLP No PSO; n=8 UCLP PSO; n=5 ICP No PSO; n=3 ICP PSO).

There were no significant differences between the No PSO and PSO groups for either cleft type for any of the feeding characteristics.

<table>
<thead>
<tr>
<th></th>
<th>Cleft Type</th>
<th>UCLP (n=13)</th>
<th>ICP (n=16)</th>
<th>p values and ci</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO</td>
<td>PSO</td>
<td>No PSO</td>
<td>PSO</td>
</tr>
<tr>
<td>Nasal regurgitation</td>
<td>N=16</td>
<td>n=17</td>
<td>n=8</td>
<td>n=8</td>
</tr>
<tr>
<td>Coughing or choking</td>
<td>11 13</td>
<td></td>
<td>3 6</td>
<td></td>
</tr>
<tr>
<td>during feed</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Gurgley breathing</td>
<td>9 13</td>
<td></td>
<td>6 5</td>
<td></td>
</tr>
<tr>
<td>during/post feed</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Signs of discomfort</td>
<td>5 5</td>
<td></td>
<td>2 4</td>
<td></td>
</tr>
<tr>
<td>during feeds</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Vomiting after feeds</td>
<td>8 8</td>
<td></td>
<td>5 3</td>
<td></td>
</tr>
</tbody>
</table>

* All confidence intervals are for differences in the percentages (PSO – No PSO)

Table 24: Summary of reported feeding characteristics at neonatal assessment
3 month assessment

The picture was similar at 3 months of age. The only difference from the neonatal assessment results (non-significant) was the decrease in the number of mothers who reported that their infant’s breathing sounded “gurgley” during or following feeds (n=18). Nasal regurgitation was reported in a similar number of cases to the neonatal assessment. It was similar in frequency, most commonly occurring once or twice a day, although in 3 cases it was reported to occur at every feed (n=1 UCLP No PSO; n=1 UCLP PSO; n=1 ICP No PSO). The extent of nasal regurgitation was trace in the majority of cases, although in 2 cases the amount of nasal regurgitation was reported to be “a thimble amount” rather than just trace. There were no significant differences in reported signs of discomfort during feeds or vomiting after feeds across the UCLP or ICP groups (Table 25).

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td>Nasal regurgitation</td>
<td></td>
<td></td>
</tr>
<tr>
<td>No PSO</td>
<td>PSO</td>
<td>p values and ci</td>
</tr>
<tr>
<td>n=15</td>
<td>n=17</td>
<td></td>
</tr>
<tr>
<td>2</td>
<td>5</td>
<td>p=0.56</td>
</tr>
<tr>
<td>Coughing or choking during feed</td>
<td></td>
<td></td>
</tr>
<tr>
<td>No PSO</td>
<td>PSO</td>
<td>p values and ci</td>
</tr>
<tr>
<td>n=15</td>
<td>n=17</td>
<td></td>
</tr>
<tr>
<td>10</td>
<td>12</td>
<td>p=0.56</td>
</tr>
<tr>
<td>Gurgley breathing during/post feed</td>
<td></td>
<td></td>
</tr>
<tr>
<td>No PSO</td>
<td>PSO</td>
<td>p values and ci</td>
</tr>
<tr>
<td>n=15</td>
<td>n=17</td>
<td></td>
</tr>
<tr>
<td>5</td>
<td>9</td>
<td>p=0.35</td>
</tr>
<tr>
<td>Signs of discomfort during feeds</td>
<td></td>
<td></td>
</tr>
<tr>
<td>No PSO</td>
<td>PSO</td>
<td>p values and ci</td>
</tr>
<tr>
<td>n=15</td>
<td>n=17</td>
<td></td>
</tr>
<tr>
<td>6</td>
<td>4</td>
<td>p=0.31</td>
</tr>
<tr>
<td>Vomiting after feeds</td>
<td></td>
<td></td>
</tr>
<tr>
<td>No PSO</td>
<td>PSO</td>
<td>p values and ci</td>
</tr>
<tr>
<td>n=15</td>
<td>n=17</td>
<td></td>
</tr>
<tr>
<td>8</td>
<td>11</td>
<td>p=0.66</td>
</tr>
</tbody>
</table>

* All confidence intervals are for differences in the percentages (PSO – No PSO)

Table 25: Summary of reported feeding characteristics at 3 months
6 month assessment

At 6 months, solids had been introduced and so feeding characteristics were evaluated for both fluids and solids. The incidence of reported nasal regurgitation for fluids had increased slightly in the UCLP groups and had remained steady within the ICP groups. As can be seen in Table 26, nasal regurgitation for solids was common in all groups. While there was no significant difference in the number of cases where nasal regurgitation was reported (Table 26), there was a trend (p=0.09) for the frequency to be more often within the PSO group, i.e. occurring at every feed rather than just occasionally, for the UCLP infants (Table 27). However overall there was a trend towards increased symptoms in UCLP infants managed with PSO.

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP No PSO</th>
<th>UCLP PSO</th>
<th>ICP No PSO</th>
<th>ICP PSO</th>
</tr>
</thead>
<tbody>
<tr>
<td>Nasal regurgitation with fluids</td>
<td>7</td>
<td>10</td>
<td>3</td>
<td>2</td>
</tr>
<tr>
<td>Nasal regurgitation with solids</td>
<td>11</td>
<td>12</td>
<td>6</td>
<td>3</td>
</tr>
<tr>
<td>Coughing or choking with fluids</td>
<td>7</td>
<td>8</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>Coughing or choking with solids</td>
<td>7</td>
<td>8</td>
<td>5</td>
<td>5</td>
</tr>
<tr>
<td>Gurgley breathing with fluids</td>
<td>1</td>
<td>3</td>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>Gurgley feeding with solids</td>
<td>4</td>
<td>3</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>Signs of discomfort during feeds (during solids and fluids)</td>
<td>1</td>
<td>3</td>
<td>3</td>
<td>2</td>
</tr>
<tr>
<td>Vomiting during or following feeds</td>
<td>6</td>
<td>6</td>
<td>1</td>
<td>2</td>
</tr>
</tbody>
</table>

* All confidence intervals are for differences in the percentages (PSO - No PSO)

*Table 26: Summary of reported feeding characteristics at 6 months*
<table>
<thead>
<tr>
<th>Frequency of nasal regurg. fluids</th>
<th>UCLP No PSO</th>
<th>UCLP PSO</th>
<th>ICP No PSO</th>
<th>ICP PSO</th>
</tr>
</thead>
<tbody>
<tr>
<td>occasional regurgitation with fluids</td>
<td>5 8</td>
<td>p = 0.48</td>
<td>3 2</td>
<td>p = 0.79</td>
</tr>
<tr>
<td>nasal regurg 1-2 x daily with fluids</td>
<td>1 2</td>
<td>p = 0.75</td>
<td>0 0</td>
<td>p = 0.77</td>
</tr>
<tr>
<td>nasal regurg at every feed (with fluids)</td>
<td>1 1</td>
<td>p = 0.74</td>
<td>0 0</td>
<td>p = 0.75</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Amount of nasal regurg. fluids</th>
<th>Trace amount of nasal regurgitation (with fluids)</th>
<th>5 10</th>
<th>p = 0.12</th>
<th>3 2</th>
<th>p = 0.70</th>
</tr>
</thead>
<tbody>
<tr>
<td>&quot;thimble&quot; amount of nasal regurgitation (with fluids)</td>
<td>2 0</td>
<td>p = 0.47</td>
<td>0 0</td>
<td>p = 0.75</td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Frequency of nasal regurg. solids</th>
<th>UCLP No PSO</th>
<th>UCLP PSO</th>
<th>ICP No PSO</th>
<th>ICP PSO</th>
</tr>
</thead>
<tbody>
<tr>
<td>occasional regurgitation with solids</td>
<td>6 3</td>
<td>p = 0.27</td>
<td>3 1</td>
<td>p = 0.41</td>
</tr>
<tr>
<td>nasal regurg 1-2 x daily with solids</td>
<td>2 1</td>
<td>p = 0.03</td>
<td>0 0</td>
<td>p = 0.51</td>
</tr>
<tr>
<td>nasal regurg at every feed (with solids)</td>
<td>3 8</td>
<td>p = 0.09</td>
<td>3 2</td>
<td>p = 0.79</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Amount of nasal regurg. solids</th>
<th>Trace amount of nasal regurgitation (with solids)</th>
<th>7 9</th>
<th>p = 0.08</th>
<th>5 1</th>
<th>p = 0.10</th>
</tr>
</thead>
<tbody>
<tr>
<td>&quot;thimble&quot; amount of nasal regurgitation (with solids)</td>
<td>3 3</td>
<td>p = 0.41</td>
<td>1 2</td>
<td>p = 0.91</td>
<td></td>
</tr>
<tr>
<td>&quot;dessert spoon&quot; amount of nasal regurgitation (with solids)</td>
<td>1 0</td>
<td>p = 0.72</td>
<td>0 0</td>
<td>p = 0.84</td>
<td></td>
</tr>
</tbody>
</table>

* All confidence intervals are for differences in the percentages (PSO – No PSO).

Table 27: Frequency and extent of episodes of nasal regurgitation during feeding 6 months
12 month assessment

Many feeding characteristics persisted 6 months after cleft palate repair. Nasal regurgitation of fluids persisted in half of the UCLP No PSO group and about a third of the UCLP PSO group. Within the ICP groups, nasal regurgitation of fluids persisted in a third of the ICP No PSO group. Nasal regurgitation of solids persisted in all groups. Coughing or choking with fluids was evident in all groups. There was a trend towards an increased incidence of coughing and choking in the UCLP No PSO group (p=0.03). With regard to discomfort during feeds and vomiting, there was a decrease in the number of cases across all groups (Table 28). Again there were no clear patterns of symptoms being more common amongst the No PSO or PSO groups for either UCLP or ICP.

<table>
<thead>
<tr>
<th></th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO p values and ci</td>
<td>No PSO p values and ci</td>
</tr>
<tr>
<td></td>
<td>n=8</td>
<td>n=8</td>
</tr>
<tr>
<td></td>
<td>n=13</td>
<td>n=6</td>
</tr>
<tr>
<td></td>
<td></td>
<td>n=7</td>
</tr>
<tr>
<td>Nasal regurgitation with fluids</td>
<td>4 5</td>
<td>4 2 2</td>
</tr>
<tr>
<td></td>
<td>p=0.41 (62 to 31)</td>
<td>p=0.22 (68 to 20)</td>
</tr>
<tr>
<td>Nasal regurgitation with solids</td>
<td>4 4</td>
<td>2 2</td>
</tr>
<tr>
<td></td>
<td>p=0.35 (68 to 22)</td>
<td>p=0.06 (67 to 9)</td>
</tr>
<tr>
<td>Coughing or choking with fluids</td>
<td>5 5</td>
<td>4 4</td>
</tr>
<tr>
<td></td>
<td>p=0.01 (90 to 6)</td>
<td>p=0.06 (67 to 9)</td>
</tr>
<tr>
<td>Coughing or choking with solids</td>
<td>6 3</td>
<td>5 4</td>
</tr>
<tr>
<td></td>
<td>p=0.01 (90 to 6)</td>
<td>p=0.06 (67 to 9)</td>
</tr>
<tr>
<td>Gurgley breathing with fluids</td>
<td>1 3</td>
<td>0 0</td>
</tr>
<tr>
<td></td>
<td>p=0.06 (67 to 31)</td>
<td>p=0.06 (67 to 9)</td>
</tr>
<tr>
<td>Gurgley feeding with solids</td>
<td>2 1</td>
<td>0 1</td>
</tr>
<tr>
<td></td>
<td>p=0.44 (66 to 22)</td>
<td>p=0.06 (67 to 9)</td>
</tr>
<tr>
<td>Signs of discomfort during feeds (both fluids and solids)</td>
<td>0 0</td>
<td>1 0</td>
</tr>
<tr>
<td></td>
<td>p=0.35 (68 to 20)</td>
<td>p=0.13 (90 to 6)</td>
</tr>
<tr>
<td>Vomiting during or following feeds</td>
<td>0 3</td>
<td>0 1</td>
</tr>
<tr>
<td></td>
<td>p=0.06 (67 to 31)</td>
<td>p=0.33 (62 to 18)</td>
</tr>
</tbody>
</table>

* All confidence intervals are for differences in the percentages (PSO - No PSO).

Table 28: Summary of reported feeding characteristics at 12 months
6.10.3 Oral motor skills

Neonatal oral motor assessment scale (NOMAS)

Each infant's feeding was rated twice with the NOMAS (Section 3.3), on a routine feed and on GOSMIF assessment. As the scale was standardised using a standard rigid feeding bottle, the NOMAS score taken from the GOSMIF study was considered to be the most valid, since this also used a rigid bottle. These ratings are now reported.

The majority of infants' feeding patterns were rated as disorganised or dysfunctional at the neonatal assessment (Table 29). Within the UCLP No PSO group, only 1 infant was rated as having a normal feeding pattern. Seven infants feeding patterns were rated as disorganised and 6 as dysfunctional. Two studies could not be rated because in 1 case a video was lost (copied over), and in the other a poor view prevented rating. Within the UCLP PSO group, no infants were rated as having a normal feeding pattern. Twelve infants displayed disorganised feeding patterns and 3 dysfunctional feeding. One infant refused the assessment feed. There were no significant differences between the UCLP No PSO and UCLP PSO groups (p=0.33). Within the ICP No PSO group, 1 infant was rated as having a normal feeding pattern, 5 as disorganised and 1 as dysfunctional. One infant refused the assessment feed. All infants in the ICP PSO group were rated as disorganised feeders, except for 2 who refused the assessment feed. There were no significant differences between the ICP No PSO and ICP PSO groups (p=1.00).
Table 29: NOMAS ratings at neonatal assessment

<table>
<thead>
<tr>
<th>NOMAS rating</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO: n=16</td>
<td>PSO: N=17</td>
</tr>
<tr>
<td>Normal</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>Disorganised</td>
<td>7</td>
<td>12</td>
</tr>
<tr>
<td>Dysfunctional</td>
<td>6</td>
<td>3</td>
</tr>
<tr>
<td>Refused or unrateable</td>
<td>2</td>
<td>2</td>
</tr>
</tbody>
</table>

Similar results were obtained at the 3 month assessment (Table 30).

No infants from any group were rated as having normal feeding patterns. Cooperation was poorer at 3 months of age with several infants from each group giving up or refusing the feed.

Within the UCLP No PSO group 5 infants were rated as having disorganised feeding patterns and 7 as having dysfunctional feeding patterns. Similarly within the UCLP PSO group 6 infants were rated as disorganised feeders and 7 as dysfunctional. Again there was no significant difference between the severity of ratings between these groups (p=1.00). Infants in the ICP No PSO and PSO groups tended to display more disorganised (ICP No PSO n=4, ICP PSO n=5) than dysfunctional feeding patterns (ICP No PSO n=1, ICP PSO n=2. Again there was no significant difference in the ratings between the groups (p=0.65).
Table 30: NOMAS ratings at 3 month assessment point

In order to evaluate whether the infant’s feeding performance was affected by the feeding bottle used, feeding was also rated during a routine feed. There was some disagreement between NOMAS ratings using the infants’ routine soft bottles and the NOMAS ratings using the GOSMIF equipment with a standard rigid bottle (Tables 31 to 39).

For all four groups combined at the neonatal assessment 54% agreement was achieved between NOMAS ratings on GOSMIF and routine soft bottle-feeds. Kappa coefficients were poor (0.295) (Table 31).
Table 31: Comparison of NOMAS scores for GOSMIF and routine adapted bottle-feeds at neonatal assessment

If the individual groups are examined a similar picture is seen. Tables 32 to 40 summarise the findings for each group. A colour-coding system is introduced to facilitate the reader's interpretation. The UCLP No PSO group is colour coded in red, the UCLP PSO group in green, the ICP No PSO group in blue and the ICP PSO group in pink.

**Neonatal assessment**

At the neonatal assessment, within the UCLP No PSO group, 10 of the 16 infants were given an identical rating for oral motor skills during both the GOSMIF feed and their routine adaptive bottle-feeds (Table 32). Two infants showed “better” oral motor skills during their routine soft bottle feed. In contrast 2 infants showed “better” oral motor skills during the GOSMIF feed. Two studies could not be rated because in one case a video was lost (copied over) and in the other a poor view prevented rating. Kappa calculations revealed moderate agreement (0.458).
Within the UCLP PSO group at the neonatal data collection point (Table 33), 7 infants were rated identically on both their routine soft bottle and GOSMIF feed. Three infants were rated “better” on their routine soft bottle feed than the GOSMIF feed. Conversely six infants were rated “better” during the GOSMIF feed. One infant accepted the routine soft bottle-feed but refused the GOSMIF feed. Since none of the GOSMIF feeds were rated as normal, it was not possible to calculate a Kappa coefficient.

Within the ICP No PSO group (Table 34), 4 infants were rated identically in both feed situations. One infant was rated with “better” oral motor skills during the GOSMIF feed. Two infants were rated with “better” oral motor skills during the routine soft bottle feed. One infant refused both feeds. Kappa calculations showed moderate reliability (0.4).

Four out of the 7 infants in the ICP PSO group (Table 35) achieved identical oral motor ratings on both the routine soft bottle-feed and GOSMIF feed. One infant achieved a “better” rating during the GOSMIF feed and another the reverse, i.e. a better rating during the routine soft bottle-feed. Two infants refused the GOSMIF feed. Kappa coefficients were not estimable, as GOSMIF assessments were never rated as normal.
<table>
<thead>
<tr>
<th>NOMAS NORMAL (SOFT) BOTTLE</th>
<th>NOMAS NORMAL (SOFT) BOTTLE</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Normal</td>
</tr>
<tr>
<td>Normal</td>
<td>1</td>
</tr>
<tr>
<td>Disorganised</td>
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<tr>
<td>Dysfunctional</td>
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<tr>
<td>Refused or unrateable</td>
<td>0</td>
</tr>
<tr>
<td>Total</td>
<td>2</td>
</tr>
</tbody>
</table>

Table 32: Comparisons of NOMAS ratings with routine adaptive bottle vs. GOSMIF for UCLP No PSO, at neonatal assessment

<table>
<thead>
<tr>
<th>NOMAS NORMAL (SOFT) BOTTLE</th>
<th>NOMAS NORMAL (SOFT) BOTTLE</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Normal</td>
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<td>Normal</td>
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<tr>
<td>Refused or unrateable</td>
<td>0</td>
</tr>
<tr>
<td>Total</td>
<td>5</td>
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</tbody>
</table>

Table 33: Comparisons of NOMAS ratings with routine adaptive bottle vs. GOSMIF for UCLP PSO, at neonatal assessment
### Table 34: Comparisons of NOMAS ratings with routine adaptive bottle vs GOSMIF for ICP No PSO, at neonatal assessment

<table>
<thead>
<tr>
<th>NOMAS GOSMIF</th>
<th>Normal</th>
<th>Disorganised</th>
<th>Dysfunctional</th>
<th>Refused or unrateable</th>
<th>Total</th>
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<td>0</td>
<td>5</td>
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<tr>
<td>Dysfunctional</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>Refused or unrateable</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>1</td>
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<tr>
<td>Total</td>
<td>1</td>
<td>4</td>
<td>2</td>
<td>1</td>
<td>8</td>
</tr>
</tbody>
</table>

### Table 35: Comparisons of NOMAS ratings with routine adaptive bottle vs GOSMIF for ICP PSO, at neonatal assessment

<table>
<thead>
<tr>
<th>NOMAS GOSMIF</th>
<th>Normal</th>
<th>Disorganised</th>
<th>Dysfunctional</th>
<th>Refused or unrateable</th>
<th>Total</th>
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<tbody>
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<tr>
<td>Disorganised</td>
<td>1</td>
<td>4</td>
<td>1</td>
<td>0</td>
<td>6</td>
</tr>
<tr>
<td>Dysfunctional</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Refused or unrateable</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>Total</td>
<td>1</td>
<td>6</td>
<td>1</td>
<td>0</td>
<td>8</td>
</tr>
</tbody>
</table>
3 month assessment

At 3 months of age 81.58% (n=31) of the infants (Table 36) were feeding similarly with both their routine soft bottles and the GOSMIF bottle. Percent agreement was 84%. Kappa coefficients were not estimable, as GOSMIF assessments were never rated as normal.

<table>
<thead>
<tr>
<th>NOMAS GOSMIF</th>
<th>NOMAS NORMAL (SOFT) BOTTLE</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Normal</td>
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<tr>
<td>Normal</td>
<td>0</td>
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<tr>
<td>Disorganised</td>
<td>3</td>
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<tr>
<td>Dysfunctional</td>
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</tr>
<tr>
<td>Refused or unrateable</td>
<td>1</td>
</tr>
<tr>
<td>Total</td>
<td>4</td>
</tr>
</tbody>
</table>

Table 36: Comparison of NOMAS scores for GOSMIF and routine adapted bottle feeds at 3 month assessment

At the 3 month assessment, similar results were found for the UCLP No PSO group (Table 37). Twelve infants were rated identically on both the routine feed and the GOSMIF feed. Three infants did not co-operate for either feed at this assessment and the remaining infant was being fed via nasogastric tube. Kappa coefficients revealed good agreement between ratings on GOSMIF and normal feeds was good (1.0)
At 3 months of age (Table 38), 10 infants in the UCLP PSO group achieved identical ratings on both routine soft bottle and GOSMIF feeds. One infant showed “better” oral motor skills during the routine soft bottle-feed and another refused the GOSMIF feed. The remaining 3 infants refused all feeds at assessment. Kappa coefficients were not estimable, as GOSMIF assessments were never rated as normal.

At 3 months of age all infants in the ICP No PSO group who accepted the feed (5) were rated identically on both the routine soft bottle-feed and the GOSMIF feed. One infant from this group was being fed by nasogastric tube and 2 infants refused to feed at this assessment (Table 39). Kappa calculations revealed good agreement between these ratings (1.0).

Four of the infants in the ICP PSO group achieved identical ratings in both feed situations. Two infants were rated as having “better” oral motor skills during the routine soft bottle-feed and another vice versa showing better oral motor skills during the GOSMIF feed. One infant refused either assessment (Table 40). Kappa coefficients could not be estimated, as GOSMIF assessments were never rated as normal.

Overall agreement between ratings obtained on routine soft bottle and GOSMIF feeds was good with slightly better agreement for the UCLP and ICP No PSO groups.

If the routine soft bottle results had been used for comparisons between the No PSO and PSO groups for each cleft type results would have been similar.
<table>
<thead>
<tr>
<th>NOMAS NORMAL (SOFT) BOTTLE</th>
<th>NOMAS NORMAL (SOFT) BOTTLE</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Normal</td>
</tr>
<tr>
<td>Normal</td>
<td>0</td>
</tr>
<tr>
<td>Disorganised</td>
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</tr>
<tr>
<td>Dysfunctional</td>
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</tr>
<tr>
<td>Refused or unrateable</td>
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</tr>
<tr>
<td>Total</td>
<td>3</td>
</tr>
</tbody>
</table>

Table 37: Comparisons of NOMAS ratings with routine adaptive bottle vs. GOSMIF for UCLP No PSO, at 3 months of age

<table>
<thead>
<tr>
<th>NOMAS NORMAL (SOFT) BOTTLE</th>
<th>NOMAS NORMAL (SOFT) BOTTLE</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
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</tr>
<tr>
<td>Normal</td>
<td>0</td>
</tr>
<tr>
<td>Disorganised</td>
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</tr>
<tr>
<td>Dysfunctional</td>
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<tr>
<td>Refused or unrateable</td>
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</tr>
<tr>
<td>Total</td>
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</table>

Table 38: Comparisons of NOMAS ratings with routine adaptive bottle vs. GOSMIF for UCLP PSO, at 3 months of age
Table 39: Comparisons of NOMAS ratings with routine adaptive bottle vs. GOSMIF for ICP No PSO, at 3 months of age

<table>
<thead>
<tr>
<th>NOMAS GOSMIF</th>
<th>NOMAS NORMAL (SOFT) BOTTLE</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
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<td>Normal</td>
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<tr>
<td>Disorganised</td>
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<td>Dysfunctional</td>
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<tr>
<td>Refused or unrateable</td>
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</tr>
<tr>
<td>Total</td>
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</tr>
</tbody>
</table>

Table 40: Comparisons of NOMAS ratings with routine adaptive bottle vs. GOSMIF for ICP PSO, at 3 months of age

<table>
<thead>
<tr>
<th>NOMAS GOSMIF</th>
<th>NOMAS NORMAL (SOFT) BOTTLE</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
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<tr>
<td>Normal</td>
<td>0</td>
</tr>
<tr>
<td>Disorganised</td>
<td>1</td>
</tr>
<tr>
<td>Dysfunctional</td>
<td>0</td>
</tr>
<tr>
<td>Refused or unrateable</td>
<td>0</td>
</tr>
<tr>
<td>Total</td>
<td>1</td>
</tr>
</tbody>
</table>
Schedule for oral motor assessment (SOMA)

The SOMA studies were rated by a dysphagia trained speech and language therapist who was not involved in the trial, and who was trained to rate SOMA studies.

6 month assessment

The SOMA has not been standardised on infants of 6 months of age and therefore for the purpose of this analysis raw scores are used rather than the authors overall rating of normal or abnormal oral motor dysfunction (Reilly et al., 1995; Skuse et al., 1995)(personal communication Reilly, 1999) (Figures 59-66).

At 6 months of age, ratings were made with a bottle-feed and puree. Although it is recommended by the authors that the assessment is video-recorded this was in practice difficult. In 5 cases the view required for scoring bottle feeds using the SOMA was obscured by the bottle.

Although sample sizes are small, the ratings obtained (raw scores) are remarkably similar between No PSO and PSO groups. Differences were non-significant using Mann Whitney U tests (Bottle-feeding: UCLP groups p=0.940 and ICP groups: p=1; Puree: UCLP groups p=0.142 and ICP groups p=0.421).

The following graphs show the range and frequency of raw scores for each group (using the same colour coding as previously; UCLP No PSO, UCLP PSO, ICP No PSO, ICP PSO).
Figure 59: SOMA scores for (bottle) UCLP No PSO (6 months)

Figure 60: SOMA scores (bottle) UCLP PSO (6 months)
Figure 61: SOMA scores (bottle) ICP No PSO (6 months)

Figure 62: SOMA scores (bottle) ICP PSO (6 months)
Figure 63: SOMA scores (puree) UCLP No PSO (6 months)

Figure 64: SOMA scores (puree) UCLP PSO (6 months)
Figure 65: SOMA scores (puree) ICP No PSO group (6 months)

Figure 66: SOMA scores (puree) ICP PSO group (6 months)
12 month assessment

The SOMA has been standardised for infants of 12 months of age and therefore the recommended coding system, of normal oral motor function or oral motor dysfunction was used.

Infant compliance was a problem at the 12 month assessment. Obtaining a complete data set was further complicated by ratings being made from videos. In some cases views of the infant’s mouth were obscured by their bottle, training beaker or cup. Each infant was offered several textures. Compliance was greatest for puree and crackers. Since the oral motor skills required to eat a cracker are more complex than those required to take puree, it was decided to use this rating as the primary outcome measure for oral motor skills at this assessment.

A total of 28 infants had rateable assessments with crackers. Six of the infants were from the UCLP No PSO group and 12 from the UCLP PSO group. In the ICP groups 5 infants from each group had rateable assessments (Table 41).

All infants were categorised as having normal oral motor function with this texture. In order to investigate the raw scores obtained prior to categorisation of normal or abnormal further, raw scores were compared across the groups (Table 41). All infants achieved scores of 0 or 1 (well below the cut off score of >9 which would indicate oral motor dysfunction) and were subsequently categorised as having normal oral function. Mann Whitney tests again revealed no significant difference between the No PSO and PSO groups for each cleft type group (UCLP groups p=0.616 and ICP groups p=1).
Table 41: SOMA raw scores for cracker

<table>
<thead>
<tr>
<th></th>
<th>UCLP</th>
<th></th>
<th>ICP</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO</td>
<td>PSO</td>
<td>No PSO</td>
<td>PSO</td>
</tr>
<tr>
<td>Cracker</td>
<td>n=6</td>
<td>n=12</td>
<td>n=5</td>
<td>n=5</td>
</tr>
<tr>
<td>mean</td>
<td>0</td>
<td>0</td>
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<td>SD</td>
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</tbody>
</table>

6.10.4 GOSMIF

Overall feeding patterns

On examination of GOSMIF studies (seen in the data review window screen; Section 4.2) collected on infants with CL and/or P and regardless of PSO status, 2 distinct overall patterns were observed.

The first of these patterns (Pattern 1) showed an initial burst of continuous sucking, followed by a consistent and rhythmical sucking burst/pause pattern. This was followed by a longer continuous burst of inconsistent sucking (Figure 67). This pattern was most commonly seen in the UCLP groups.

The second pattern (Pattern 2) was characterised by an extended inconsistent sucking with a less clearly identifiable sucking burst/pause pattern. At first glance it often appeared that there were no clearly identifiable sucking bursts and it was not until the data was examined more closely that bursts were identified (Figure 68). This pattern was most common in the ICP groups.
Figure 67: GOSMIF study, pattern 1

Figure 68: GOSMIF study pattern 2
It is acknowledged that these descriptions are gross but nevertheless an interesting observation. More objective measurements were obtained following identification of rateable sucking bursts and automated analysis (Section 4.2).

**Physiological measures of feeding (GOSMIF measurements)**

The first 3 rateable sucking bursts within each infant’s study were selected for measurement. The measure for each sucking burst is represented by a dot on the following graphs. There should therefore be 3 dots aligned vertically for each infant. However, it was not always possible to measure 3 sucking bursts and in some cases there are only 1 or 2 dots, indicating that only 1 or 2 sucking bursts were measured. The previously used colour coding system (UCLP No PSO, UCLP PSO, ICP No PSO, ICP PSO) is used again together with the infant identification numbers, facilitating interpretation. The details of these measures are described in Section 4.2. T tests were used to compare the median measures for each case across PSO status groups. Separate comparisons were made for each cleft type.

**Length of sucking bursts**

At the neonatal assessment the length of sucking bursts was similar across cleft type groups regardless of PSO status (Figure 69). The majority of sucking bursts were between 2 and 20 seconds; however there were several bursts which were nearly as long as 30 seconds. This was a consistent pattern across all groups. There was no significant difference between UCLP No PSO and UCLP PSO groups or between ICP No PSO and ICP PSO groups (Table 42).

At the 3 month assessment there was a trend towards longer sucking bursts (Figure 70). The majority of sucking bursts were between 2 and 20 seconds in length, however there were several sucking bursts lasting between 20 and 50 seconds. The infants in the ICP groups used slightly longer sucking bursts,
however there is no significant difference between the No PSO and PSO groups for either UCLP or ICP (Table 42).

At the 6 month assessment the infants were using longer sucking bursts, up to 55 seconds in duration (Figure 71). While the majority of sucking bursts were still between 2 and 20 seconds there were more bursts between 20 and 55 seconds (Figure 71). Again there were no significant differences between the groups. Confidence intervals were wide particularly for the ICP groups where numbers were small (Table 42).

Figures 72 to 75 show individual trajectories within UCLP No PSO, UCLP PSO, ICP No PSO and ICP PSO groups.

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO (mean)</td>
<td>PSO (mean)</td>
</tr>
<tr>
<td>Neonatal assessment</td>
<td>9.64</td>
<td>8.75</td>
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<td>3 month assessment</td>
<td>6.76</td>
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<tr>
<td>6 month assessment</td>
<td>7.45</td>
<td>12</td>
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</table>

*Confidence intervals are for differences in the mean (PSO-No PSO)

*Table 42: Mean length of sucking bursts, p values and confidence intervals for No PSO and PSO groups for UCLP and ICP at neonatal, 3 and 6 month assessments*
Figures 69-71: Length of first 3 suckling bursts at neonatal assessment, 3 months and 6 months.
Figures 72 and 73: Changes in median length of sucking bursts for individual infants within UCLP No PSO group and UCLP PSO group.
Figures 74 and 75: Changes in median length of suckling bursts for individual infants within ICP No PSO group and ICP PSO group.
**Peak to peak intervals**

At the neonatal assessment, the majority of infants were using sucks lasting between 0.4 and 0.7 seconds (Figure 76). There was a wider range of peak to peak intervals in the ICP groups, but no significant difference between the No PSO and PSO groups for either cleft type (Table 43).

At the 3 month assessment there was no change in the peak-to-peak interval within sucking bursts, although the extreme measures were slightly longer (Figure 77). There were no significant differences between the No PSO and PSO groups for either cleft type (Table 43).

By 6 months of age, the peak-to-peak intervals for all groups were longer (Figure 78). Again there were no significant differences between the No PSO and PSO groups for either cleft type (Table 43).

Figures 79 to 82 show individual trajectories within UCLP No PSO, UCLP PSO, ICP No PSO and ICP PSO groups.

Although the intervals tend to be shorter for the UCLP PSO group at the neonatal assessment, there is no similar trend at later ages.

<table>
<thead>
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<th>Cleft Type</th>
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<th>ICP</th>
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</thead>
<tbody>
<tr>
<td></td>
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<td>Mean (mean)</td>
</tr>
<tr>
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<td>3 month assessment</td>
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<tr>
<td>6 month assessment</td>
<td>0.53</td>
<td>0.56</td>
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</table>

Table 43: Mean peak to peak intervals, p values and confidence intervals for No PSO and PSO groups for UCLP and ICP at neonatal, 3 and 6 month assessments
Figures 76 to 78: Peak to peak intervals at neonatal assessment, 3 and 6 months
Figures 79 and 80: Changes in peak to peak interval for individual infants over time, within UCLP No PSO group and UCLP PSO group.
**Figure 81 (ICP No PSO)**

**Figure 82 (ICP PSO)**

Figures 81 and 82: Changes in peak to peak interval for individual infants over time, within ICP No PSO group and ICP PSO group.
**Rate of sucking**

At the neonatal assessment, the rate of sucking for infants with UCLP varies from 70 to 150 sucks per minute (Figure 83). The range was slightly higher for infants with ICP with sucking rates as fast as 190 sucks per minute. However there was no significant difference between groups based on PSO status for either cleft type (Table 44).

Similarly at 3 months of age there was a wide range in the rate of sucking but no significant difference between the groups dependent on PSO status for either cleft type (Table 44) (Figure 84).

Again at 6 months of age there was a wide range in the rate of sucking and no significant difference between the PSO groups for either cleft type (Table 44) (Figure 85).

Figures 86 to 89 show individual trajectories within UCLP No PSO, UCLP PSO, ICP No PSO and ICP PSO groups.

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO (mean)</td>
<td>PSO (mean)</td>
</tr>
<tr>
<td>Neonatal assessment</td>
<td>102.93</td>
<td>113.84</td>
</tr>
<tr>
<td></td>
<td>23.75</td>
<td>105.58</td>
</tr>
<tr>
<td>3 month assessment</td>
<td>123.80</td>
<td>118.26</td>
</tr>
<tr>
<td></td>
<td>13.02</td>
<td>26.28</td>
</tr>
<tr>
<td>6 month assessment</td>
<td>117.56</td>
<td>108.60</td>
</tr>
<tr>
<td></td>
<td>8.88</td>
<td>41.11</td>
</tr>
</tbody>
</table>

*Confidence intervals are for differences in the mean (PSO-No PSO)

**Table 44:** Mean rate of sucking, p values and confidence intervals for No PSO and PSO groups for UCLP and ICP at neonatal, 3 and 6 month assessments
Figures 83 to 85: Rate of sucking at neonatal assessment, 3 and 6 months
Figures 86 and 87: Changes in rate of sucking over time, for individual infants, within UCLP No PSO group UCLP PSO group.
Figures 88 and 89: Changes in rate of sucking over time, for individual infants, within ICP No PSO group and ICP PSO group.
Suck swallow ratio

As with all the physiological measures there was a wide range of suck swallow ratios (Figures 90 to 92). There was little change over time for this measure (Table 45). There were no significant differences between No PSO and PSO groups for either cleft type at any assessment point (Table 45).

Figures 93 to 96 show individual trajectories within UCLP No PSO, UCLP PSO, ICP No PSO and ICP PSO groups.

<table>
<thead>
<tr>
<th></th>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td>No PSO (mean)</td>
<td>PSO (mean)</td>
</tr>
<tr>
<td>Neonatal assessment</td>
<td></td>
<td>2.93</td>
<td>3.59</td>
</tr>
<tr>
<td>3 month assessment</td>
<td></td>
<td>2.31</td>
<td>2.57</td>
</tr>
<tr>
<td>6 month assessment</td>
<td></td>
<td>1.91</td>
<td>2.13</td>
</tr>
</tbody>
</table>

*Confidence intervals are for differences in the mean (PSO-No PSO)

Table 45: Mean suck swallow ratios, p values and confidence intervals for No PSO and PSO groups for UCLP and ICP at neonatal, 3 and 6 month assessments.
Figures 90 to 92: Suck swallow ratios at neonatal assessment, 3 and 6 months
Figures 93 and 94: Changes in suck swallow ratios over time, for individual infants, within UCLP No PSO group and UCLP PSO group.
Figures 95 and 96: Changes in suck-swallow ratios over time, for individual infants, within ICP No PSO group and ICP PSO group.
Percent pressure generation above baseline pressure in feeding bottle

The infants in this cohort generated a high proportion of positive pressures during bottle-feeding, although there was great variability (Table 46) (Figures 97 to 99). Again there were no significant differences or consistent patterns between the No PSO and PSO groups in either the UCLP or ICP groups with the exception of UCLP groups at the 3 month assessment where the % of pressure generation above the baseline was greater in the No PSO group (Table 46).

Figures 100 to 103 show individual trajectories within UCLP No PSO, UCLP PSO, ICP No PSO and ICP PSO groups.

<table>
<thead>
<tr>
<th>Cleft Type</th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO (mean)</td>
<td>PSO (mean)</td>
</tr>
<tr>
<td>Neonatal assessment</td>
<td>80.70</td>
<td>0.34</td>
</tr>
<tr>
<td>3 month assessment</td>
<td>84.20</td>
<td>63.55</td>
</tr>
<tr>
<td>6 month assessment</td>
<td>81.06</td>
<td>83.68</td>
</tr>
</tbody>
</table>

*Confidence intervals are for differences in the mean (PSO-No PSO)

Table 46: Mean % positive pressure generated above baseline, p values and confidence intervals for No PSO and PSO groups for UCLP and ICP at neonatal, 3 and 6 month assessments
Figures 97 to 99: % pressures generated above baseline pressure in bottle at neonatal assessment, 3 and 6 months
Figures 100 and 101: Changes in % pressure generated above baseline pressure in bottle, over time, for individual infants, within UCLP No PSO group and UCLP PSO group.
Figures 102 and 103: Changes in % pressure generated above baseline pressure in bottle, over time, for individual infants, within ICP No PSO group and ICP PSO group.
6.10.5 Videofluoroscopy findings

Videofluoroscopic assessment of swallowing was carried out on 20 consecutive cases (UCLP No PSO, 5 infants; UCLP PSO 8 infants; ICP No PSO, 4 infants and ICP PSO 4 infants) and was rated independently by the Researcher and another Speech and Language Therapist experienced in the videofluoroscopic assessment of paediatric dysphagia.

At the oral stage, all infants regardless of PSO status, demonstrated abnormal tongue configuration during sucking, with uncoordinated stripping of the tongue, and reduced compression of the teat. Bolus formation was abnormal in all cases but one, with milk pooling in the lateral sulci and in the floor of the mouth. Milk entered the nasopharynx in 16 of the 20 infants; no infants however demonstrated frank nasal regurgitation with milk leaking from the nose.

At the pharyngeal stage, 19 infants triggered the swallow after the bolus had left the valleculae or when the bolus was in the piriform sinuses. Fourteen of the 20 infants demonstrated adequate airway protection with no material entering the larynx. However, 3 infants displayed silent aspiration or material entering the airway, passing below the vocal folds with no reflexive cough or attempt to eject the material (rated as 7 on the Rosenbek scale (Rosenbek et al., 1996)). This aspiration occurred prior to triggering of the swallow and was associated with delayed triggering of the swallow. Another 3 infants displayed reduced airway protection with material entering the airway, contacting the vocal folds and being ejected from the airway (rated as 3 on the Rosenbek scale (Rosenbek et al., 1996)).

Pharyngeal residue was evident in about half of the infants (11 infants). All infants cleared this spontaneously with subsequent swallows.

There was no significant difference between the No PSO and PSO groups for either cleft type, for number of abnormal behaviours rated (Table 47).
<table>
<thead>
<tr>
<th>Oral and Pharyngeal behaviours/events</th>
<th>Number rated abnormal</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>UCLP (n=13)</td>
<td>ICP (n=8)</td>
</tr>
<tr>
<td></td>
<td>No PSO (n=5)</td>
<td>No PSO (n=4)</td>
</tr>
<tr>
<td>Lip closure</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Tongue configuration during sucking</td>
<td>5</td>
<td>8</td>
</tr>
<tr>
<td>Compression of teat</td>
<td>5</td>
<td>8</td>
</tr>
<tr>
<td>Bolus formation</td>
<td>5</td>
<td>7</td>
</tr>
<tr>
<td>Nasal regurgitation</td>
<td>4</td>
<td>6</td>
</tr>
<tr>
<td>Appropriate triggering of the swallow</td>
<td>5</td>
<td>7</td>
</tr>
<tr>
<td>Airway protection</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Pharyngeal clearance</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>Cricopharyngeal function</td>
<td>0</td>
<td>0</td>
</tr>
</tbody>
</table>

*All confidence intervals are for differences in the percentages (PSO - No PSO)*

Table 47: Oral and pharyngeal behaviours rated as abnormal on videofluoroscopy

### 6.10.6 Anthropometry

Changes in z scores for weight, height, head circumference and body mass index are shown in Figures 104 to 119 respectively. In order to enable identification of individual infants across groups, the same colour is used to represent the same infant within each of the 4 diagnostic and randomisation groups. These colours also relate to previous graphs where individuals are shown.
Figures 104 to 107 show the infant’s reported birth weight and subsequent measurements collected (Section 5.6.1). Height, head circumference and body mass index standard score measurements are shown for each assessment point but not birth (Figures 108 to 119).
Figures 104 and 105: "Z" scores for weight over time for UCLP No PSO group and UCLP PSO group
Figures 106 and 107: "Z" scores for weight over time for ICP No PSO group and ICP PSO group.
Figures 108 and 109: “Z” scores for height over time for UCLP No PSO group and UCLP PSO group.
Figures 110 and 111: "Z" scores for height over time for ICP No PSO group and ICP PSO group.
Figures 112 and 113: "Z" scores for head circumference UCLP No PSO group and UCLP PSO group
Figures 114 and 115: "Z" scores for head circumference ICP No PSO group and ICP PSO group.
Figures 116 and 117: "Z" scores for BMI UCLP No PSO group and UCLP PSO group
Figures 118 and 119: "Z" scores for BMI ICP No PSO group and ICP PSO group.
The patterns were remarkably similar between infants allocated to No PSO or PSO groups for all growth outcomes within each cleft type.

Comparisons were made between groups for the final assessment standardised scores of each infant. The majority of these measurements were those collected at the 12 month assessment. Some infants did not reach 12 months of age during the data collection period, and as a result the measurements used in the comparisons were those collected at the neonatal, 3 month or 6 month assessment points (Table 48).

<table>
<thead>
<tr>
<th></th>
<th>UCLP</th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO</td>
<td>PSO</td>
</tr>
<tr>
<td>Neonatal assessment</td>
<td>n=16</td>
<td>n=17</td>
</tr>
<tr>
<td>2000</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td>3 month assessment</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>6 month assessment</td>
<td>6</td>
<td>3</td>
</tr>
<tr>
<td>12 month assessment</td>
<td>7</td>
<td>13</td>
</tr>
</tbody>
</table>

*Table 48: Table showing the last assessment points used in comparisons of anthropometric measures*

There were no significant differences between No PSO and PSO groups for weight, height, head circumference or bmi for either cleft type (Table 49).

If PSO were found to have any effect on feeding efficiency and hence growth it would have been expected that infants managed with PSO would have shown higher Z scores for all the anthropometric measures. There was
however no consistent pattern. Infants with UCLP and PSO had lower average Z scores for weight, head circumferences and body mass index than those with UCLP and No PSO. The reverse was found for height (Table 49). Within the ICP groups a similarly non consistent pattern was found with those infants managed with PSO having higher average Z scores for weight, head circumference and bmi but not height (Table 49). None of the differences were statistically significant. However confidence intervals were wide. Therefore it is not possible to discount differences of potential clinical significance.

Comparisons were also made for all infants who reached 12 months of age during the data collection period. Fewer infants reached this point (UCLP No PSO n=7, UCLP PSO n=13, ICP No PSO n=4 and ICP PSO n=7). Infants in the UCLP PSO group had lower average Z scores for all the anthropometric measures than those in the UCLP No PSO group. The reverse was found for infants with ICP. Again all differences were not statistically significant and confidence intervals even wider than at last assessment due to the further reduction in numbers. Therefore, although the trends suggest that PSO has not lead to an improvement in anthropometric measures, clinically important differences cannot be completely discounted.
<table>
<thead>
<tr>
<th></th>
<th>UCLP</th>
<th></th>
<th>ICP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No PSO (mean)</td>
<td>PSO (mean)</td>
<td>p values</td>
</tr>
<tr>
<td>Weight</td>
<td>-0.08</td>
<td>-0.16</td>
<td>-0.08</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Height</td>
<td>0.21</td>
<td>0.35</td>
<td>0.14</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Head circumference</td>
<td>-0.42</td>
<td>-0.74</td>
<td>0.43</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Body Mass Index</td>
<td>-0.18</td>
<td>-0.55</td>
<td>0.38</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

*All confidence intervals are for differences in the percentages (PSO – No PSO)*

Table 49: Means, p values and confidence intervals for differences between UCLP (No PSO and PSO) and ICP (No PSO and PSO) for weight, height, head circumference and body mass index z scores at last assessment point
<table>
<thead>
<tr>
<th>Body Measurement</th>
<th>UCLP (No PSO n=7 mean)</th>
<th>PSO (n=13 mean)</th>
<th>p values</th>
<th>Average difference</th>
<th>ci</th>
<th>ICP (No PSO n=4 mean)</th>
<th>PSO (n=7 mean)</th>
<th>p values</th>
<th>Average difference</th>
<th>ci</th>
</tr>
</thead>
<tbody>
<tr>
<td>Weight</td>
<td>0.50</td>
<td>0.08</td>
<td>0.51</td>
<td>-0.43</td>
<td>0.91</td>
<td>-0.70</td>
<td>-0.23</td>
<td>0.63</td>
<td>0.47</td>
<td>2.58</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>-1.76 to</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>-1.64 to</td>
<td></td>
</tr>
<tr>
<td>Height</td>
<td>0.81</td>
<td>0.53</td>
<td>0.63</td>
<td>-0.28</td>
<td>0.90</td>
<td>0.67</td>
<td>-0.32</td>
<td>0.28</td>
<td>-0.98</td>
<td>0.97</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>-1.46 to</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>-2.94 to</td>
<td></td>
</tr>
<tr>
<td>Head circumference</td>
<td>-0.38</td>
<td>-0.68</td>
<td>0.61</td>
<td>-0.31</td>
<td>0.92</td>
<td>-1.64</td>
<td>-0.57</td>
<td>0.28</td>
<td>1.08</td>
<td>3.17</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
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<td>-1.54 to</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>-1.03 to</td>
<td></td>
</tr>
<tr>
<td>Body Mass Index</td>
<td>0.06</td>
<td>-0.39</td>
<td>0.49</td>
<td>-0.45</td>
<td>0.88</td>
<td>-2.06</td>
<td>-0.77</td>
<td>0.16</td>
<td>1.98</td>
<td>4.91</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>-1.78 to</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>-0.95 to</td>
<td></td>
</tr>
</tbody>
</table>

*All confidence intervals are for differences in the percentages (PSO - No PSO)*

Table 50: Means, p values and confidence intervals for differences between UCLP (No PSO and PSO) and ICP (No PSO and PSO) for weight, height, head circumference and body mass index z scores at 12 month assessment point
6.11 Per protocol analyses

Compliance within the PSO groups was relatively poor, hence further analyses of the primary outcome measures were carried out, accounting for compliance. Those who comply may be a biased group and any difference shown between those infants who complied and the control group can less readily be applied to PSO. However some estimated measure of partial efficacy may be obtained. Two comparisons were made. Firstly infants allocated to PSO groups who complied completely (i.e. wore their PSO for 23-24 hours a day until 6 months of age) were compared with those infants allocated to the No PSO groups. Secondly infants allocated to PSO and who complied completely were compared with infants allocated to PSO and who had at least partial compliance wearing the plate for a part of the prescribed 6 month period.

6.11.1 Oral motor skills

As all infants were found to have normal oral motor skills at 12 months of age, further analysis was not warranted.

6.11.2 Anthropometry

Figures 121, 123, 125, 127, 129, 131 133, and 135 show the anthropometric measures along with the degree of compliance for infants allocated to the PSO groups. Cases where infants complied completely are shown in aqua, cases where the infants were wearing their plates at the 3 month assessment but had abandoned wearing it by the 6 month assessment are shown in orange and those who were not wearing their plate at the 3 month assessment are shown in green. One infant had some initial problems with tolerating the plate for continuously but was wearing it by 2 weeks of age (shown in pink), and another case never tolerated the plate for more than a few hours and abandoned use before 1 month of age (shown in purple). For ease of comparison the graphs showing the No PSO groups (shown in black) are
shown side by side (Figures 120, 122, 124, 126, 128, 130, 132 and 134). The graphic representations suggest that there is no significant difference between patterns over time, related to the degree of compliance.

There was no significant difference between infants allocated to PSO and who complied fully and those allocated to No PSO groups for any anthropometric measures at the final assessment point for each infant (UCLP: Weight: p= 0.55, mean difference -0.46, ci -2.07 to 1.15, Height: p= 0.65, mean difference -0.30, ci -1.71 to 1.10, Head circumference: p= 0.53, mean difference -0.47, ci -2.03 to 1.01, BMI: p=0.69, mean difference -0.30, ci -1.86 to 1.27. ICP: Weight: p=0.87, mean difference 0.18, ci -2.32 to 2.68, Height: p= 0.23, mean difference -1.29, ci -3.58 to 1.00, Head circumference: p=0.34, mean difference 1.15, ci -1.50 to 3.80, BMI: p= 0.35, mean difference 1.50, ci -2.00 to 5.00).

It therefore appears that lack of compliance had no significant effect on the primary outcome measures of oral motor skills or anthropometry.
Figure 120 (UCLP No PSO)  

Figure 121 (UCLP PSO variable compliance)  

Legend
- * complete compliance
- * abandoned use after 3 but before 6 months
- * initial problems but wearing by 2 weeks
- * never wore consistently
- * no PSO

Figures 120 and 121: "Z" scores for weight in UCLP no PSO and UCLP PSO group taking into consideration PSO compliance group
Figures 122 and 123: "Z" scores for height in UCLP no PSO group and UCLP PSO group taking into consideration PSO compliance.
Figures 124 and 125: "Z" scores for head circumference in UCLP no PSO and UCLP PSO group taking into consideration PSO compliance.
Figures 126 and 127: "Z" scores for BMI in UCLP no PSO group and UCLP PSO group taking into consideration PSO compliance.
Figures 128 and 129: "Z" scores for weight in ICP no PSO group and ICP PSO group taking into consideration PSO compliance.
Figures 130 and 131: "Z" scores for height in ICP no PSO group and ICP PSO group taking into consideration PSO compliance.
Figures 132 and 133: “Z” scores for head circumference in ICP no PSO group and ICP PSO group taking into consideration PSO compliance.
Figures 134 and 135: "Z" scores for bmi in ICP no PSO group ICP PSO group taking into consideration PSO compliance
6.12 Summary

Unfortunately the small numbers recruited limit the power of the results of this trial, particularly at the later assessments (e.g. 12 months) when numbers were smaller than at the earlier assessments (UCLP No PSO n=8, UCLP PSO n=13, ICP No PSO n=6 and ICP PSO n=7).

It was initially calculated that in order to obtain 80% power at the 5% significance level for the continuous variable, anthropometry, 25 infants would be required in each group. However even in the larger UCLP groups these numbers were not achieved at the primary outcome measure point of 12 months (UCLP No PSO n=8 and UCLP PSO n=13). Figure 53 was introduced in Section 5.11 to show the differences detectable at 5% significance level, with power 80%, 90% and 85% for different sample sizes. Figure 136 a modified replication of Figure 53, shows that 50 infants distributed evenly between 2 groups, would have shown a difference of 0.8 SD at 80% power (shown in black). When the number of infants is reduced to 13 in each group, the difference detected at 80% power is 1.1 SD (shown in blue). For two groups of 7 the difference detected is only 1.5 SD (shown in red). Hence the power to detect an 0.8 SD difference was less than the anticipated 80% and differences of this magnitude may not have been identified within the trial undertaken. Because of reduced sample size, the trial had sufficient power to detect only larger shifts in mean outcomes between the No PSO and PSO groups.
The majority of results obtained are remarkably similar across the four groups, although confidence intervals were wide for some measures due to the unexpectedly small recruited numbers.

While there were some small differences in the background variables including demographics, gestational age, birth-weight, risk factors for feeding difficulties or developmental delay, between the UCLP and ICP groups, there were no major differences between the No PSO and PSO groups for each cleft type.

Similarly, there were no differences across the No PSO and PSO groups for each cleft type, for later risk factors of developmental delay or feeding difficulties including family stress, general health and general development.

---

*Figure 136: Graph showing the standardised difference in means that can be detected with 80, 90 and 95% power at 5% significance with specified numbers per group*
There were no significant differences between the No PSO and PSO groups for either cleft type, for the primary outcomes measures of oral motor skills. It might be expected that if PSO had a significant impact on feeding this would translate into differences in anthropometric measures. There were however no consistent improvements among the PSO groups for either cleft type.

Similarly there were no significant differences in the secondary outcome measures of parental report about feeding methods, acceptance of age appropriate solids, time taken to complete feeds or feeding characteristics at 12 months of age. There were however occasional trends observed. There was no significant difference in the feeding methods used or the number of adaptations to teats required for bottle feeding. All infants accepted age appropriate solids, however there was a trend towards a greater proportion of infants with UCLP and PSO accepting mashed and chopped table foods. This was not the case for infants with ICP and PSO. There was a trend towards infants from UCLP PSO group to completing bottle feeds in a approximately 3 minutes less time than those in the UCLP No PSO group. A similar but stronger tendency was evident within the ICP groups, with those infants managed with PSO feeding completing a bottle feed in approximately 15 minutes less time than those managed without PSO. An interesting phenomenon was evident with the time taken to complete solid feeds at 12 months. As might be expected given the above results the time taken for infants with ICP and PSO to complete a solid feeds was approximately 6 minutes shorter than those managed without PSO. In contrast infants in the UCLP PSO group took approximately 5 minutes longer than the UCLP No PSO group to complete solid feeds. None of these differences were however significant. There were no significant differences or consistent patterns between infants managed with or without PSO irrespective of cleft type for the feeding characteristics including nasal regurgitation and coughing and choking. Consistent with the majority of these reported findings, there were
no significant differences between the No PSO and PSO groups for oral motor skill assessment.

Similarly at 6 months of age there were some differences between the PSO groups for both cleft types, for the secondary outcome measures of parental report about feeding methods and feeding characteristics. If PSO do allow infants to develop better oral motor skills, acceptance of a wider range of more complex textures might be expected in the PSO groups. While there was a trend towards an increased acceptance of thicker solids, including 7 month solids and lumpy puree amongst infants managed with PSO for both cleft types, at 6 months of age this difference was not significant. However within the UCLP PSO group there were significantly more infants who accepted thick puree. The reported length of time taken to complete bottle feeds tended to be shorter for infants managed with PSO. This trend was not however statistically significant. There were no significant differences between the No PSO and PSO groups for either cleft type in feeding characteristics of coughing and choking or gurgley breath sounds during or after feeding. While there was no significant difference in the number of infants displaying nasal regurgitation it did appear that the frequency of these episodes was greater for infants managed without PSO. Consistent with most of the parent reports there were no significant differences between the No PSO and PSO groups for either cleft type, for the physiological measures of sucking as assessed with the GOSMIF or oral motor skills assessed with the SOMA.

One might have expected that if these trends had clinical implications differences in growth would have been evident. This was not however the case.

When analysis was repeated taking into account compliance, whether comparing infants who complied completely with PSO and those not randomised to the PSO groups or between infants who complied partially and those not randomised to PSO groups, there were still no significant
differences in the primary outcome measures of oral motor skills and anthropometric measures at 12 months of age.
Chapter 7

DISCUSSION AND CONCLUSIONS

This chapter focuses on three main areas. The first section discusses the results of the trial with reference to the questions posed. The second discusses factors that may have affected these results including subjects recruited, the trial methodology, selection and application of assessment tools, and adherence to the specified protocol. The third discusses areas of further research and the conclusions drawn.

7.1 Results

This trial set out to answer the following primary research questions.

a. Is there a significant difference in the oral motor skills of infants (with ICP or UCLP) managed with or without PSO post palate repair at 12 months of age?

b. Is there a significant difference in general body growth (weight, length, head circumference and body mass index) of infants (with ICP or UCLP) managed with or without PSO after palate repair at 12 months of age?

Oral motor skills were assessed at 12 months of age using the SOMA. All infants were rated as having normal oral motor skills at this age, irrespective of PSO grouping. Most of the literature about feeding in infants with CL and/or P focuses on feeding prior to surgical intervention. This literature suggests that feeding difficulties are common and are due to the infant’s inability to generate the sucking pressures required to transfer milk from the
bottle or teat. Although there are no formal reports of feeding after palate repair, it is generally accepted that the feeding problems resolve. The finding of this study that all infants had normal oral motor skills at 12 months of age, 6 months following palate repair, adds support to this belief. It has been suggested that PSO improve the oral motor movements for speech (Stuffins, 1983; Gnoinski, 1990; Gruber, 1990; Konst et al., 1999; Konst, 2002) and feeding (Lifton, 1956; Williams et al., 1968; Jones et al., 1982; Balluff and Udin, 1986; Goldberg et al., 1988; Osuji, 1995) by decreasing the development of abnormal compensatory oral motor movements. Konst et al (1999; 2002) recently completed a randomised control trial (Dutchcleft) investigating the effect of PSO on early speech development in 49 children with UCLP and showed that the use of PSO does improve or normalise the acquisition of early speech sounds and results in better speech and language development at 2 ½ and 3 years of age. They suggested that this might be the result of more normal sensori-motor patterns being laid down during infancy when the PSO was worn (Stuffins, 1983; Folkins, 1985; Konst et al., 1999; Konst, 2002). While it would be of interest to compare the findings of the Dutchcleft study with those of this trial there is a fundamental difference between the two studies relating to the timing of surgery and length of use of PSO (Kuijpers-Jagtman and Prahl-Andersen, 1997; Konst et al., 1999; Konst et al., 2000; Kuijpers-Jagtman et al., 2001; Konst, 2002). In this trial palate repair (both soft and hard) was carried out at 6 months of age, whereas in the Dutchcleft trial a two-stage repair was used with repair of the soft palate undertaken at 12 months of age and the hard palate not until 9 years of age. Infants recruited to both trials wore PSO from birth. In this trial, PSO were worn for 6 months, whereas in the Dutchcleft trial PSO were worn for 12 months. Hence infants in the Dutchcleft trial had an unrepaird palate during the critical period for babbling development between 6-12 months of age (Russell and Grunwell, 1993; Harding, 1993; Russell and Harding, 2000) and the use of PSO is possibly more critical for speech development during this
time. To compare directly the Dutch cleft population with that of this trial is therefore inappropriate.

The second question addressed was whether there were differences in the anthropometric measures of weight, height, head circumference and body mass index between the groups of infants managed with and without PSO at 12 months of age. If PSO had an effect on feeding efficiency then differences in growth measurements would have been expected. There were, however, no significant differences in growth, height, head circumference or the overall measure of body mass index. Although some of the confidence intervals were wide, indicating imprecision of the study results due to limited numbers of infants, there were no consistent trends.

It is widely accepted that infants with CL and/or P may be slow to gain weight while their feeding regime is being established but return to, or catch-up to approximately their birth centile by the age of 2 years (Ranalli and Mazaheri, 1975; Avedian and Rubery, 1980; Lee et al., 1997). In this trial, the majority of infants in all groups showed catch-up growth for weight by 12 months of age, returning to, or surpassing their birth-weight Z score. There were occasional differences between the Z scores for weight, height, head circumference or body mass index for individual infants however these differences were less than 1 SD and were not considered to be clinically important.

The World Health Organisation (WHO) recommend that weight and height are collected allowing weight-for-height measures or bmi to be calculated (1995). It is suggested that if an infant shows a drop in Z scores for both weight and length, long term malnutrition or poor health is indicated, whereas a drop in weight but not height suggests recent or continuing weight loss. The WHO (1995) suggests that changes of two Z scores for weight-for-height or body mass index are significant and require intervention. Within this trial
there were no differences greater than one Z score for body mass index, and therefore would not be considered clinically significant.

Four secondary research questions were addressed.

c. Is there a significant difference in the oral motor skills of infants with ICP or UCLP managed with or without PSO, prior to surgical repair of the palate at 3 and 6 months of age?

d. Are there any significant differences in physiological measures of feeding in infants with ICP or UCLP managed with or without PSO, prior to surgical repair of the palate at 3 and 6 months of age?

e. Are there any significant differences in reported feeding characteristics (length of feed, nasal regurgitation, number of adaptations of feeding equipment) of infants with ICP or UCLP managed with or without PSO, prior to surgical repair of the palate at 3 and 6 months of age?

f. Is there any significant difference in general body growth (weight, length, head circumference and body mass index) of infants with ICP or UCLP managed with or without PSO, prior to surgical repair of the palate at 3 and 6 months of age?

In answer to the first of these questions, the results showed no significant differences in the oral motor skills of the infants whether managed with or without PSO for either cleft type at either age.

Oral motor skills were assessed at 3 months of age with the NOMAS and at 6 months of age with the Soma. In contrast to Clarren et al’s (1987) and Brogan’s (1987) reports that the oral motor skills of infants with CL and/or P are normal, few of the infants in this trial demonstrated normal patterns at 3 or 6 months of age. The predominant abnormal features identified on the
NOMAS were inconsistent range of tongue and jaw movements, arrhythmic tongue and jaw movements, altered rate of sucking and inco-ordination of the suck swallow and breathe triad. There was also evidence of abnormal tongue movements on the videofluoroscopic assessments of a subgroup of the trial cohort. All infants in the trial cohort failed to groove or cup the tongue around the teat and the range of tongue movements was reduced. Eishima (1991) interestingly also reported a lack of grooving or cupping around the teat in non-cleft infants when they were fed with a fast flow teat. This might suggest that where flow rate is increased such as with the use of soft bottles, and less effort is required on the part of the infant to transfer the milk from the teat or nipple, stabilisation of the teat by cupping of the tongue is less necessary. Interestingly, some of the earliest descriptions of feeding problems in infants with CL and/or P allude to the findings of this trial in that infants with CL and/or P fail to compress the teat or nipple with their tongue, and instead use a biting or chomping action on the teat. For example, Zickefoose (1957) described infants “chewing the nipple/teat” in an attempt to obtain milk from a bottle and Tisza et al (1962) described infants as “milking and squeezing the teat”. Clarren et al (1987) reported a similar pattern for infants with Pierre Robin Sequence but not ICP and UCLP.

Osujii (1995) hypothesised that if an opposing surface, in the form of PSO was provided, infants with CL and/or P would produce more normal tongue movements, including compression of the teat. The findings of this trial do not support this hypothesis as all infants failed to compress the teat with the tongue and so PSO did not offer any advantage.

Tongue movements also contribute to the formation and posterior transfer of in the oral cavity. While Brogan et al (1987) suggested that tongue movements for this purpose are normal in infants with CL and/or P, the findings of this trial suggest otherwise. All infants, regardless of cleft type or PSO status, showed poor bolus formation and transfer posteriorly. As seen from the videofluoroscopic evidence, residue was frequently present under the tongue,
an indication of reduced tongue movements. The reduced range of tongue movements is difficult to explain given that anatomically the tongue is unaltered. However as discussed earlier, it is postulated that the abnormal tongue movements may have developed in utero as a result of their altered anatomy. Non-cleft infants have been found to produce sucking movements in utero (Bosma, 1986). It might be hypothesised that the tongue movements of infants with CL and/or P are reduced even in utero, with the tongue sitting in the cleft rather than actively moving for sucking and swallowing. Alternatively it might be that there is early recognition on the part of infants with CL and/or P, that there is no opposing surface for the tongue to contact during sucking and this movement pattern is therefore not established.

To address the question of whether there were significant differences in the physiological measures of bottle feeding between the groups of infants managed with or without PSO prior to palate repair, GOSMIF assessments were carried out neonatally, and at 3 and 6 months of age. It was predicted that if PSO were facilitating more efficient feeding, patterns more closely resembling those of normal infants might be found. In order to address this, comparisons of the results of the non-cleft cohort assessed in the trial were made with the CL and/or P groups at the neonatal assessment.

There were no significant differences in the physiological measures of bottle-feeding, between the groups of infants managed with or without PSO, for either cleft type before palate repair at any time point.

More specifically, the measures identified from the literature as contributing to feeding efficiency were length of sucking bursts, rate of sucking, length of individual sucks or peak to peak to peak intervals, percentage of positive pressure generated and the suck swallow ratio. All these measures are closely related to the motor components of feeding discussed above.
There was no difference between the overall patterns of feeding between infants with CL and/or P for either cleft type or PSO status suggesting that the behaviours of the cleft group were similar as a whole.

There were, however, differences between the non-cleft infants and those with CL and/or P. The non-cleft infants completed a 5-minute assessment and produced an organised and rhythmic sucking/pause pattern (Figure 137). In contrast, infants with CL and/or P, irrespective of cleft type and PSO status, used one of two patterns of sucking. In the first, the infants did not produce any identifiable sucking bursts, but used a continuous and disorganised pattern (Figure 138). Pattern 2 was characterised by the generation of a short but rhythmic sucking burst/pause pattern. This pattern was maintained for the first part of the study (generally 2 to 3 minutes), followed by a pattern of disorganisation (Figure 139). Interestingly, both patterns closely resemble those reported for premature infants (Wollf, 1969; Gewolb et al., 2001), perhaps suggesting there may be a developmental component to the sucking pattern.

Figure 137: Data review screen for normal infant GOSMIF trial
Some changes were evident between the neonatal assessment and 6 months of age in the sucking patterns of the infants with CL and/or P, irrespective of
cleft type or PSO status. A typical example is shown in Figure 140. At the neonatal assessment, this infant showed an energetic feeding pattern characterised by the generation of clearly defined sucking bursts and energetic positive-pressure generation (Figure 140 a). By 3 months of age, however, there were less defined sucking bursts and pauses, and less energetic attempts at sucking (Figure 140 b). These changes may reflect adaptive patterns developed by the infant due to the habitual use of soft bottles. It may be that as the infant gradually became familiar with his/her mother’s use of the soft bottle, he/she realised that less effort was required on his/her part. The principles of this lend support to the hypothesis put forward by Ardran et al (1951) that “if a nipple with a large hole is given repeatedly the movements of the tongue, lips, jaw and cheeks would become lazy”. Given that infants in this trial demonstrated overall passive feeding patterns, the lack of differences between the infants managed with or without PSO was not surprising.
Figure 140: GOSMIF neonatal and 3 month assessments showing differences in feeding patterns between non cleft infants and trial cohort
There were no differences between the “within sucking bursts” of infants managed with or without PSO, irrespective of cleft type. There were however some differences between the non-cleft infants and the groups of infants with CL and/or P.

Infants with CL and/or P used shorter sucking bursts (Figure 141), but their rate of sucking was significantly faster (Figure 142) and the length of individual sucks (Figure 143) was shorter than in the normal infants. The majority of CL and/or P infants sucked at rates reported to be associated with non-nutritive sucking (Wolff, 1968; Lau and Kusnierczyk, 2001). Ardran (1951), Christensen et al (1976), Eishima (1991), Mathew (1988; 1991) and Mathew and Bhatia (1989) have all shown that when the flow rate of bottle feeds is increased, the infant’s rate of sucking slows (with the length of individual sucks lengthening) and becomes more typical of the nutritive sucking pattern. Conversely, if the flow rate is decreased, the rate of sucking increases with the length of individual sucks decreasing, and is more typical of non-nutritive sucking (Ardran et al., 1958a; Christensen et al., 1976; Mathew and Bhatia, 1989; Eishima, 1991; Mathew, 1991a). So for infants with CL and/or P, where the bolus size is smaller and the flow rate slower than in non-cleft infants, it is not surprising that the rate of sucking is faster and the length of individual sucks is shorter. If there had been a difference in the sucking efficiency of infants with CL and/or P managed with or without PSO, it would have been expected that those managed with PSO would have demonstrated slower rates or sucking, resembling those of the non-cleft infants.
Figure 141: Scatter plots showing length of sucking bursts for non-cleft and all groups of infants with CL, and/or P at neonatal assessment

Figure 142: Scatter plots of rate of sucking for normal infants and all groups of infants with CL, and/or P at neonatal assessment
To transfer fluid from the breast or bottle, infants must generate a combination of positive and negative pressures. The group of non-cleft infants generated more negative pressure to facilitate this transfer. The infants with CL and/or P, whether managed with PSO or without PSO, generated a significantly higher proportion of positive pressures (Figure 144). This predominance of positive pressure generation can be attributed to the characteristic chomping or biting action on the teat. Shwenzener and Grimm (1981), Hotz (1983), Komposch (1986), Kogo et al (1997) and Trankmann (2000) hypothesised that PSO allow infants with CL and/or P to generate the negative intra-oral pressures required to transfer fluid from the breast or bottle. In contrast, Choi et al’s (1991) investigation concluded that PSO did not facilitate the production of negative intra-oral pressures. The findings of this study support those of Choi et al in that no difference in intra-oral pressures between infants managed with or without PSO were found.
The suck/swallow ratio describes the number of sucks produced prior to the triggering of a swallow during breast or bottle-feeding. In this trial infants with CL and/or P, irrespective of cleft type or PSO status, produced higher suck/swallow ratios than the non-cleft infants (Figure 145). This increase in the number of sucks required to obtain an adequate bolus is probably an indicator of an inefficient suck. This pattern remained at 3 and 6 months of age, with wide variability in the range being a predominant feature (Section 6.10.5). This was in contrast to the age-matched non-cleft infants where the suck/swallow ratio was always in the range of 1 to 3 (Vice et al., 1995; Qureshi et al., 2002).
Figure 145: Scatter plots showing the suck/swallow ratios for non-cleft and all groups of infants with CL and/or P at neonatal assessment

It is widely believed that infants with CL and/or P swallow normally (Shelton et al., 1966; Clarren et al., 1987). There are no reports suggesting otherwise. However, videofluoroscopic results confirmed that the pharyngeal stage of swallowing differed from that reported as normal in the literature. Non-cleft infants trigger their swallow at the level of the valleculae (Newman et al., 1991; Arvedson and Lefton-Greif, 1998). In contrast, the majority of infants within this study did not trigger the swallow until the bolus had reached the piriform sinuses. The reasons for this delay are unclear. Miller (1986) suggests that initiation of the swallow is dependent on sensory feedback from a number of areas including the faucial arches, uvula, soft palate and posterior tongue and pharynx. Given the abnormal anatomy and reports of altered oral stereognosis in cleft palate (Hockberg and Kabcenell, 1967; McAllister et al., 2001) the delay in triggering might be attributable to reduced sensory input. Swallowing is a highly co-ordinated and complex process and relies on the
accurate timing and co-ordination of over 20 different muscles. It seems reasonable to hypothesise that the delay observed is a result of abnormal patterns of tongue movements and inco-ordination of the oral phase that subsequently has an impact on pharyngeal stage muscle movements. If PSO did facilitate more efficient or normal feeding patterns at the oral stage there would be a subsequent improvement at the pharyngeal stage. This was found not to be the case for either cleft type.

Delay in triggering of the swallow places infants and children at increased risk of aspiration (Shapiro and Healy, 1988; Rogers et al., 1993a; Taniguchi and Moyer, 1994; Arvedson et al., 1994; Arvedson and Brodsky, 2002). Twenty percent of infants assessed with videofluoroscopy were found to aspirate silently in that the bolus entered the larynx and passed through the vocal folds into the trachea without the infant showing any signs such as coughing. It is well recognised that this phenomenon places the infant at high risk of developing chest infections and aspiration pneumonia. Silent aspiration is usually associated with neurological impairment (Mirrett et al., 1994; Taniguchi and Moyer, 1994) or reduced sensation (Arvedson, 1998), or may be developmental in nature (Leith, 2000). It has also been suggested that infants may become desensitised to chronic aspiration, thereby reducing the cough reflex (Arvedson et al., 1994; Arvedson and Brodsky, 2002). Given that the infants in this trial had not been diagnosed with neurological impairment, and yet were aspirating silently, the concept of desensitisation provides a possible explanation. It is interesting that again this characteristic is associated with reduced sensation and sensori-motor changes.

In order to answer the question as to whether there are any differences in the feeding characteristics of infants with ICP or UCLP, managed with or without PSO prior to surgical repair of the palate, parents were asked what equipment and adaptations they used in feeding their infant, the length of feeds and what feeding symptoms or difficulties their infant had.
If PSO had improved feeding, a proportion of infants managed with PSO might have breast-fed at least partially or might have taken bottle-feeds with standard bottles or with unadapted teats. In contrast to the reports of Crossman (1998), Grady (1977), Danner (1992) and Kogo et al (1997), this was found not to be so in this trial.

Exclusive breastfeeding is the recommended method of feeding newborns and is widely encouraged (World Health Organisation, 2001a; World Health Organisation, 2001b). Unfortunately, it is rarely possible for infants with CL and/or P to be breastfed exclusively, although it is reported that PSO may facilitate breastfeeding (Crossman, 1998; Hemingway, 1972; Kogo et al., 1997; Biancuzzo, 1998). Much importance is given to the advantages of breastfeeding by professionals and others such as extended family, parent groups (including the National Childbirth Trust), and the media. Many parents of infants with CL and/or P plan to breastfeed. A large number of the mothers in this trial attempted breastfeeding (UCLP No PSO n=11 69%, UCLP PSO n=11, 65%; ICP No PSO n=7, 87%, ICP PSO n=5, 62%), comparing surprisingly favourably with patterns seen in the general population (71% in England and Wales, (Hamlyn et al., 2002). A national survey of infant feeding patterns is carried out every four years, providing detailed analysis of the influence of factors such as birth order, parental education, socio-economic rating and ethnicity (Hamlyn et al., 2002). The demographic characteristics of the UK survey population are similar to this trial cohort. The items in the trial questionnaires were broadly based on those of the UK survey, thus allowing some comparisons of feeding methods and symptoms between the trial group of infants and the UK population. In the UK survey, “incidence of breast feeding” was defined as “the proportion of sampled babies who were put to the breast, even if this was on one occasion only”. While the incidence of breast-feeding in this study is similar to that in the UK survey, the duration of breastfeeding (whether exclusive or combined with bottle-feeding) was quite different. In the UK survey it was reported that 65% of infants were still breastfed at 6 weeks of age. In this trial no infants with CL and/or P,
irrespective of PSO status, were breastfed at 6 weeks of age. The average length of time mothers persisted with breast-feeding was 14 days. Parents were asked why they ceased breast-feeding in both studies. In the UK survey reasons given mostly related to the mother including insufficient milk, time consuming or tiring, painful breasts or nipples, the infant could not be fed by others, mother’s ill health, the infant rejected the breast and infants’ medical problems (Hamlyn et al., 2002). In this trial, mothers’ reasons centred on the infant, such as the inability of the infant to latch on, inability of the infant to suck, the infant rejecting the breast, and lack of certainty about the volume taken. In addition, all mothers reported that they had been informed by medical or nursing staff and also by other parents, that infants with CL and/or P are often unable to breast-feed.

All infants with CL and/or P in this trial were bottle fed at some point in time. The majority were given bottle feeds in the immediate postnatal period. However, two infants were fed initially with a cup and spoon on the recommendation of their midwife. This advice was based on the widely held and yet unconfirmed view that early introduction of bottle feeds and/or dummies contributes to nipple confusion (Walker, 1993; Neifert et al., 1995; Righard and Alade, 1997). One of these infants was fed using this method for two weeks only. The parents found this method time consuming and inconvenient and found that their infant was successful in latching on to the mother’s breast. The second infant continued for an additional week at the encouragement of the infant’s grandmother. At about 3 weeks of age the infant began fussing and refusing cup and spoon-feeds. After further discussion between the mother and researcher about the concept of nipple confusion vs. the infant’s need for nutrition and comfort associated with sucking, a standard soft bottle was introduced. The fussing behaviours ceased and the infant thrived. Another infant was showing weight loss and dehydration at a two-week medical review requested by the paediatrician. Feeding assessment confirmed that this infant did not show any sucking attempts with a teat. As the infant did not show clinical signs associated with
risk factors for aspiration, a scoop bottle was introduced. Both parents and infant managed this bottle safely: the infant was rehydrated and began to gain weight. All other bottle fed infants were fed routinely with the widely recommended soft/squeezable bottles (Shaw et al., 1999). Mothers of two infants also reported occasionally feeding their infants with standard rigid bottles, as their infants “managed the rigid bottles”. This was an interesting subgroup. As these infants were feeding without the assistance of the soft bottles it was expected that they would be more efficient feeders and perhaps demonstrate patterns with regards to pressure generation and suck swallow ratios, similar to the non cleft group. However, this was not the case. It is likely that there were no differences because their bottles had been adapted with extra-enlarged holes, which were not present in the teats used during the GOSMIF assessments.

The finding that there were no patterns of breast and/or bottle feeding across the PSO and No PSO groups for either cleft types supports the findings of the objective oral motor and feeding assessments.

Three main adaptations of teats were used. These included enlarging the pre-existing hole, using a fast flow version of the teat and/or creating additional holes in the teat. There were no differences in the number or types of adaptations made to infants’ teats. If PSO had a positive effect on feeding, fewer adaptations would have been expected in the PSO groups. It was also anticipated that the number and type of adaptations would be related to the ease of feeding measured by the time it took to complete a feed. This was not the case. The type of adaptation used was related to the Clinical Nurse Specialist’s (CNS) personal preferences. Parents of infants managed by one CNS tended to adapt teats by enlarging the existing hole and/or creating additional holes. In contrast, the other CNS had concerns about parents adapting teats as the process of doing so is not only difficult but is also not easy to replicate in all teats. She therefore recommended the use of fast-flow
teats. These adaptations produced a similar effect of increasing the flow rate of milk through the teat.

The time taken for an infant to complete a bottle feed is often used as an indication of feeding efficiency (Wolf and Glass, 1992; Arvedson and Brodsky, 2002). It is generally accepted that non-cleft infants complete their bottle feed in thirty minutes (Wolf and Glass, 1992; Bannister, 2001; Arvedson and Brodsky, 2002) and that the time taken to complete feeds shortens with age. Similarly, when infants with CL and/or P complete the prescribed amount of feed in thirty minutes they are considered to be efficient (Bannister, 2001; Nassar et al., 2001). In the non-cleft cohort studied the mean length of a bottle-feed in the first month of life was 26.5 minutes (SD 17.33, Range 10 to 90 minutes) (Section 4.5). The infants with CL and/or P took a longer the mean time to complete a feed (using a soft bottle with or without adapted teat) with an average of 35.9 minutes (SD 15.93, Range 10 to 90 minutes). The wide range in time taken to complete a feed for both groups reflects the high variability of infants’ feeding patterns. As reported in Section 6.10.2 the infants with CL and/or P, irrespective of PSO status, showed shorter feed times with increasing age in common with the literature for normal infants (McGowan et al., 1991; Medoff-Cooper, 1991; Arvedson and Brodsky, 2002). These shorter feed times may be for similar reasons to normal infants such as neuro-developmental maturity, improving oral motor skills or practice (Ardran and Kemp, 1951; Arvedson and Brodsky, 2002). For infants with CL and/or P the shorter times might also reflect the parents increasing skill with using soft bottles and the infant’s increasing ability to compensate for their anatomical abnormalities. It is likely that the shorter times found at 12 months are the result of corrected anatomy following surgery.

Several authors have claimed that PSO reduce the time taken to complete feeds (Lifton, 1956; Jones et al., 1982; Balluff and Udin, 1986; Goldberg et al., 1988; Osuji, 1995). The majority of infants in this trial completed bottle feeds
within the 30 minutes usual feeding time for non cleft infants at both 3 and 6 months of age. However there was a trend towards infants managed with PSO, in both cleft type groups, to complete their bottle feeds in a shorter time than the groups managed without PSO. Although the differences were not statistically significant at either age when taken independently, there were statistically significant trends within infants with increasing age. Infants who had been allocated to the PSO groups decreased the time taken to complete bottle feeds at neonatal, 3 and 6-month assessments significantly faster than those who were allocated to the No PSO groups. This finding might be explained by an increased ability to compensate for their feeding difficulties when PSO are in situ. It is however unlikely that this finding is clinically significant given that the majority of the infants completed feeds within 30 minutes.

At 6 months of age all infants were taking solids. As reported in Section 6.10.2, there was no difference in the mean time infants took to complete these solid feeds between the No PSO and PSO groups for either cleft type. In order to manage solids infants require more complex oral motor skills and with reduced oral motor skills longer feed times would have been expected. The finding that there was no difference in the time taken to complete solid feeds lends support to the previous finding that there was no significant difference in the oral motor skills of the trial cohort irrespective of cleft type or PSO status.

**Weaning**

All infants took age-appropriate solids at 6 and 12 months of age whether managed with or without PSO. However, at 6 months of age significantly more infants in the UCLP PSO group (87%) accepted thicker puree than in the UCLP No PSO group (28%) (95% ci for the difference 21 to 87%). A similar, although nonsignificant difference, was seen in the ICP infants (ICP No PSO 86%, ICP PSO 43%, 95% ci for the difference −16 to 83%). By 12
months of age all infants were accepting age-appropriate solids. There was however a non significant trend for more infants in the UCLP PSO group (as compared to the UCLP No PSO group) to accept mashed or chopped table foods. Tables 8 and 22 show an overall tendency for the infants with PSO to take textures earlier than those without PSO. However in the overall context of acceptance of solids this finding was not considered to be clinically significant, as all infants were accepting more orally challenging solids such as chopped table foods and soft solids.

*Nasal regurgitation*

It has been suggested that PSO reduce nasal regurgitation (Huddart and Ziberman, 1977; Jones et al., 1982). In this study however there was no difference in the incidence or amount of nasal regurgitation reported by mothers for the No PSO and PSO groups, for either cleft type, at both 3 and 6 months of age.

In normal feeding an intact palate is considered important to prevent nasal regurgitation (Section 2.4.2). During swallowing the soft palate elevates, contacting the posterior pharyngeal wall, and prevents fluid or food from entering the nasopharynx. In this study, nasal regurgitation was minimal in both UCLP and ICP groups irrespective of PSO status. In the neonatal period nasal regurgitation was reported in only 24% of cases, occurring at most twice a day, and in trace amounts. This proportion remained unchanged at 3 months of age (23%). The distribution changed slightly at this assessment point with a greater proportion of infants with ICP demonstrating nasal regurgitation. This suggests that infants with UCLP were better able to adapt to their altered anatomy and minimise the nasal regurgitation. At 6 months of age an interesting phenomenon occurred. The proportion of infants with nasal regurgitation of fluids increased markedly to approximately 50% in both the UCLP and ICP groups, irrespective of whether PSO were in situ or not. While the amount of nasal regurgitation remained trace, it occurred at every
feed. It could be expected that by 12 months of age, 6 months after palate repair, nasal regurgitation would have settled down. Fifty percent of infants with UCLP and 33% of infants with ICP still showed trace nasal regurgitation of fluids every day. The low incidence of nasal regurgitation in the early months might be explained by parents’ cautious approach to feeding their infants (e.g. paying close attention to their infant, careful pacing of the feed and use of the soft bottles) and the use of strategies reported to minimise nasal regurgitation such as upright positioning. These strategies might become less strictly applied as parents’ anxieties decrease and the infants take increasing amounts of feed. Around 3 months of age infant feeding becomes less reflexive and more participative. By 6 months of age the infants are actively involved in feeding. This was evident in several cases. Several infants held and squeezed their mothers’ hands to indicate that they wanted more milk. One infant supplemented his mother’s use of the soft bottle by poking his finger into the teat or between the teat and collar resulting in a rapid flow of fluid from the teat. These patterns of less controlled feeds with the majority of mothers reporting nasal regurgitation was worse when the infants were “being greedy” or “guzzling” suggest that feed rate might influence nasal regurgitation. Where nasal regurgitation persisted at 12 months of age, all infants had small fistulae in the hard palate. In all cases the surgeon had reviewed the fistulae but no immediate further surgery was planned, in view of the minimal amount of nasal regurgitation and the surgeons preferred conservative approach to management of fistulae at this age.

All parents reported some initial and short-term problems with at least a teaspoon of solid feeds being regurgitated on a daily basis. At 6 months of age trace nasal regurgitation was reported in the majority of infants. The frequency of this was highly variable but there was no significant difference between the No PSO and PSO groups. By 12 months of age the incidence of nasal regurgitation of solids had decreased to 50% for the UCLP groups and 33% for the ICP groups. As with nasal regurgitation of fluids, all infants with nasal regurgitation of solids at 12 months of age had fistulae.
Huddart and Zibemian (1977) claim that PSO reduce ulceration of the nasal septum by teats or nipples. In this study there was no difference in the incidence of ulceration between infants managed with PSO or not. Given that infants in the trial cohort demonstrate abnormal tongue movements and do not compress the teat against the palate or nasal septum, this finding was not remarkable.

Huddart and Ziberman (1977) also claim that PSO decrease choking in infants with CL and/or P. In this trial mothers were asked about indicators of airway protection, including coughing and choking during feeding and gurgley breathing during or immediately after feeding. A surprising number of infants were reported to show these risk factors for reduced airway protection, whether managed with PSO or not. The similarities in these reports between the No PSO and PSO groups for both cleft types support the findings of the videofluoroscopic assessments, that the closed system required for safe swallowing is not created with PSO in situ.

**Growth**

The final question addressed in this trial was whether PSO had an effect on the anthropometric measures of weight, length, head circumference and body mass index at 3 and 6 months of age. If PSO did have any benefit before palate repair, the growth patterns for infants managed with PSO would be expected to be better than those managed without PSO, perhaps more closely resembling normal infants. The majority of infants had decreased Z scores in the first month of life while their feeding routine was being established. However all infants showed improved growth by 6 months of age whether they were assigned to the No PSO or PSO groups. There were no significant differences in any of the mean Z scores (weight, length, head circumference) for the PSO and No PSO groups within each cleft type, at any assessment points. Hence these results provided no evidence that PSO were effective in improving feeding to any extent that was clinically relevant.
7.2 Factors which may have affected the results of the trial

There are many factors that might have affected the results of the study. These fall into the following categories: the trial design, the small numbers recruited, variations in the standardised care package, compliance with PSO and assessment tools.

7.2.1 Study design

The clinical team initially suggested a non-randomised trial comparing infants referred to St Andrews (where PSO were routinely used) to those referred to GOSH (where PSO were not routinely used). This had the advantage of allowing clinicians to continue with the regular treatment method they believed to be most effective. In addition it would avoid problems with parents requesting the routine treatment for their local team's site. This design could have confounded the results by introducing variables associated with the management site routine and style. Such systematic differences or bias have been reported to over or underestimate the differences between control and treatment groups (Altman, 1999). The use of the “gold standard” randomised control trial (RCT) design was chosen to avoid these biases (Altman, 1999).

PSO are clearly visible and therefore it was not possible to blind the researcher, parents or the cleft team to whether infants were randomised to the No PSO or PSO groups, allowing potential for differences in management. To minimise bias, the plan of care for nursing, orthodontic (with the exception of PSO), and surgical management of all infants was standardised. Documentation highlighted the first potential for bias with infants allocated to the PSO groups attending significantly more orthodontic appointments that those allocated to the No PSO groups. Unfortunately documentation designed to allow recording of feeding advice given by CNSs proved time consuming and was not completed. It was therefore not possible
to confirm that the frequency and type of input were entirely comparable for
the No PSO and PSO groups. One of the strengths of this study however
was that all surgery was carried out by the Lead Surgeon, thus excluding the
variable of surgeon's skill on outcome.

The researcher administered all research feeding assessments including the
oral motor assessments, GOMMF and videofluoroscopy perhaps introducing
the possibility of bias. The questionnaires used to obtain information about
demographics, and the medical and feeding histories offered multiple-choice
answers, thus reducing the risk of bias. The researcher made all
anthropometric measurements, ensuring consistency in the technique used.
While this was advantageous it did allow a potential for bias as the researcher
was not blinded to which PSO group the infant had been allocated. However
all feeding assessments were rated blindly by staff not involved in the trial and
who were unaware of the allocation of the infant. This minimised the
potential for bias in scoring the assessments.

7.2.2 Subjects

Cleft types are heterogeneous and it is recommended that studies in this area
should therefore take this into account (Fraser, 1970; World Health
Organisation Expert Committee, 2002). However, one of the difficulties of
research in the field is the variability in cleft presentation even within each
cleft type group (World Health Organisation Expert Committee, 2002;
Craniofacial Anomalies Network, 2003). There is also evidence that there
might be etiological differences within cleft type groups, such as between
isolated clefts of the soft palate only and clefts of the hard and soft palate
(Christensen and Fogh-Andersen, 1994). In order to ensure that samples are
homogeneous careful classification is essential (Harkins et al., 1960; World
Health Organisation Expert Committee, 2002; Craniofacial Anomalies
Network, 2003). The CRANE classification system, widely used throughout
the UK, was adopted. UCLP and ICP were chosen as they are among the
highest prevalence groups in which PSO are used in the UK (Sommerlad, 1994; Craniofacial Anomalies Network, 2003). As clefts of the soft palate only and those including both the hard and soft palates are considered separate entities with potential for different a response to treatment techniques, the inclusion criteria for this trial specified that clefts include the soft and at least 2/3rds of the hard palate.

Exclusion criteria included infants born prematurely before 36 weeks gestation and infants diagnosed at birth with a syndrome. Given that a syndrome diagnosis is not always possible within the neonatal period, infants who had major medical problems often associated with syndromes, such cardiac anomalies requiring surgical intervention and neurological impairment, were also excluded. More importantly, these factors are known to affect infant feeding and may therefore have influenced the results if included.

To ensure that groups were balanced for factors known to affect the majority of outcomes of the wider trial (including feeding), minimisation was used to ensure the balance of the groups for sex and birth order.

In addition, there are several other background variables known to affect feeding, which include ethnicity, social class and medical factors. As reported in Section 6.2, the groups were approximately balanced for these factors at the beginning of the trial (Altman, 1985). The infants’ medical status and family stress factors were also monitored throughout the trial period. It was fortuitous that there were no major differences either in the background variables that might have increased risk for developmental or feeding problems, between the No PSO and PSO groups, initially, or during the course of the trial.

Recruitment, unexpectedly, was a significant problem. The initial refusal of the parents of eight infants to enter the trial encouraged the team to review the way in which participation was approached. Although all members of the team were aware of their own biases and the need to
maintain equipoise, concern was raised as to whether this bias might have been subtly evident to parents.

The issue of refusal to participate in RCTs is often reported as an area of difficulty. It was complicated in this trial by the historical practice of the two sites involved (GOSH and St Andrews) and in particular the maternity units in the catchment areas of these units. The maternity units within the catchment area of St Andrews were fully aware of the routine use of PSO. Staff at these units often explained to parents that their infant would be fitted with PSO, detailing how this would help establish feeding, before a member of the cleft team was able to discuss the trial. In occasional cases, the member of the cleft team introducing the trial had to address this with parents. This only affected participation in the trial in one case.

Another factor influencing recruitment was an overestimation of the number of infants who would be eligible to participate.

This study took place against a background of major changes in the provision of care for children with CL and/or P in the UK.

Following concerns raised about variations in the standards of treatment of children with CL and/or P, the Clinical Standards Advisory Group (a statutory committee, which advises Health Ministers in England and Wales) requested a review of services to enable them to advise on any changes in practice, which would improve clinical standards (1998; Sandy et al., 1998; Bearn et al., 2001; Sandy et al., 2001; Sell et al., 2001; Williams et al., 2001). Following this review, the committee recommended that service provision be centralised into a small number of regional centres, thus ensuring multidisciplinary teams were able to treat high volumes of patients to a standardised protocol, carry out long-term follow-up and audit, and participate in inter-centre research. It was anticipated that this reorganisation, initially proposed to start April 2000, would result in a significant increase in the numbers of infants referred to the NTRCU.
However re-organisation proved to be a very difficult and complex process and designation of the centre did not occur until April 2002. Although the numbers of referrals did increase following this, it was not as much as anticipated. It was expected that approximately 25% of the increased caseload would have been UCLP and 41% ICP (although this group includes all clefts palate cases only) (Sommerlad, 1994; World Health Organisation Expert Committee, 2002; Craniofacial Anomalies Network, 2003). However this was not the case. Thirty two infants were born with clefts during the time frame. Eleven of the infants had ICP however only one had a cleft meeting the inclusion criteria of extending into 2/3rds of the hard palate. Another eleven infants were born with cleft lip and palate, however interestingly the majority (8) were born with BCLP. An additional ten infants had cleft lip only, thereby not meeting the inclusion criteria. One infant was found to have a submucous cleft. Two infants were also diagnosed with syndromes and were therefore excluded. Recruitment therefore did not increase.

7.2.3 Numbers of infants recruited

As presented in Section 6.11, in order to obtain 80% power at the 5% significance level 25 infants would have been required in each group. Given that the numbers recruited were smaller the power was less, and it is possible that differences of 0.8 SD were not identified. At the primary outcome measure point of 12 months, when the number of infants in the UCLP groups were 8 (No PSO) and 13 (PSO), only differences greater than 1.1 SD would have been detected. Similarly, within the ICP groups where the numbers of infants were 4 (No PSO) and 7 (PSO), only differences greater than 1.5 SD would have been detected. As a result, smaller but clinically significant differences may not have been detected.

There were also several instances where confidence intervals were wide. These were the result of the small number of infants recruited. The wide
intervals suggest that any differences between the groups (no PSO and PSO) are uncertain and it would be inadvisable to make definitive conclusions based on them.

7.2.4 Variations in aspects of standardised care

The first aspect of care where there was some variation was the provision of feeding advice. One of the two CNSs saw all parents of newborn infants for counselling. Parents were given the advice outlined in the protocol. There were, however, two differences in the provision of this advice. In cases where mothers wished to pursue breast-feeding, infants seen at GOSH received additional advice from the hospital’s “breast-feeding specialist” who has experience in helping mothers of infants with medical problems breastfeed. This additional advice was made available in two cases (UCLP PSO and UCLP No PSO). This resource was not available at St Andrew’s, and rather mothers were given advice by the community midwives and health visitors. The second difference was in the provision of equipment. Infants seen at St Andrew’s received four sets of bottles whereas those at GOSH received two sets only. This would be unlikely to have any impact on the results of the trial and was not considered important since randomisation was used within the centres. There were slight differences in advice about, and management of, the adaptation of teats, according to the individual preference of the CNS. The CNS at St Andrews routinely enlarged holes in teats and created additional holes. In contrast the CNS at GOSH preferred to give parents unadapted teats, and only to make teat adaptations if indicated. Her experience had been that parents found the adaptation of teats difficult, and costly because of failed attempts, and that they worried about the safety of modifying teats against the manufacturer’s advice. GOSH had therefore organised for the manufacturers to supply teats with enlarged holes. It is difficult to know the impact of these different adaptations on the infants’ feeding patterns, although in principle both techniques should have served the same purpose of increasing the flow rate. According to the protocol any
additional advice given by the CNS would be recorded. Unfortunately this aspect of the protocol was not adhered to. It is feasible that caregivers/parents of some infants were given advice that has not been recorded.

7.2.5 Orthodontics

The protocol for the trial required that infants with UCLP be seen for a total of nine orthodontic reviews in the first six months, and infants with ICP four times in the first six months. This requirement was however difficult to adhere to and there was a significant difference in the number of appointments attended by the UCLP groups. Infants in the UCLP PSO group had an increased number of appointments compared to those in the UCLP No PSO group. Theoretically, parents of the former group might have benefited from additional contact with professionals allowing opportunity for extra support. However the increased number of appointments could also be interpreted as adding to the burden of care, with increased costs to the family of travel for appointments, additional day care for siblings and time off work.

7.2.6 Adherence to protocol specified assessment times

As discussed in Section 6.3, there was some variability, but no significant difference between groups for the mean age of infants at assessment. To minimise the effect of variability within and between the groups, scores were used in comparisons of anthropometric measures. The knowledge of the developmental changes in feeding which occur in the first 12 months of life was a major factor in requiring that assessment times were adhered to as closely as possible. The most significant changes in feeding patterns occur within the first 3 months of life (Pollitt et al., 1981; McGowan et al., 1991; Wolf and Glass, 1992; Qureshi et al., 2002). At the neonatal assessment the mean age and age range at assessment were similar across the No PSO and PSO groups. At the 3-month assessment, the infants' ages were again similar
for the UCLP groups. However, one subject in the ICP PSO group was assessed later than the others. Despite this, the infant performed similarly to others in the group. The validity of his NOMAS results might, however, be questioned as he was outside the standardised age range for the assessment tool. The age ranges at the 6- and 12-month assessment points were similar. Overall adherence to the specified assessment times was excellent and no statistical adjustments were required for analysis.

7.2.7 Assessment tools

*Structured interviews*

The structured interview questionnaires allowed systematic collection of demographics, medical and feeding history. The majority of questions were factual and closed and multiple-choice answers were offered. Parents were instructed to answer the questions based on the current situation, rather than to recall previous performance or events. It is recognized that the answers to questions in the feeding questionnaires may not always have been accurate. One example would be when parents were asked how long it typically took their infant to complete a bottle-feed. Difficulties sometimes arose when infants had highly variable feeding patterns and mothers found it difficult to report a typical pattern. Nevertheless, parents were often highly focussed on their infant’s feeding and many had very concise food diaries allowing averages to be identified. Several questions were asked about symptoms that might suggest reduced airway protection. One was “Does your baby sound gurgley during or after feeds?” This question is routinely asked in clinical feeding evaluations (Wolf and Glass, 1992; Arvedson and Brodsky, 2002) and rarely causes confusion for parents. However parents of infants with CL and/or P found it difficult to differentiate between the commonly reported nasal snuffle and a true pharyngeal gurgle. Another issue was the use of the questionnaires with families where English was not their first language. Wherever possible, professional interpreters were used to administer the
questionnaires but there were situations where family members acted as interpreters. This had the advantage of the parents generally being comfortable and familiar with "the interpreter". However there were situations where mothers appeared uncomfortable discussing their medical history with an older male member of the family functioning as the interpreter.

**Administration of assessments**

The majority of feeding assessments were carried out in the infant's home. This setting had the advantage of ensuring that the environment was familiar for mother and infant. A common challenge was the co-ordination of assessment times with the infant's feed times. The majority of mothers attempted to anticipate the time at which their infant would feed, however on many occasions routines went awry and the researcher would need to spend extended times waiting for infants to wake or be ready for feeds. There were also occasions when mothers forgot appointments and visits had to be rescheduled. Occasional difficulties occurred with siblings and pets interfering with equipment and the assessment procedure. Some assessments were administered either at St Andrews or GOSH. These appointments were usually arranged to co-ordinate with ward admissions or outpatient clinics and rarely occurred as planned. Feeding assessments were therefore sometimes delayed. On several occasions invasive medical procedures such as blood tests were administered prior to feeding assessments, upsetting the infants. As a result the feeding assessments may have been less than optimum.

**Oral motor assessment scales**

Given the developmental changes in infant feeding patterns over the first year of life two different oral motor assessment scales were required, the Neonatal Oral Motor Assessment Scale (NOMAS) and the Schedule of Oral Motor Assessment (SOMA).
No oral motor/feeding scale exists for children with structural anomalies. As discussed in Chapter 3, the NOMAS was originally developed for use with premature infants. Its use in research to date has been limited to this area. Prior to using this tool several issues regarding the administration of the scale needed clarification with one of the authors (Meyer Palmer, 1999). It was advised that the infants’ own adapted feeding bottle and teat could be used. In using the scale with infants with CL and/or P, several weaknesses became evident. As discussed earlier the majority of the infants were fed with modified teats. Given that flow rate is known to have an effect on sucking patterns, comparing infants with different feeding equipment would have introduced flow rate as a potential variable. In order to ensure that this was not the case the equipment used was standardised as per the GOSMIF protocol. Two assessments were therefore carried out. The first was made from the standardised GOSMIF assessment. The second assessment was administered with the infant’s own soft bottle and adapted teat. While the feeding pattern assessed with the infant’s own bottle allowed for individual variation in the number and type of adaptations made to the teat as well as the mother’s technique in using a soft bottle, it did allow the infant’s “best” feeding patterns to be observed. Hence the comparisons of results obtained under each condition are presented in Section 6.10.3. At the neonatal assessment, there was poor agreement of the oral motor rating in both the unadapted GOSMIF assessment and the routine feeding assessments. One possible explanation for these differences might be that the infants had, or were in the process of mastering feeding with adapted bottles and developing alternative oral movement patterns to compensate for their anatomical abnormalities. It was predicted that infants might be rated as having less severely impaired oral motor skills when feeding with their own adapted bottle. They would be receiving an appropriately sized bolus, as their sucking was supplemented by their mother’s use of the soft bottle. Thus the need for compensatory behaviours such as fast vigorous sucking (in an attempt to transfer milk from the bottle) would be minimised. However this was not
found to be the case. Several infants were rated as having better oral motor skills when feeding with the unfamiliar GOSMIF bottle and teat. These infants apparently had less disorganised feeding with the GOSMIF bottle and teat. This was perhaps attributable to the infant having more control over the flow rate of the feed, allowing the infant more time to co-ordinate the suck and swallow, as compared to the faster flow rate of the adapted bottle and teat. Crucially however, in spite of the better sucking patterns with unadapted bottles and teats, the infants took very little fluid during the GOSMIF assessment and hence this method was inadequate nutritionally.

Given the high proportion of infants categorised as having dysfunctional oral motor skills a closer analysis of the 28 items in the scale was undertaken (Appendix 13). In order to be rated as dysfunctional only one item within the individual category had to be present. Typically four main features within the dysfunctional category were scored: reduced or excessive degree of jaw movements, arrhythmic jaw movements during sucking, arrhythmic tongue movements during sucking, and an inability to maintain a sucking pattern for two minutes.

A further item was whether there was a difference in the rate of sucking between the non-nutritive dummy sucking and the nutritive bottle-feeding. A normal rating is achieved on this item if the infant is able to change their rate of sucking from approximately 2 sucks/second with a dummy, to 1 suck/second with a bottle. If there is no change in the rate of sucking a dysfunctional rating is given. The majority of infants were sucking at the faster non-nutritive rate throughout the feed. Dummies are rarely given to infants with CL and/or P as surgeons usually advise against their use. They are believed to interfere with healing and possibly to contribute to surgical breakdown following palate repair. The few infants who did accept the dummy were unable to hold it within their mouths. This was probably due to no experience of having one, abnormal tongue movements and/or lack of an opposing surface against which to position the teat. It might also be that the
bulk of the dummy, which is greater than that of a teat, interfered with the infants' habitual airway, resulting in reflexive expulsion of the teat from the mouth. Given that so many infants refused the dummy for assessment of non-nutritive sucking there may have been a failure to assess correctly the difference between the non-nutritive and nutritive sucking rate. This was a limitation of the classification system. Nevertheless, it was the only published standardised scale available.

The NOMAS is only applicable for infants up to 3 months of age. For assessments at 6 and 12 months of age there was only one standardised oral motor assessment scale available, the SOMA. As discussed in Section 3.3 the SOMA gives an overall rating of normal or abnormal oral motor skills based on scores of individual items for a variety of food textures. All infants were rated as having normal oral motor skills at 12 months of age (Section 6.10.3). In addition, there were several infants from all four groups who were uncooperative during the assessment and who refused one or more textures. Unfortunately, it was impossible to determine whether this refusal pattern was attributable to taste preferences, difficulty managing the texture or to behavioural factors. The SOMA prescribes the foods to be used in the assessment. Although the schedule was developed and standardised on a British cohort, a large group of the infants in this study refused these foods. This may have been because of their ethnic background. Substitutions of similarly textured foods were made as ethnically appropriate e.g. dhal mixed with rice replacing the item baked beans. Of those infants who refused one or more textures, all were reported by their parents to take these or similar foods, suggesting that the refusal was behavioural in nature. Another area of difficulty was adherence to the administration guidelines as to how the various textures should be given to the infants. At 6 months of age, infants allowed their mothers to spoon-feed them purees according to the administration guidelines and took fluids from either their own bottles or training beakers. Unfortunately however, approximately 60% of the video views of the infants' faces were obstructed by the mother's hand, or the bottle or training beaker,
preventing the studies from being rated. At 12 months of age a different problem arose. The infants had reached the developmental stage of self-feeding. Few infants allowed their mothers to feed them according to the administration guidelines and insisted on attempting self feeding. As a result, there were individual items, which relied upon specific presentation of a spoon, for example those related to lip movement in clearing the bolus from the spoon (which required that the spoon was presented horizontally and not “scraped clean” on the top lip) could not be scored. This difficulty was discussed with one of the authors, who advised on how to score accommodating for this (Reilly, 2002). This modified scoring did not change the overall ratings of normal or abnormal oral motor skills at 12 months of age.

**GOSMIF**

The GOSMIF allowed objective and reliable assessment of several aspects of infant bottle-feeding including patterns of sucking, pressure generation during sucking and swallow identification. The development of the GOSMIF and the resolution of technical difficulties are discussed in detail in Sections 4.1-4.4.

Assessment of a group of non-cleft infants allowed collection of data on patterns of normal feeding and comparisons with the trial cohort increasing our knowledge of the nature of feeding in infants with CL and/or P.

The procedure for these assessments was standardised ensuring that comparisons could be made between groups and the non-cleft trial. Identical equipment was used throughout and all infants were given 5% glucose solution avoiding any variation in feeding patterns associated with the composition of formula given.

Very few problems arose with the use of the GOSMIF in the trial cohort. There was a proportion of infants who would not co-operate for GOSMIF
assessments. Initially, the number of infants refusing GOSMIF assessment was small. The refusers were those infants who had previously undergone invasive medical interventions and were unsettled. However, at the 6-month assessment point, when the infants were actively involved in feeding, few infants completed the 5-minute trial feed, the majority co-operating for only 1 or 2 minutes. At this age, many infants insisted on holding the bottle, pulling at the wires and in several cases appearing to encourage their mothers to squeeze the bottle (by squeezing her hand). In addition the majority of infants became extremely frustrated and began wriggling and fussing. This reduced co-operation may have been due to the infants' awareness that they were not able to achieve an adequate bolus with the rigid bottle. An increased bolus may have been achieved by replacing the medium-flow teats with the fast-flow version. However this may have been dangerous for those infants unable to cope with the faster flow rate, increasing their risk of aspiration. It might also be that the infants did not like the taste of the glucose solution at this age.

The reliability of the GOSMIF has been discussed in detail in Section 4.6.

Although the validity of the GOSMIF has not been formally assessed, it was reassuring that measures collected in the non-cleft sample were similar to those published for age-matched infants. In addition, the simultaneous videofluoroscopic and GOSMIF assessment confirmed that pressure transducer traces indicating sucks were such, and auscultation traces were appropriately matched to swallows. However only one study of this type was carried out and further studies are essential. The non-cleft data for this trial was only collected at the neonatal assessment point. Follow-up at 3 and 6 months would provide additional information about the developmental changes in sucking patterns.

Given that the GOSMIF proved easy to use, reliable, well tolerated by infants and their mothers and provided accurate information about infant bottle-
feeding, it will be a useful assessment technique with many groups of infants who have bottle-feeding difficulties.

**Anthropometry**

The measurement of weight, length and head circumference is reported to be reliable with some training (World Health Organisation, 1986; World Health Organisation Expert Committee, 1995). The age at which anthropometric data were collected varied to a small extent. Growth increases rapidly during the first year of life and potentially these differences may have affected the results. To account for this Z scores were calculated. The Z score system expresses the anthropometric raw measurement as a number of standard deviations or Z scores below or above the reference mean, taking into account the infant's age and sex. In order to ensure that the reference means used were culturally appropriate to the trial cohort the British 1991 Growth Reference Charts were used in the calculation of Z scores (Freeman et al., 1995; Child Growth Foundation, 1999). The World Health Organisation recommends that when assessing response to an intervention in infancy, weight, length, age and sex are recorded allowing weight for age and length for age or Z scores to be assessed (World Health Organisation Expert Committee, 1995). In this trial, Z scores for weight, length and head circumference were collected, adhering to the recommendations of the WHO. Body Mass Index scores were also calculated providing an overall measure of body mass relative to length.

**7.2.8 Compliance with PSO**

Compliance was monitored closely in this study. Parents were questioned by the researcher at each assessment point about the length and frequency of PSO wear. It was felt that parents might not acknowledge inconsistent use of the PSO to the orthodontist, as he was directly involved in its provision and
adjustment. To avoid this potential bias use of the PSO was determined by the researcher.

Compliance was reasonably good immediately post-fitting and at 3 months of age. However by 6 months of age compliance was poor with only 14 of the 23 infants wearing their plates all day. The parents of many of these infants reported that there had been occasions when they had forgotten to replace the appliance after cleaning for feeding. They did not notice any deterioration in feeding and had therefore made an active decision to abandon the PSO use.

7.2.9 Medical problems over time

Few infants developed medical problems over the period of data collection. The majority of infants with ICP had been diagnosed with Pierre Robin Sequence and it was therefore not surprising that a proportion of them developed respiratory difficulties requiring nasopharyngeal airways (Caouette-Laberge et al., 1994; Heaf et al., 1982; Pashayan and Lewis, 1984; Robertson, 1988). All of the infants requiring airway support also required total or supplementary feeds. A further 2 infants with UCLP developed respiratory problems. Rather than being obstructive in nature these respiratory difficulties were related to recurrent coughs and colds, requiring antibiotic treatment. These infants were both found to be aspirating on the videofluoroscopic assessments. One case was managed with nasogastric feeding and the other with thickening the feeds.

Another infant was diagnosed with biliary atresia and multiple fractures associated with child abuse. Knowledge of feeding patterns in infants with biliary atresia suggested that this infant’s feeding might be abnormal (Arvedson and Brodsky, 2002) and growth therefore affected. It might be argued that this infant should therefore have been excluded from the
analysis, however in order to adhere to the protocol inclusion and exclusion criteria he was included in all analysis.

A common but unexpected problem across both cleft types, irrespective of PSO status, was the presence of symptoms suggestive of, or a confirmed diagnosis of, gastro-oesophageal reflux. Gastro-oesophageal reflux (GOR) is defined as retrograde flow of gastric secretions from the stomach into the oesophagus. It can have a significant impact on infant feeding (Hyman, 1994; Mathisen et al., 1999). It can be difficult to manage and may be a contributory factor in infants developing behavioural or hypersensitivity feeding problems (Hyman, 1994; Putman, 1997; Arvedson and Brodsky, 2002). This problem is known to occur in infants with Pierre Robin Sequence (Poole et al., 1982; Elliot et al., 1995; Dudkiewicz et al., 2000; Baujat et al., 2001). It was therefore not surprising that consistent vomiting (a common symptom of GOR) was reported in 63% of the ICP group within the first month of life. However it has not been reported in infants with UCLP, making it more of a surprising finding that 50% of infants in this cohort vomited consistently within the first month of life. Gastro-oesophageal reflux requiring medication was diagnosed in 6 (18%) infants in the UCLP groups and 3 (18%) infants in the ICP groups. The percentages of infants requiring medications for management of GOR were similar at 3 and 6 months of age, but by 12 months of age none of the infants required medications. In contrast, the UK survey found that only 2.3% of mothers reported that their infants vomited regularly (Hamlyn et al., 2002). There is the possibility that parents’ views of what constitutes normal and abnormal vomiting may have affected these findings. Parents of infants with CL and/or P are very focussed on their infant’s feeding and growth, and it may be that they seek advice and intervention earlier than mothers of non cleft infants do. However medications would not have been prescribed unless the paediatrician was convinced of the diagnosis.
7.2.10 Analysis

A plethora of comparisons were made between the No PSO and PSO groups for each cleft type. While these resulted in there being many p values, the significant values were not considered in isolation but rather in the overall context of the results. It is understood that with so many tests some will be falsely statistically significant. For example parents were asked if their infant coughed or choked when eating solids. As presented in Table 28, there was a significant difference at 0.05, between the number of infants in the UCLP No PSO and UCLP PSO groups reported to show this behaviour. However when the other risk factors for reduced airway protection such as gurgley breathing and coughing and choking with fluids are considered there were no other significant patterns suggesting that this was not probably clinically important. If PSO were having an affect patterns of differences would have been expected rather than isolated findings.

7.3 Generalising these findings to other cleft types and protocols for management of infants with CL and/or P

This trial assessed the effect of PSO on the feeding patterns of infants with UCLP and ICP within the clinical setting of the NTRCU. Following CSAG recommendations (1998), many aspects of care of infants with CL and/or P are standardised. Hence the findings of this study should be applicable for other cleft teams following this care protocol. Where differences in practice exist, such as in surgical techniques and timing of surgery, the results may be less applicable. Infants in this trial underwent surgical repair of the cleft palate at approximately 6 months of age. Feeding patterns change rapidly over the first year of life and it may be that if surgery is carried out considerably before or after 6 months, developmental aspects of feeding may interact with the effect of PSO. The majority of infants with CL and/or P are fed with soft bottles and the

- 342 -
findings of this study would therefore be directly applicable in this respect. Surgical skill has been shown to have a significant effect on outcomes such as speech and facial growth (CSAG Report 1998) and it therefore seems likely that it may also affect feeding development. In this trial one surgeon carried out all the surgery according to his defined protocol, thus eliminating the surgical variables of technique, timing and surgeon skill.

7.4 Further research

It was unfortunate that the numbers of subjects in the study were fewer than envisaged. The power of the study would be increased by recruiting an additional 17 infants to the UCLP groups and an additional 34 infants to the ICP groups. Such numbers should provide definitive answers to the questions posed.

The trial has also highlighted other areas of possible research. The nature and progression of feeding in CL and/or P is poorly understood. Management is based on subjective and anecdotal findings and is rarely objectively assessed. To ensure that practice is evidence based, further research into the nature and management of feeding, using objective assessment techniques, as used in this study, is essential. Studies comparing the feeding patterns of infants with CL and/or P to those of non-cleft infants over time will provide further information about the nature and history of feeding patterns in infants with CL and/or P. Research should attempt to tease out the aspects of feeding in infants with CL and/or P which are innate and aspects which may be learned in order to compensate for anatomical defects. The finding that the pharyngeal stage of swallowing is also affected was unexpected and further investigations into the effects of management techniques and surgery are warranted. Co- incidental findings of this trial suggest that the management techniques utilised routinely, such as soft bottles, may contribute to the development of abnormal sucking patterns and pharyngeal stage problems. This urgently requires further investigation since this is routine practice by
clinical nurse specialists. Given that recent research has highlighted
behavioural feeding problems in toddlers with CL and/or P (Daryanani,
2003), another priority should be to assess whether the early management
 techniques utilised may have impacted on these later problems.

7.5 Conclusions

Unfortunately the small numbers of infants recruited to the trial limited the
power of the results. As result only differences in excess of 1.1 SD for the
UCLP groups and 1.5 SD for the ICP groups would have been detected with
reasonable power. It may be that smaller and yet clinically significant
differences were not detected.

The findings of this trial were however that PSO did not improve feeding
efficiency or general body growth post palate repair at 12 months of age,
either in infants with UCLP or ICP. If feeding efficiency had been improved,
a significant difference in the growth of infants randomised to the PSO
groups would have been expected. This was found not to be so.

Similarly, before palate repair at 3 and 6 months of age, no significant
differences in feeding patterns or growth were found. The many hypotheses
as to how PSO give more efficient feeding by acting as an obturator and
providing an opposing surface prior to repair of the palate, are similarly not
supported.

Infant feeding difficulties increase the burden of care for families (Adams et
al., 1999). Feeding is a highly emotive area and mothers of infants with
feeding difficulties often experience feelings of inadequacy when unable to
feed their infants as they had planned. Constant concern about the amount of
feed the infant takes and subsequent growth places stress on the family unit.
In addition, special feeding equipment and techniques are often used.
Relatives and friends who are often readily available to offer support to
mothers of healthy infants can be reluctant and anxious about helping with
feeding infants with CL and/or P. Frequent medical and hospital appointments may be costly and time consuming and may foster a dependency on the professionals. PSO can add to an already significant burden of care by increasing the number of hospital appointments and adding care of yet another piece of equipment during a time when parents are often struggling to come to terms with having a baby with a cleft (Solnit and Stark, 1962). This would be unwarranted unless there was evidence that the benefits of the treatment outweighed the increased burden (World Health Organisation Expert Committee, 2002). Basing a decision on the use of PSO purely on feeding outcomes may, however, be inappropriate as it is could be that PSO have other advantages, such as increased ease of surgery, improved facial growth outcomes and speech development. It will be several years before surgical and speech outcomes are known for the infants enrolled in this trial.
Pressure Sensors
Absolute Unamplified Noncompensated

24PC Series

FEATURES
• Absolute pressure measurement
• Miniature package
• 2-15 and 2-30 psi pressure ranges
• 2 mA constant current excitation significantly reduces sensitivity shift over temperature

24PC PERFORMANCE SPECIFICATIONS

<table>
<thead>
<tr>
<th>Parameter</th>
<th>Range</th>
<th>Min</th>
<th>Typ</th>
<th>Max</th>
<th>Units</th>
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<tr>
<td>Excitation</td>
<td>bar</td>
<td>10</td>
<td>12</td>
<td>VDC</td>
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<tr>
<td>Null Shift</td>
<td>2-15</td>
<td>±2.0</td>
<td>±4.0</td>
<td>mV</td>
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<td></td>
<td>2-30</td>
<td>±2.0</td>
<td>±5.5</td>
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<tr>
<td>Linearity</td>
<td>2-15</td>
<td>10</td>
<td>20</td>
<td>% Span</td>
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<td></td>
<td>2-30</td>
<td>15</td>
<td>30</td>
<td></td>
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<td>Sensitivity Shift</td>
<td>All</td>
<td>±5.0</td>
<td>±6.5</td>
<td>% Span</td>
<td></td>
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<tr>
<td>Repeatability &amp; Hysteresis</td>
<td>All</td>
<td>±0.5</td>
<td></td>
<td>% Span</td>
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<td>Input Resistance</td>
<td>4.0 K</td>
<td>5.0 K</td>
<td>6.0 K</td>
<td>Ohms</td>
<td></td>
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<tr>
<td>Output Resistance</td>
<td>4.0 K</td>
<td>5.0 K</td>
<td>6.0 K</td>
<td>Ohms</td>
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<tr>
<td>Weight</td>
<td>2.6</td>
<td></td>
<td></td>
<td>grams</td>
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</table>

ENVIRONMENTAL SPECIFICATIONS

Operating Temperature: -40 to +85°C (-40 to +185°F)
Storage Temperature: -55 to +100°C (-67 to +212°F)
Shock Qualification tested to 150 G
Vibration Qualification tested to 0 to 2 Hz, 2G sine
Media Compatibility: Limited only to those media which will not attack polyetharimide, silicon, fluorosilcone and silicone seals.

*Span: the algebraic difference between output endpoints
**B.F.S.L.: Best Fit Straight Line

24PC ABSOLUTE ORDER GUIDE

<table>
<thead>
<tr>
<th>Catalog Number</th>
<th>Pressure Range</th>
<th>Min.</th>
<th>Typ.</th>
<th>Max.</th>
<th>Null Offset</th>
<th>Sensitivity</th>
<th>Over- pressure</th>
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<tr>
<td></td>
<td>psi</td>
<td></td>
<td></td>
<td></td>
<td>mV</td>
<td>mV/psi</td>
<td>psi Typ.</td>
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<td>24PCD</td>
<td>2-15</td>
<td>-140</td>
<td>-200</td>
<td>-260</td>
<td>-46</td>
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<td></td>
<td>2-30</td>
<td>-180</td>
<td>-300</td>
<td>-340</td>
<td>-61</td>
<td>+29</td>
<td>11</td>
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</table>

*Non-compensated pressure sensors, excited by constant current instead of voltage, exhibit temperature compensation of Span. Application Note #1 briefly discusses current excitation.

Constant current excitation has an additional benefit of temperature measurement. When driven by a constant current source, a silicon pressure sensor's terminal voltage will rise with increased temperature. The rise in voltage not only compensates the Span, but is also an indication of die temperature.

Honeywell • MICRO SWITCH Sensing and Control • 1-800-537-6945 USA • +1 815-236-6847 International • 1-800-737-3390 Canada
Pressure Sensors
Absolute Unamplified Noncompensated

24PC SERIES ABSOLUTE PRESSURE SENSOR OUTPUT CURVE

EXCITATION SCHEMATIC

TERMINATION STYLE
Style 6 - 1 x 4
Pin 1 = Vs (+)
Pin 2 = Output (+)
Pin 3 = Ground (-)
Pin 4 = Output (-)
Pin 1 is notched
Pin 2 is next to Pin 1, etc.

SENSOR SELECTION GUIDE

<table>
<thead>
<tr>
<th>Product</th>
<th>4 Circuit Type</th>
<th>Pressure Transducer</th>
<th>C Pressure Range</th>
<th>F*+ Type of Seal</th>
<th>D* Type of Port (P1)</th>
<th>6 Termination Style</th>
<th>A Pressure Measurement</th>
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</thead>
<tbody>
<tr>
<td>220PC</td>
<td>Standard</td>
<td>C 2-15 psia</td>
<td>1 bar</td>
<td>F Fluoro-</td>
<td>A Straight</td>
<td>6 x 4 (600' long)</td>
<td>A Absolute</td>
</tr>
</tbody>
</table>

**Media seal is on P1 side and will not be in contact with media**

Example: 24PCF6DA
Non-compensated 15 psig Absolute sensor with fluorosilicone seal, modular port, 1 x 4 terminals, 600' long.

MOUNTING DIMENSIONS (for reference only)

Honeywell MICRO SWITCH Sensing and Control • 1-800-537-6945 USA • 1-815-235-6847 International • 1-800-737-3360 Canada
Appendix 2

Knowles

• Ceramic Vibration Transducer
• High Vibration Sensitivity And Small Size
• Wide Frequency Range
• Internal FET Preamplifier
• High Mechanical Durability

Typical Response Curve

Open Circuit vibration sensitivity

FREQUENCY IN HERTZ

RESPONSE MEASURED WITH 15VDC SUPPLY IN 2 WIRE MODE AND 1G ACCELERATION. RESPONSE REMAINS FLAT DOWN TO 20Hz.
BU 1771

Specification Data

- **Output Impedance**: 5200 ohms (Nominal) in two wire circuit
- **Supply Voltage**: 1.5 to 10Vdc
- **Current Drain**: 50μA (Maximum) in two wire circuit with 1.5Vdc
- **dc Resistance between case and negative terminal**: 100 ohms (Maximum)
- **Weight**: 0.28 grams (Nominal)

Outline Drawing

- Dimensions shown in millimeters (inches)

Application Circuits

- **Three Wire Circuit**
- **Two Wire Circuit**

NOTE: BU 1771 is qualified as a two wire circuit device and also usable as a three wire hook up with approximately 10% increase in sensitivity.

This information on this Data Sheet reflects typical performance. Specific test methodologies of each model are available by requesting Outline Drawing Sheets and Performance Specification Sheets. Knowles' responsibility is limited to compliance with the Outline Drawing and the Performance Specifications applicable to the subject model at time of manufacture.
Appendix 3

Knowles®

EK series

- Electret Microphone
- Uses A-Vibro-Metric (AVM™) Construction To Give:
  Low Vibration Sensitivity
  The Highest Knowles Electroacoustical Sensitivity
- Low Noise Level
- Inbuilt FET Preamplifier
- Rugged Construction To Withstand Severe Temperature
  And Humidity Conditions
- High Resistance To Mechanical Shock
- Various Responses Available

Typical Response Curve

![](Typical_Response_Curve.png)

Knowles Electronics Co.
73 Victoria Road, Seygate, P.A.
West Sussex, BN13 1FF, England
Phone: 01444 235432
Fax: 01444 240724

Knowles Electronics, Inc.
1151 Mountain View
Itasca, Illinois 60143, U.S.A.
Phone: (630) 293-5100
Fax: (630) 293-2875

- 350 -
EK series

Other Response Curves

Sloped Response
6dB - Solid Line
12dB - Broken Line

Stepped Response
3021 Solid Line
3115 Broken Line

Flat Response
3033 - 3132 - 3163 - 3133
### Respiration cables and assemblies

<table>
<thead>
<tr>
<th>Order number</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>M92100</td>
<td>Capable/Kemp Cable open end, Length = 1.20 mtr.</td>
</tr>
<tr>
<td>M92102</td>
<td>Capable/Kemp Cable with 2mm female connectors, Length</td>
</tr>
<tr>
<td>M92110</td>
<td>Adapter cable for Capable/Kemp cable with bridge adapter</td>
</tr>
<tr>
<td>M92115</td>
<td>Respiband set Premature</td>
</tr>
<tr>
<td>M92120</td>
<td>Respiband set Infant/Baby</td>
</tr>
<tr>
<td>M92125</td>
<td>Respiband set toddler (14&quot; - 20&quot;)</td>
</tr>
<tr>
<td>M92135</td>
<td>Respiband set small adult (22&quot; - 28&quot;)</td>
</tr>
<tr>
<td>M92145</td>
<td>Respiband set regular adult (28&quot; - 36&quot;)</td>
</tr>
<tr>
<td>M92147</td>
<td>Respiband set large adult (32&quot; - 40&quot;)</td>
</tr>
<tr>
<td>M92155</td>
<td>Respiband set junior adult (34&quot; - 43&quot;)</td>
</tr>
<tr>
<td>M92165</td>
<td>Respiband set x large adult (48&quot; - 66&quot;)</td>
</tr>
<tr>
<td>M92175</td>
<td>Respiband set senior adult (42&quot; - 54&quot;)</td>
</tr>
<tr>
<td>M92185</td>
<td>Respiband set medium adult (20&quot; - 32&quot;)</td>
</tr>
<tr>
<td>M92190</td>
<td>Respiband Clip-adapter (70cm) for ambulatory recorder wire</td>
</tr>
</tbody>
</table>

### Contact Information

TEMEC can be contacted directly through its team of international staff in the Netherlands or via one of our distributors.

**INTERNATIONAL:**

TEMEC Instruments B.V.  
Speikofstraat 2  
NL-6466 LZ KERKRADE  
The Netherlands

**Telephone:** +31-45-5428888  
**FAX:** +31-45-5428584
Development of a System to Study Infant Feeding.

J. I. Veness¹ A. Masarei²

¹Biomedical Engineering Department, ²Speech and Language Therapy


Abstract — A system has been developed that is portable and simple to use to study signals associated with feeding, these being sucking, swallowing and breathing. Three transducers are attached to the patient and one is mounted in the feeding bottle. Swallowing is monitored by a microphone and vibration transducer, breathing is detected with an impedance band fitted round the chest. Signals are conditioned and fed into a laptop PC where the data is displayed and stored along with a simultaneous video picture. Test results can then be reviewed and studied at a later date. Preliminary results have shown that the device is well tolerated by the baby and during feeding the suck/swallow/breathe ratio of 1:1:1 has been observed which is consistent with the literature.

1. Introduction

The process of infant feeding can be broken down into three main functions, these being Suck, Swallow and Breathe. These processes are tightly integrated with each other and share related functionality and anatomical structures. For example some structures have a dual role in providing oxygen and nourishment to sustain the child.
1.1 Suck

During the suck the mouth is used as a pump. Two types of pressure are created, these being positive and negative. The infant compresses the teat with the tongue causing the fluid to pass from the high pressure inside the bottle, to the lower pressure mouth. The infant can also create a negative pressure in the mouth by sealing the oral cavity and dropping the jaw and tongue thus enlarging the cavity. Both pressure differential generation methods have been found to play a role in infant feeding. (Brake et al, 1988).

1.2 Swallow

The swallowing process is coordinated by a collection of motor nerves which excite a large number of muscles in the mouth, pharynx, larynx and esophagus. It can be split into 3 phases, 1) Oral phase 2) Pharyngeal phase and 3) Esophageal phase.

1.2.1 Oral Phase

The oral phase prepares the food ready for swallowing. This ensures that the food is small enough to pass into the stomach, and forms the food into a bolus. The bolus is transported from the front of the mouth to the back in readiness for initiation of the swallowing sequence. In babies the oral stage mainly consists of sucking and passing the liquid to the back of the mouth. This continuous process still maintains the bolus so that a swallow is not initiated by fluid leaking into the pharynx.

1.2.2 Pharyngeal Phase

The pharyngeal phase is controlled by twenty six muscles and six cranial nerves, and is initiated by the impact of the bolus on sensory receptors in the back of the mouth. First the nasal, laryngeal and oral openings close to
prevent leakage, and to channel the bolus. Secondly the cricopharyngeal sphincter opens, and finally the bolus moves through the pharynx by the combined action of peristalsis and the changing pressure gradient within the pharynx.

1.2.3 Esophageal Phase

The final stage of transferring food from the mouth to the stomach is the esophageal phase. Initially the upper sphincter relaxes to allow the bolus to pass into the esophagus. Peristaltic action then propels the bolus down the esophagus to the lower sphincter which relaxes to allow the bolus to pass into the stomach.

The process of evaluating a patient's disorder tries to establish where the feeding process is breaking down and thus where the problem lies. The severity of disorder can then be ascertained. Examples of disorders are Gastroesophageal reflux, neurology problems such as inadequate motor control and lack of coordination, muscle disorders such as muscular dystrophy, structural congenital problems like cleft lip and palette and airway breathing difficulties.

To date many devices and techniques have been developed to study the complex process of feeding. These include pressure transducers and manometers used to measure intra-oral pressures during sucking, ultrasonography and videofluoroscopy which provide real time images of the pharyngeal swallow, cervical auscultation - listening to the feeding sounds with a stethoscope, and oximetry which gives an indication of the baby's response to work. (Braun and Plamer, 1985; Wolf, 1968; Eishima et al, 1991; Bosma et al 1990; Newman et al 1991; Vice et al, 1990; Ackemann, 1993).

Many of these techniques are unsuitable for the routine clinical environment, and do not lend themselves to portability due to their bulk. They also require operation by specialist staff from other departments. To carry out a valid
assessment the child must be ready for a feed and be in a relaxed and quiet environment as similar as possible to its normal routine. Few of these procedures can be carried out simultaneously adding to patient intrusion, time and expense.

To solve these problems an easily transportable, minimally invasive system is required, that can simultaneously acquire signals from the three most important feeding parameters: sucking, swallowing, and breathing.

2 Instrumentation

An overview of the system designed to collect the various signals from the patient can be seen in the block diagram below:

![Swallow Study Recording System Block Diagram](image)

The signal transducers chosen for the project are as follows:

Pressure, Honeywell 24PC 1psi peizoressitive sensor.

Acceleration, Knowles BU1771 ceramic vibration transducer.

Sound, Knowles EK3132 Electret Microphone.
Respiration, Respitrace expanding impedance band.

Video recording was carried out using a Sony DCR-TRV900E Digital camera.

The notebook computer used is a Dell Latitude 366XT Pentium 2 with the following cards installed:

Nogatech CaptureVision PCMCIA type 2 Video capture card. Max resolution 384x288.

Amplicon PCM DAS08 PCMCIA type 2 ADC Card, 12 bit resolution.

2.1 Electronic Design

The electronic interface between transducer and computer serves two main functions. It amplifies and conditions the small signals from the patient and provides an isolation safety barrier, required to meet the BS5724 Part 1 electrical safety standard. The circuit can be divided into four channels as described below.

The pressure transducer is mounted in the end of the bottle and forms a resistive bridge circuit which is connected to an instrumentation amplifier with a gain of 1000. This removes any common mode signals coupled to the patient from the surrounding environment. The signal is then low pass
filtered and boosted a further 2.5 times, then passed through the isolation amplifier to the output.

The microphone circuit uses an Automatic Gain Control amplifier at the input. The output from this is amplified a further ten times before passing through another output isolator. The overall frequency response of the channel is 30Hz – 1.2Khz.

The amplification for the Vibration stage is an AC coupled operational amplifier with a gain of 1000. Again the output is isolated.

The respiration circuit uses a standard impedance strap consisting of a wire stitched to an expandable cloth band. As the wire ring is deformed the impedance changes thus indicating expansion and contraction of the chest cavity. The changes are extremely small as is the impedance of the band. To make the most of available impedance a high excitation frequency is used. The band is connected in a balanced bridge arrangement, with a 10Khz excitation frequency supplying it. This frequency is as high as possible whilst staying within the instrumentation amplifiers Gain Bandwidth Product (GBP) specification. A signal generator chip is used to provide 10KHz at 5v. The sine wave was buffered with an op-amp to drive the belt. Belt impedance at 10Khz is 3.14ohms ( 50uH).

After initial amplification the modulated 10Khz signal is precision rectified and smoothed to give a DC level representing belt impedance. Offset adjustment is provided to allow zeroing of the signal by the operator, once the belt is attached to the patient. The output signal is then isolated before passing into the ADC (Analog to Digital Converter) card.

A surface mount technology, printed circuit board was designed and built to provide a small, robust and lightweight interface between transducers and ADC card.
2.2 Software Design

Software for capturing, displaying, recording and analyzing data was written using Borland’s Windows development tool, Delphi 5.

The software has four basic windows for data capture, display of recorded results and two trace analysis windows.

The capture window controls the collection of data over a specified period which is user selectable. This window controls both PCMCIA cards for simultaneous data and video capture. The ADC card comes with universal driver software routines to facilitate control of the card and data stream. Only particular sample rates can be selected as per the hardware manual specifications. The minimum sample frequency attainable is 622Hz.

When a recording session is in progress the waveforms and Video are simultaneously updated in the windows shown.

Optimum frame rate for the video capture was found to be 5 frames per second. Faster than this caused frames to be dropped thus producing capture time drift resulting in out of sync pictures with data signals during playback.
Once the data has been captured it can be reviewed in the window shown below:

In this window the data from the whole test can be studied. The test can be played through in real time or sections can be jumped to and frozen for investigation. If actual pressures and waveform measurements need to be carried out then the analysis window can be entered, and is shown below.

Here cursors can be dragged across to enable amplitude or frequency measurements. The display gain can also be increased to allow viewing of small signals. Using the scroll bar at the top of the page, the data segment time base can be varied to enable viewing of complete bursts. The user can
then select a portion of the data to import into the automatic analysis window via the use of the two cursors.

The analysis result page can be seen below:

![Analysis Results](image)

On entry to the window an automarking algorithm places marks on each suck trough. The user can adjust the amplitude of deflection that is marked enabling fine tuning of the automarking process. Calculations are made for each of the parameters in the results tables, with the exception of the swallow count which is entered by the user. Automatic analysis ensures reproducibility between studies and enables data analysis to be carried out very swiftly.

### 3 Procedure

Assessments can be carried out in the infant’s home, on the ward or in the clinic as part of their clinical evaluation, ideally at a time that coincides with a feed. Feeding can be carried out as normal from the bottle provided, with the microphone and vibration transducer taped to the throat, and the respiration belt around the chest.
The feeding liquid used is a 5% glucose solution, this eliminates any variability induced by other liquids such as taste, texture and fat content, which the literature suggests may affect performance.

The length of the assessment can be up to 20 minutes, typically a five minute period is used. This helps to keep the data collected to a manageable size, as the video clips consume large amounts of computer memory.

4 Results / Case Study

After obtaining informed consent, a healthy term infant aged three months was assessed at a routine feed time. All other aspects of his feeding routine were adhered to. He was fed his standard formula, using a teat type he was familiar with and was fed by his mother. The parent reported that his behavior did not vary from that of his routine and she did not feel that he was distressed by the procedure. His state of alertness before, during and after the assessment was coded according to Brazelton's descriptive list of states of alertness (Brazelton, 1984) : "State 5 ; Active alert".

A certified user assessed his oral motor skills, with the Neonatal Oral Motor assessment scale as "Normal".

Results obtained with the system are consistent with those reported in the literature. Early in the feed he used a suck/swallow/breathe ration of 1:1:1. As the feed progressed this changed to a ratio of 2 or 3:1:1.( Wolff, 1968; Mathew, 1986; Shivpuri et al, 1983; Weber et al, 1986). Pressures generated within the bottle varied from 15.4mm H₂O to 18.71 mmH₂O.

5 Discussion

Preliminary results obtained from the data collection system are consistent with those presented in the literature with regards to suck/swallow/breathe ratios and co-ordination. The system has the added advantage of allowing
simultaneous video capture of the feed. This is useful in facilitating accurate assessment of the feeding process and ensures that inappropriate conclusions are not drawn where waveforms may show artefacts caused by struggling or transducer movement. It also allows the use of observational, behavioural and feeding scales to be used on the same feed sample. The combination of objective information obtained with the system, and the use of standardized rating scales provide additional information about the feeding process. As the system is a non-invasive assessment tool it also has the advantage that it can be repeated regularly and can be used to assess an entire feed, unlike other procedures which assess for a short period.

Future developments for the system are planned to include post data collection analysis tools to statistically analyze the signals and data study in the frequency domain.
Appendix 6

Swallow Study
Auto-Mark
Algorithm
Flow Diagram
PATIENT / PARENT INFORMATION SHEET

We would like to ask permission to include your child in this project.

TITLE OF PROJECT

An investigation of the effects of pre-surgical orthodontics (baby plates) on feeding and speech in children with cleft lip / palate.

WHY IS THE STUDY BEING DONE?

Baby plates are small acrylic plates which, when fitted inside the mouth, block the cleft. The plate is worn continuously until surgical repair of the cleft palate (usually at approximately 6 months). Uncertainty exists over the use of baby plates. Some clinicians / researchers believe that they help with feeding and speech development. There is however no evidence to support this.

THE AIM OF THE STUDY

The aim of this study is to evaluate whether baby plates help in the early management of cleft lip / palate and if they benefit feeding and speech development.

HOW IS THE STUDY TO BE DONE?

General description

If you agree to participate in this study, your child will be assigned either to have a baby plate or not to have a baby plate. Rather like tossing a coin there will be a 50% chance of your child having a plate and 50 % chance of him/her not having a plate. Your child's usual medical and surgical management will continue normally. This will usually involve about 4 - 8 visits to the Orthodontic Department and Cleft Lip and Palate Clinic. For the
purposes of the project your child will be seen, either in Cleft Lip and Palate Clinic or at home, on 6 occasions (at day 5, 3 months, 6 months, 12 months, 18 months and 24 months) to evaluate his or her feeding and speech development. Wherever possible we will endeavour to link these to clinic appointments however there may be some extra visits required.

Details of what the study will involve

Routine management of your child’s feeding would involve regular interviews, and observation of your child feeding. In some cases an X-ray of your child’s swallowing may be taken. Assessment for the purposes of this project will be similar.

The feeding assessment is done in 2 or 3 parts.
1. A short interview about how your child is feeding
2. Your child will then be observed and videorecorded being fed. We will also measure the strength of your child’s sucking by having your child suck on a specially modified teat.
3. An X-ray of your child swallowing may also be taken if clinically appropriate.

Routine management of your child’s speech would generally involve assessment at 18 months with ongoing monitoring / treatment if required. However for the purposes of this project you child will have additional assessments as below.

From 6 months your child’s babbling and speech will be assessed. This will be done in 2 parts.
1. A short interview about your child’s speech development.
2. Observation of your child playing. This will be video recorded and babbling and speech samples analysed.

WHAT ARE THE RISKS AND DISCOMFORTS?

There may be some additional hospital visits required for assessments, but we hope that we will be able to co-ordinate your child’s assessments with clinical appointments.

WHAT ARE THE POTENTIAL BENEFITS?

If the use of baby plates is shown to be beneficial in the management of feeding and speech development in children with cleft lip and / or palate, they may be adopted for use with more children. If however they are not
shown to provide any benefit the time commitment associated with hospital visits (for parents) and costs to the NHS may be reduced and we can redirect the money and time to other ways of helping children with cleft lip and/or palate.

**WHAT OTHER TREATMENTS ARE AVAILABLE?**

There are no additional treatments routinely used in the management of these children at either Great Ormond Street Hospital or St Andrew’s Centre for Plastic Surgery.

**WHO WILL HAVE ACCESS TO THE CASE / RESEARCH RECORDS?**

Only the research team and a representative from the hospital’s Ethics Committee will have access to data collected.

**WHAT ARE THE ARRANGEMENTS FOR COMPENSATION?**

This project has been approved by an independent research ethics committee who believe that it is of minimal risk to you. However, research can carry unforeseen risks and we want you to be informed of your rights in the unlikely event that any harm should occur as a result of taking part in this study.

Only the work of academic staff on this project is covered by a compensation scheme which may apply in the event of any significant harm occurring to your child as a result of taking part in this study. Under this scheme it would not be necessary for you to prove fault. You also have the right to claim damages in a court of law. This would require you to prove fault on the part of the Hospital / Institute and / or any manufacturer involved.

**DO I HAVE TO TAKE PART IN THIS STUDY?**

No. If you decide that you do not wish your child to participate in this study the team’s management of your child will continue as routine. The decision about whether a baby plate is used will be dependent on the Cleft Lip and Palate Team’s decision.

If you decide, now or at a later stage, that you do not wish to participate in this research project, that is entirely your right, and will not in any way prejudice any present or future treatment.
WHO DO I SPEAK TO IF PROBLEMS ARISE?

If you have any complaints about the way in which this research project has been, or is being conducted, please, in the first instance, discuss them with the researcher. If the problems are not resolved, or you wish to comment in any other way, please contact the Chairman of the Research Ethics Committee, by post via the Research and Development Office, Institute of Child Health, 30 Guildford Street, London, WC1N 1EH, or if urgent by telephone on 0171 242 9789 ext. 2620, and the Committee administration will put you in contact with him.

DETAILS OF HOW TO CONTACT THE RESEARCHER

Ms Anthea Masarei
Research Fellow
Institute of Child Health
Honorary Senior Specialist Speech and Language Therapist
(Cleft Lip and Palate Team)

Correspondence via;
Speech and Language Therapy Department
Great Ormond Street Hospital NHS Trust
Great Ormond Street
London, WC1N 3JH

Telephone; 0171 405 9200 ext. 5043
Great Ormond Street Hospital for Children NHS Trust and Institute of Child Health Research Ethics Committee

Consent Form for PARENTS OR GUARDIANS of Children Participating in Research Studies

Title: An investigation of the effects of presurgical orthodontics (baby plates) on feeding and speech outcomes in children with cleft lip and/or palate.

NOTES FOR PARENTS OR GUARDIANS

1. Your child has been asked to take part in a research study. The person organising that study is responsible for explaining the project to you before you give consent.

2. Please ask the researcher any questions you may have about this project, before you decide whether you wish to participate.

3. If you decide, now or at any other stage, that you do not wish your child to participate in the research project, that is entirely your right, and if your child is a patient it will not in any way prejudice any present or future treatment.

4. You will be given an information sheet which describes the research project. This information sheet is for you to keep and refer to. Please read it carefully.

5. If you have any complaints about the way in which this research project has been or is being conducted, please, in the first instance, discuss them with the researcher. If the problems are not resolved, or you wish to comment in any other way, please contact the Chairman of the Research Ethics Committee, by post via The Research and Development Office, Institute of Child Health, 30 Guilford Street, London WC1N 1EH or if urgent, by telephone on 0171 242 9789 ext 2620 and the committee administration will put you in contact with him.

CONSENT

I/We _____________________________, being the parent(s)/guardian(s) of _____________________________ agree that the Research Project named above has been explained to me to my/our satisfaction, and I/We give permission for our child to take part in this study. I/We have read both the notes written above and the Information Sheet provided, and understand what the research study involves.

SIGNED (Parent(s)/Guardian(s)) DATE

SIGNED (Researcher) DATE
Appendix 9

PSO project: Guidelines for Initial Feeding Advice

The following are guidelines for advice that might be given to parents of babies recruited to the PSO and feeding project.

Baby's name: ________________________________

Baby's hospital number: ____________________

Date seen: _____________________________ (dd/mm/yyyy)

Advice given by: ________________________________

1. Discussion of problems babies with cleft palate +/- lip might have with their feeding:
   - weak and inefficient sucking
   - extended length feeds
   - fatigue
   - ingestion of air with increased need for burping

2. Discussion of feeding management strategies
   - "squeezy" bottles why and how
   - NUK teat (lip seal ...)
   - placement of teat in baby's mouth
   - adaptations to teat that may be useful e.g. extra holes, holes in side of teat ....
   - burping
   - positioning

3. Discussion re breast feeding (while not actively discouraged, need to monitor baby's growth to ensure that baby is getting enough milk; can be used in conjunction with bottle feeds for experience and bonding)

4. Where / how to get additional bottles / teats... (CLAPA)
Appendix 10

PSO project: Additional Feeding Advice Sessions
(Speech and Language Therapist / Clinical Nurse Specialist)

Baby’s name: ____________________________________________

Baby’s hospital number: __________________________________

Date seen: _____________________________________________

Problems:
1. ______________________________________________________

2. ______________________________________________________

3. ______________________________________________________

4. ______________________________________________________

Advice given / intervention:
1. ______________________________________________________

2. ______________________________________________________

3. ______________________________________________________

4. ______________________________________________________

Follow-up: _____________________________________________

Signed: _______________________________________________
PSO project: Guidelines for Maxillofacial / Dental Review
Appointments

All babies will be seen for these appointments. Although there will be differences in the content of these appointments related to whether the baby has a plate or not, we must aim to ensure some consistency in the appointments. *Italicised areas are specific to babies with PSO.*

Suggested content;

1. How is baby generally?

2. Intra-oral examination:
   - any areas of redness / irritation / ulceration
   - any infection or thrush
   - *does plate fit securely*
   - *any rubbing?*
   - *any adjustments to plate required?*

   .................................................................if yes please document

3. Any problems with feeding?
   ............................................if yes please advise Anthea Masarei for follow-up

*Record forms for progress notes attached.*
PSO Project; Maxillofacial / Dental Review Appointment Progress Notes

Baby's name: ________________________________

Baby's hospital number: ________________________________

<table>
<thead>
<tr>
<th>Appointment 1. (Impression taken)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Date of appointment:</td>
</tr>
<tr>
<td>Any problems:</td>
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<tr>
<td>Advice given / intervention:</td>
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<td>Signed:</td>
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</table>
**Appointment 2. (fitting of plate if appropriate)**

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<td>Signed:</td>
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**Appointment 3. (review)**

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<tr>
<td>Advice given / intervention:</td>
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<tr>
<td>Signed:</td>
<td></td>
</tr>
</tbody>
</table>
Appointment 4. (review)

Date of appointment: ________________________________

Any problems: ______________________________________
____________________________________________________
____________________________________________________
____________________________________________________

Advice given / intervention: __________________________
____________________________________________________
____________________________________________________
____________________________________________________

Signed: ____________________________________________

Appointment 5. (review)

Date of appointment: ________________________________

Any problems: ______________________________________
____________________________________________________
____________________________________________________
____________________________________________________

Advice given / intervention: __________________________
____________________________________________________
____________________________________________________
____________________________________________________

Signed: ____________________________________________
Appointment 6. (review)

Date of appointment: __________________________

Any problems: 

____________________________________________________________________________________

Advice given / intervention: 

____________________________________________________________________________________

Signed: ________________________________

Appointment 7. (review)

Date of appointment: __________________________

Any problems: 

____________________________________________________________________________________

Advice given / intervention: 

____________________________________________________________________________________

Signed: ________________________________
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<th><strong>Appointment Review</strong></th>
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<td><strong>Date of appointment:</strong></td>
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<tr>
<td><strong>Advice given / intervention:</strong></td>
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<tr>
<td><strong>Signed:</strong></td>
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</table>

<table>
<thead>
<tr>
<th><strong>Appointment Review</strong></th>
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<tbody>
<tr>
<td><strong>Date of appointment:</strong></td>
</tr>
<tr>
<td><strong>Any problems:</strong></td>
</tr>
<tr>
<td><strong>Advice given / intervention:</strong></td>
</tr>
<tr>
<td><strong>Signed:</strong></td>
</tr>
</tbody>
</table>
## NOMAS®

Neonatal Oral-Motor Assessment Scale (NOMAS)

Copyright © 1990 Marjorie Meyer Palmer

<table>
<thead>
<tr>
<th>Jaw</th>
<th>Normal</th>
<th>Disorganization</th>
<th>Dysfunction</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>consistent degree of jaw depression</td>
<td>inconsistent degree of jaw depression</td>
<td>excessively wide excursion that interrupt the intra-oral seal on the nipple</td>
</tr>
<tr>
<td></td>
<td>rhythymtical excursions</td>
<td>arrhythmical jaw movements</td>
<td>minimal excursion; clenching</td>
</tr>
<tr>
<td></td>
<td>spontaneous jaw excursions occur upon tactile presentation of the nipple up to 30 minutes prior to a feed</td>
<td>difficulty initiating movements: inability to latch on</td>
<td>asymmetry, lateral jaw deviation</td>
</tr>
<tr>
<td></td>
<td>jaw movement occurs at the rate of approximately one per second (1/2 the rate of NNS)</td>
<td>small, tremor-like start-up movements noted</td>
<td>absence of movement (% of time)</td>
</tr>
<tr>
<td></td>
<td>sufficient closure on the nipple during the expression phase to express fluid from the nipple</td>
<td>does not respond to initial cue of nipple until jiggled</td>
<td>lack of rate change between NNS and NS (NNS = 2/sec; NS = 1/sec)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Tongue</th>
<th>Normal</th>
<th>Disorganization</th>
<th>Dysfunction</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>cupped tongue configuration (tongue groove) maintained during sucking</td>
<td>excessive protrusion beyond labial border during extension phase of sucking without interrupting sucking rhythm</td>
<td>flaccid, flattened with absent tongue groove</td>
</tr>
<tr>
<td></td>
<td>extension-elevation-retraction movements occur in anterior-posterior direction</td>
<td>arrhythmical movements</td>
<td>retracted; humped and pulled back into oro-pharynx</td>
</tr>
<tr>
<td></td>
<td>rhythymtical movements</td>
<td>unable to sustain suckle pattern for two minutes due to: habituation</td>
<td>asymmetry, lateral tongue deviation</td>
</tr>
<tr>
<td></td>
<td>movements occur at the rate of one per second</td>
<td>poor respiration</td>
<td>excessive protrusion beyond labial border before/after nipple insertion with out/down movement</td>
</tr>
<tr>
<td></td>
<td>liquid is sucked efficiently into the oro-pharynx for swallow</td>
<td>fatigue</td>
<td>absence of movement (% of time)</td>
</tr>
</tbody>
</table>

Summary and impression:

Recommendations:

__________________________
Certified Examiner

Certificate #
## SOMA - OMC category - Purée

<table>
<thead>
<tr>
<th>Name:</th>
<th>Examiner:</th>
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<tbody>
<tr>
<td>Date of birth:</td>
<td>Diagnosis</td>
</tr>
<tr>
<td>Date of assessment:</td>
<td></td>
</tr>
<tr>
<td>Age:</td>
<td></td>
</tr>
<tr>
<td>Body position:</td>
<td></td>
</tr>
<tr>
<td>Support required:</td>
<td>Head position:</td>
</tr>
</tbody>
</table>

### Purée

<table>
<thead>
<tr>
<th>Fromage frais</th>
<th>mousse</th>
<th>pureed fruit</th>
<th>other</th>
<th>(Circle choice)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Non rateable</td>
<td>Rateable</td>
<td>Yes</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>React 1</td>
<td>head orientation to spoon</td>
<td>y</td>
<td>n</td>
<td></td>
</tr>
<tr>
<td>Sequence 1</td>
<td>smooth rhythmic sequence</td>
<td>y</td>
<td>n</td>
<td></td>
</tr>
<tr>
<td>Lip 1</td>
<td>lower lip draws inwards around spoon</td>
<td>y</td>
<td>n</td>
<td></td>
</tr>
<tr>
<td>Lip 2</td>
<td>upper lip removes food from spoon</td>
<td>y</td>
<td>n</td>
<td></td>
</tr>
<tr>
<td>Lip 3</td>
<td>lower/upper lip assist in cleaning</td>
<td>y</td>
<td>n</td>
<td></td>
</tr>
<tr>
<td>Lip 11</td>
<td>lower lip active during suck/munch/chew</td>
<td>y</td>
<td>n</td>
<td></td>
</tr>
<tr>
<td>Tongue 11</td>
<td>consistent/considerable protrusion</td>
<td>y</td>
<td>n</td>
<td></td>
</tr>
<tr>
<td>Tongue 12</td>
<td>protrusion beyond incisors</td>
<td>y</td>
<td>n</td>
<td></td>
</tr>
<tr>
<td>Jaw 1</td>
<td>graded jaw opening</td>
<td>y</td>
<td>n</td>
<td></td>
</tr>
</tbody>
</table>

**Sum of shaded boxes**

**Cutting score:**
- > 3 indicates oral motor dysfunction
- < 3 normal oral motor function
POSTURE AND SEATING:

No oral motor assessment would be complete without consideration being paid to the child's position whilst eating and drinking. However any assessment of posture is beyond the scope of the SOMA and the authors recommend that examiners use the SOMA in association with a recognised assessment of seating/posture.

The examiner always elects to administer the SOMA in the child's optimum seating position to provide the infant with the most appropriate and adequate seating support. However facilities depend entirely on the family or clinic resources available and to an extent on the infant's co-operation.

General ratings of body and head position are therefore made during the administration as indicated below, however, this aspect of the SOMA has not been standardised in any way.

Body positioning:

Body position during each OMC category is marked in the place provided on the score sheets and rated as follows:

1. **Supine** the infant is positioned on its back.
2. **Prone** the infant is positioned on its front.
3. **Supine/prone with elevated head** the infant is in either of the above two positions with the head raised. This may be with the support of an adult or cushions, etc. note type of support.
4. **Side lying** the infant is lying on either side - note if support given.
5. **Semi-sitting with support** the infant is sitting in a semi-upright position with full trunk support. This may be provided by either an adult, a chair or by cushions.
6. **Sitting upright with trunk support** the child is seated in an upright position but with the trunk supported by cushions, a harness, padding, etc.
7. **Sitting upright with back support** the infant is seated in an upright position but with back support only. The child may be leaning back against the chair for support.
8. **Sitting upright without support**
9. Walking around

10. Special seating - describe

11. Other

HEAD POSITIONING (score only if abnormal)

Head position is rated during each OMC category and is marked in the place provided on the score sheets. Ratings are as follows:

1. Forward flexion - The child's head flops or is bent forward in forward flexion onto the chest.

2. Side flexion - The child's head flops or is bent sideways to the right or to the left.

3. Extension backwards - The child's head is extended backwards or flops backwards.

4. Extension forwards - The child's head is extended forwards away from the body with the chin pointing upwards.

DEGREE OF HEAD SUPPORT PROVIDED

1. None - The examiner does not need to assist the child in any way with head support during the assessment.

2. Partial - The examiner needs to provide the child with some degree of support such as steadying the child's head momentarily to enable the child to bite or begin drinking. However this help is not continuous.

3. Moderate - The examiner provides the child with some degree of support for at least half of the time during the trial. The child may need assistance with head control or jaw support etc.

4. Total support - Without the examiner's support he child would not be
Oral Motor Challenge Category - Puree

Rating the discrete oral motor behaviours

There are nine discrete oral motor behaviours which make up the screening index for the OMC category, purée. A description and definition of each behaviour is given below.

React1 - HEAD ORIENTATION TO FOOD
The infant moves his/her head, body or trunk towards the spoon or drink. This movement may involve trunk or head extension or a variety of other movements. This movement should be carefully checked in slow motion on the video if it is not obvious. In children with neuro-motor impairments the movement is often very subtle.
Score Yes if present. This a normal response.

Sequence 1 - SMOOTH SEQUENCE
A smooth sequence of at least 3 or more suck swallows, munching actions or chewing actions are seen. There are no co-ordination difficulties with integrating suck swallow or chew/munch-swallow, etc.
Score yes if present. This a normal response.

Lip1 - LOWER LIPS DRAWS INWARDS AROUND SPOON
Susan Evans - Morris, a clinician and researcher, describes the ability for the lower lip to draw inwards around the spoon as part of the process of separation of movement and the development of skill and precision. That is, the lips no longer move in unison with the jaw or tongue and the lower lip can mould around the spoon independently and draw inwards to help keep food in the mouth when the spoon is withdrawn.
Score yes if present. This a normal response.

Lip2 - UPPER LIP ACTIVELY REMOVES FOOD FROM THE SPOON
The upper lip is able to move forwards and downwards to help clean the spoon of food or remove food from the spoon. The lips may mould completely around the spoon or the midpoint of the upper lip may make contact only.
Score yes if present. This a normal response.

Lip3 - LOWER/UPPER LIP ASSIST IN CLEANING
The lower and upper lips assist to clean food from lips. For example, the lower lip is moved against the upper teeth or gums or upper lip in order to clean and retrieve small pieces of food.
Score yes if present. This a normal response.
Lip 11 LOWER LIP ACTIVE DURING SUCKING/ CHEWING/MUNCHING
The lower lip is active during the sucking, munching or chewing sequence. Early in development this movement is not separated from the total movement patterns of the jaw and tongue, however, this separation takes place and the upper and lower lip can function independently. This movement may be to help in the cleaning process such as moving down in order to clean with the lower lip or assists in keeping food within the mouth and preventing spillage.
Score yes if present. This a normal response.

Tongue 11 CONSISTENT/CONSIDERABLE PROTRUSION
The tongue protrudes consistently throughout the sucking/munching or chewing sequence (more than 50% of the time) representing a more infantile pattern of extension/retraction. The tongue may protrude to different degrees, either beyond the lower dentition or beyond the lower lip.
Score yes if present. This is an abnormal response

Tongue 12 TONGUE PROTRUSION BEYOND THE INCISORS
The tongue protrudes between the incisors but not beyond the lower lip.
Score yes if present. This is an abnormal response.

Jaw 1 GRADED JAW OPENING TO ACCEPT SPOON
The jaw is opened sufficiently to accept a loaded spoon. There is neither too wide or too narrow an excursion. In Cerebral Palsy and young babies often the opening is exaggerated or the jaw excursion may be too narrow and not allow placement of the spoon.
Score yes if present. This a normal response.
Great Ormond Street Hospital
for Children NHS Trust
and the Institute of Child Health
Speech and Language Therapy Department

"Dysphagia Swallow" / Videofluoroscopy Report
For use in "Baby Plate" (Anthea Masarei et al) project

Patient name / code:__________________________________________

Patient status: _____________________________________________

Aversive behaviours:________________________________________

Airway / chest status:

☐ no problems ☐ congestion/ chest infection ☐ other __________

☐ Oxygen dependent via ______________________________________

☐ nasopharyngeal airway ☐ tracheostomy (+/- speaking valve ________)

☐ ventilator dependent _____________________________

Seating:

☐ Tumble form ☐ supine ☐ other _____________________________

Presenter of food:

☐ parent/carer ☐ speech and language therapist ☐ nurse
Utensils:

- bottle
- teat type

Imaging views taken:

- lateral
- other

Texture:

- liquid
- other

Summary of Results:

<table>
<thead>
<tr>
<th>ORAL STAGE</th>
<th>NORMAL</th>
<th>ABNORMAL</th>
</tr>
</thead>
<tbody>
<tr>
<td>lip closure</td>
<td></td>
<td></td>
</tr>
<tr>
<td>tongue configuration during sucking</td>
<td></td>
<td></td>
</tr>
<tr>
<td>bolus formation / control</td>
<td></td>
<td></td>
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<tr>
<td>oral residue</td>
<td></td>
<td></td>
</tr>
<tr>
<td>nasal regurgitation</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

** PHARYNGEAL STAGE **

- point at which swallow is triggered
- laryngeal penetration / aspiration **
- post swallow / sucking burst residue
- cricopharyngeal dysfunction

*Descriptions of normal swallow events definitions attached*

** Rosenhek aspiration penetration scale (attached)**
### Rating Scale; description of normal swallow behaviours

<table>
<thead>
<tr>
<th>Oral and Pharyngeal behaviours/events</th>
<th>Descriptors of normal</th>
</tr>
</thead>
<tbody>
<tr>
<td>Lip closure</td>
<td>No loss or dribbling of fluid</td>
</tr>
<tr>
<td>Tongue configuration</td>
<td>Rhythmical stripping of the tongue during sucking</td>
</tr>
<tr>
<td>Compression of teat</td>
<td>Compression of the teat during sucking</td>
</tr>
<tr>
<td>Bolus formation</td>
<td>Bolus well controlled and propelled posteriorly</td>
</tr>
<tr>
<td>Nasal regurgitation</td>
<td>Bolus does not enter nasopharynx</td>
</tr>
<tr>
<td>Triggering of the swallow</td>
<td>Swallow is triggered at the level when the bolus head passes the tongue base or at the level of the valleculae</td>
</tr>
<tr>
<td>Pharyngeal residue</td>
<td>Pharynx is cleared other than trace coating</td>
</tr>
<tr>
<td>Cricopharyngeal function</td>
<td>Well timed and complete opening</td>
</tr>
</tbody>
</table>
**Laryngeal penetration-aspiration scale** *(Rosenbek et al., 1996)*

<table>
<thead>
<tr>
<th>Rating</th>
<th>Description</th>
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<tbody>
<tr>
<td>0</td>
<td>Material does not enter the airway</td>
</tr>
<tr>
<td>1</td>
<td>Material enters the airway, remains above the vocal folds and is ejected from the airway</td>
</tr>
<tr>
<td>2</td>
<td>Material enters the airway, remains above the vocal folds and is not ejected from the airway</td>
</tr>
<tr>
<td>3</td>
<td>Material enters the airway, contacts the vocal folds and is ejected from the airway</td>
</tr>
<tr>
<td>4</td>
<td>Material enters the airway, contacts the vocal folds and is not ejected from the airway</td>
</tr>
<tr>
<td>5</td>
<td>Material enters the airway, passes below the vocal folds and is ejected into the larynx or out of the airway</td>
</tr>
<tr>
<td>6</td>
<td>Material enters the airway, passes below the vocal folds and is not ejected from the trachea despite effort</td>
</tr>
<tr>
<td>7</td>
<td>Material enters the airway, passes below the vocal folds and no effort is made to eject</td>
</tr>
</tbody>
</table>
PSO project: Additional Feeding Advice (given by Feeding Specialist Speech and Language Therapist)

Baby's name: ________________________________

Baby's hospital number: ____________________

Date seen: ____________________ (ddmmyyyy)

Advice given by: ________________________________

1. Information about normal breast / bottle feeding (diagram attached)
   - Sucking; positive pressure generation or compression
     - negative pressure generation or suction
   - swallow is then triggered as the milk reaches the back of the throat
   - airway closes and food is sent through the pharynx (throat) into the oesophagus

2. Discussion as to why babies with cleft palates may experience difficulties (pointing out that generally these babies only have difficulties with)
   - weak sucking; reduced / altered opposing surface
     - inability to generate negative pressure
   - perhaps difficulty co-ordinating breathing with sucking and swallowing
   - fatigue

3. Revision and / or clarification re feeding equipment given (Mead Johnson with NUK teat) why and how …

4. Revision re teat placement (particularly important in non cleft babies)

5. Discussion re need to adapt teat
Great Ormond Street Hospital
for Children NHS Trust
and the Institute of Child Health
Speech and Language Therapy Department

“Use of PSO in feeding management of children with Cleft Lip and Palate” project

Data collection forms

*General Health Questionnaire (Demographics)*

1. Date of data collection (ddmmyyyy)
2. Baby’s name: ____________________________
3. Baby’s hospital number: __________________
4. Baby’s DOB: (ddmmyyyy)
5. Sex: male (1), female (2)
6. Location: GOSH (1), St Andrew’s (2)
7. Mother’s name: __________________________
8. Mother’s DOB: (ddmmyyyy)
9. What level of education have you completed?
   - Left school prior to GCSEs (1)
   - GCSEs (2)
   - A levels (3)
   - NVQ / certificate (4)
   - University (5)
   - Post graduate (6)
10. Do you undertake any paid employment? Yes (1), No (0)
11. What is the title of your job? __________________
12. What does your job involve? __________________

- 390 -
13. Marital status:  married (1)  
                  living together (2)  
                  single (3)  
                  widowed, divorced or separated (4)  

14. Husband's / partner's name: ________________________  

15. Husband's / partner's DOB: (ddmmyyyy) ___________  

16. What level of education did he complete?  
                  Left school prior to GCSEs (1)  
                  GCSEs (2)  
                  A levels (3)  
                  VQ / certificate (4)  
                  University (5)  
                  Post graduate (6)  

17. Does your partner undertake any paid employment?  
               Yes (1), No (0)  

18. What is the title of his job? ____________________  

19. What does his job involve? ____________________  

20. Birth order for this child:  
                  1 2 3 4 5 .......  

(coding as per Registrar General's scale)
“Use of PSO in feeding management of children with Cleft Lip and Palate” project

Data collection forms

*General Health Questionnaire 1 (Medical) (to be collected from medical notes and clarified with mother as appropriate)*

| Baby’s hospital number: ___________________________ |
| Date of data collection (ddmmyyyy) |

**Mother’s health during pregnancy**

1. Mother’s health during pregnancy:

| generally well, no real problems (1) |
| unwell with “expected” pregnancy difficulties for short periods of time but managed by GP (2) |
| unwell with more than expected difficulties associated with pregnancy (3) |
| unwell requiring “specialist management” (4) |
| hospitalisation required (5) |
| other (6) |

2. Any problem’s with the following during pregnancy?

| infection Yes (1), No (0) |
| toxemia Yes (1), No (0) |
| bleeding Yes (1), No (0) |
| thyroid disease Yes (1), No (0) |
| polyhydramnios Yes (1), No (0) |
| other Yes (1), No (0) |

3. Smoked during pregnancy? Yes (1), No (0)

4. How much? ___________________________ / week
5. Medication / drugs during pregnancy?  
   Yes (1), No (0)  

6. If yes, what medications and how much?  

7. Units of alcohol per week during pregnancy  
   _____________________________________ units per week  

Father's health  

8. Father's health:  

   generally well (1)  
   occasionally unwell but no visits to GP required (2)  
   one or more visits to GP required (3)  
   regular visits to GP for ongoing illnesses (4)  
   specialist management or hospitalisation required (5)  

Baby's delivery  

9. Gestational age: __________________________ weeks  

10. Birth weight: ___________________________ kg  

11. Was induction required?  
   Yes (1), No (0)  

12. Any assistance e.g. forceps/suction required?  
   (includes breech)  
   Yes (1), No (0)  

13. Caesarean section required?  
   Yes (1), No (0)  

14. Pain relief required?  
   Yes (1), No (0)  

15. Length of delivery.  
   ___________________________ hours  

16. Any foetal distress?  
   Yes (1), No (0)  
   if yes outline:  
   _____________________________________  
   (to be coded at a later date)
17. APGAR scores:
   1 minute
   5 minutes

18. Admission to the neonatal unit? Yes (1), No (0)

19. Length of admission to neonatal unit; ___________ days

20. Resuscitation required? Yes (1), No (0)

21. If required, how long for? ________________ minutes

22. Ventilation required? Yes (1), No (0)

23. If required, how long for? ________________ days

24. Is ventilation ongoing? Yes (1), No (0)

25. Does baby have any other medical problems as below:
   jaundice Yes (1), No (0)
   cardiac Yes (1), No (0)
   respiratory Yes (1), No (0)
   gastrointestinal Yes (1), No (0)
   neurological Yes (1), No (0)
   other Yes (1), No (0)

26. Cleft type:
   □ isolated cleft of the soft palate and 2/3 of the hard palate Yes (1), No (0)
   □ complete unilateral lip and palate (right) Yes (1), No (0)
   □ complete unilateral lip and palate (left) Yes (1), No (0)

27. Syndrome diagnosed? Yes (1), No (0)
   (to be coded at a later date)
"Use of PSO in feeding management of children with Cleft Lip and Palate" project

Data collection forms

Feeding Questionnaire (Data collection point 1)
(this information is collected by interview)

Baby's hospital number: ____________________________

Date of data collection (ddmmyyyy)

General

1. How are you feeding your baby?
   - Breast
   - Bottle
   - Feeding tube

2. How often are you feeding your baby?
   - 4 hourly
   - 3 hourly
   - 2 hourly
   - on demand ________________

3. What are you feeding your baby?
   - expressed breast milk
   - standard formula
   - “special” formula
   - other

4. Are you adding anything to your baby’s feeds?
   - Yes (1), No (0)
5. What are you adding?
- Rice cereal
- Other solids
- Supplements
- Medication
- Flavours e.g. sugar

Bottle feeding

6. What type of bottle are you currently feeding your baby with?
- Mead Johnson
- Soft Plas
- Chicco
- Standard
- Rosti
- Haberman feeder
- Other e.g. cup and spoon

7. What type of teat are you using?
- NUK orthodontic, vented (size 2)
- NUK orthodontic, vented (size 1)
- Standard term / newborn latex teat e.g. Cow and Gate
- Standard term / newborn silicone teat
- Premature teats
- Extended length teat e.g. Lambs teat
- NUK Cleft Palate teat
- Cross cut teat
- Other

8. Have you made any adaptations to your baby's teat?
- Enlarged hole
- Cross cut teat
- Created a “new” hole on the side
- Use the teat upside down
- Other

9. How much feed does your baby take at each feed?

10. How long does it take your baby to take this amount?

Breast feeding

11. Have you tried breast-feeding your baby?  Yes (1), No (0)

12. If you have not tried breast feeding why did you decide not to?

<table>
<thead>
<tr>
<th>Reason</th>
<th>Yes (1), No (0)</th>
</tr>
</thead>
<tbody>
<tr>
<td>mother had inverted or painful nipples</td>
<td></td>
</tr>
<tr>
<td>embarrassment</td>
<td></td>
</tr>
<tr>
<td>unable to tell how much feed baby is taking</td>
<td></td>
</tr>
<tr>
<td>baby has been unwell</td>
<td></td>
</tr>
<tr>
<td>advised by CLAPC team that breast feeding is generally not successful with CLAP babies</td>
<td></td>
</tr>
<tr>
<td>decided after speaking to other mothers of CLAP babies</td>
<td></td>
</tr>
<tr>
<td>other</td>
<td></td>
</tr>
</tbody>
</table>

13. If you have tried breast feeding are you continuing with it or are you planning to do so?  Yes (1), No (0)

14. If you are not continuing breast feeding why not?

<table>
<thead>
<tr>
<th>Reason</th>
<th>Yes (1), No (0)</th>
</tr>
</thead>
<tbody>
<tr>
<td>baby is/was unable to &quot;latch onto breast&quot;</td>
<td></td>
</tr>
<tr>
<td>baby is/was unable to generate or maintain adequate suck</td>
<td></td>
</tr>
<tr>
<td>mother had inverted or painful nipples</td>
<td></td>
</tr>
<tr>
<td>embarrassment</td>
<td></td>
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<tr>
<td>decided after speaking to other mothers of CLAP babies</td>
<td></td>
</tr>
<tr>
<td>other</td>
<td></td>
</tr>
</tbody>
</table>

15. How long do you breast-feed your baby for at each feed?  mins
Tube feeding

16. How long has or did your baby have a feeding tube for? ________________ days

17. Is tube feeding ongoing? Yes (1), No (0)

18. How much feed does your baby have through the tube at each feed? _____________________mls

Feeding observations

19. Have you noticed any feed in or coming from your baby’s nose during feeds? Yes (1), No (0)

20. How often do you notice feed in the nose or coming from your baby’s nose?
- occasionally Yes (1), No (0)
- once or twice a day Yes (1), No (0)
- at every feed Yes (1), No (0)

21. How much feed returns through the nose?
- trace amounts only Yes (1), No (0)
- about a thimble full Yes (1), No (0)
- about a dessertspoon full Yes (1), No (0)

22. Have you noticed your baby coughing, choking or gagging during feeds? Yes (1), No (0)

23. Have you noticed your baby’s breathing becoming “gurgley” during feeds? Yes (1), No (0)

24. Does your baby ever show signs of discomfort during feeds? e.g. crying, arching, drawing legs to chest Yes (1), No (0)

25. Does your baby vomit after feeds? Yes (1), No (0)

26. How often does this occur?
- rarely Yes (1), No (0)
- once or twice a day Yes (1), No (0)
- during or after every feed Yes (1), No (0)

27. Does your baby have any medicine for this? Yes (1), No (0)
28. What medicine is your baby having for this?

<table>
<thead>
<tr>
<th>Medicine</th>
<th>Yes (1), No (0)</th>
</tr>
</thead>
<tbody>
<tr>
<td>thickener added to feeds</td>
<td></td>
</tr>
<tr>
<td>Gaviscon</td>
<td></td>
</tr>
<tr>
<td>Ranitidine</td>
<td></td>
</tr>
<tr>
<td>Domperidone</td>
<td></td>
</tr>
<tr>
<td>Cisapride</td>
<td></td>
</tr>
<tr>
<td>Other</td>
<td></td>
</tr>
</tbody>
</table>

29. Does anyone other than yourself feed your baby?

Yes (1), No (0)

30. Who else feeds your baby?

<table>
<thead>
<tr>
<th>Feeder</th>
<th>Yes (1), No (0)</th>
</tr>
</thead>
<tbody>
<tr>
<td>husband / partner</td>
<td></td>
</tr>
<tr>
<td>other adult relative</td>
<td></td>
</tr>
<tr>
<td>sibling</td>
<td></td>
</tr>
<tr>
<td>carer</td>
<td></td>
</tr>
<tr>
<td>friend</td>
<td></td>
</tr>
<tr>
<td>nursing staff</td>
<td></td>
</tr>
<tr>
<td>Other</td>
<td></td>
</tr>
</tbody>
</table>
Great Ormond Street Hospital for Children NHS Trust
and the Institute of Child Health
Speech and Language Therapy Department

"Use of PSD in feeding management of children with Cleft Lip and Palate" project

Data collection forms

General Health Questionnaire (Data collection point 2, 3, 4, 5)
(this information is collected by interview)

Baby's hospital number: ________________________________

Date of data collection (ddmmyyyy)

Demographics

1. Has anything changed with regards to your homelife?
   Yes (1), No (0)

2. If yes what has changed?

☐ husband / partner has left

☐ husband / partner is no longer working

☐ mother has returned to work full time

☐ mother has returned to work part time

☐ other ________________________________

☐ Yes (1), No (0)
3. How has your general health been since the birth of _____________?

☐ generally well
☐ occasionally unwell but no visits to GP required
☐ one or more visits to GP required
☐ regular visits to GP for ongoing illness
☐ specialist management or hospitalisation required

4. How has your husband's / partner's health been?

☐ generally well
☐ occasionally unwell but no visits to GP required
☐ one or more visits to GP required
☐ regular visits to GP for ongoing illness
☐ specialist management or hospitalisation required

5. How are your other children?

☐ generally well
☐ occasionally unwell but no visits to GP required
☐ one or more visits to GP required
☐ regular visits to GP for ongoing illness
☐ specialist management or hospitalisation required
Medical information

1. Has your baby had any medical problems since I last saw you?  
   (if yes outline below)  Yes (1), No (0)  
   cardiac  Yes (1), No (0)  
   respiratory  Yes (1), No (0)  
   gastrointestinal  Yes (1), No (0)  
   neurological  Yes (1), No (0)  
   other  Yes (1), No (0)  

2. Syndrome diagnosed?  Yes (1), No (0)  
   (to be coded at a later date)  

3. What medical interventions has your child had?  
   (details to be confirmed from medical notes and results to be coded at a later date)  

4. Other information:  

5. Medications:  

- 402 -
Great Ormond Street Hospital
for Children NHS Trust
and the Institute of Child Health
Speech and Language Therapy Department

"Use of PSO in feeding management of children with Cleft Lip and Palate" project

Data collection forms

**Anthropometric data (Data collection point 1)**

Baby's hospital number: __________________________

Date of data collection (ddmmyyyy)

1. Length: ________________________________ cm
   (SD: ________________________________ )

2. Weight: ________________________________ kg
   (SD: ________________________________ )

3. Head circumference: ________________________________ cm
   (SD: ________________________________ )
### Data collection forms

**Feeding Questionnaire (Data collection point 2)**

*This information is collected by interview*

<table>
<thead>
<tr>
<th>Baby’s hospital number: __________________________</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Date of data collection <em>(ddmmyyyy)</em></td>
<td></td>
</tr>
</tbody>
</table>

**General**

1. How are you feeding your baby?
   - [ ] Breast          Yes (1), No (0)
   - [ ] Bottle          Yes (1), No (0)
   - [ ] Feeding tube    Yes (1), No (0)

2. How often are you feeding your baby?
   - [ ] 6 hourly        Yes (1), No (0)
   - [ ] 4 hourly        Yes (1), No (0)
   - [ ] 3 hourly        Yes (1), No (0)
   - [ ] 2 hourly        Yes (1), No (0)
   - [ ] on demand       Yes (1), No (0)

3. What are you feeding your baby?
   - [ ] expressed breast milk Yes (1), No (0)
   - [ ] standard formula   Yes (1), No (0)
   - [ ] “special” formula  Yes (1), No (0)
   - [ ] other             Yes (1), No (0)

4. Are you adding anything to your baby’s feeds? Yes (1), No (0)
5. What are you adding?
- Rice cereal: Yes (1), No (0)
- Other solids: Yes (1), No (0)
- Supplements: Yes (1), No (0)
- Medication: Yes (1), No (0)
- Flavours e.g. sugar: Yes (1), No (0)

Bottle Feeding

6. What type of bottle are you currently feeding your baby with?
- Mead Johnson: Yes (1), No (0)
- Soft Plas: Yes (1), No (0)
- Chicco: Yes (1), No (0)
- Standard: Yes (1), No (0)
- Rosti: Yes (1), No (0)
- Haberman feeder: Yes (1), No (0)
- Other e.g. cup and spoon: Yes (1), No (0)

7. What type of teat are you using?
- NUK orthodontic, vented (size 2): Yes (1), No (0)
- NUK orthodontic, vented (size 1): Yes (1), No (0)
- Standard term / newborn latex teat: Yes (1), No (0)
- Standard term / newborn silicone teat: Yes (1), No (0)
- Premature teats: Yes (1), No (0)
- Extended length teat e.g. Lambs teat: Yes (1), No (0)
- NUK Cleft Palate teat: Yes (1), No (0)
- Cross cut teat: Yes (1), No (0)
- Other: Yes (1), No (0)

8. Have you made any adaptations to your baby’s teat?
- Enlarged hole: Yes (1), No (0)
- Cross cut teat: Yes (1), No (0)
- Created a “new” hole on the side: Yes (1), No (0)
- Use the teat upside down: Yes (1), No (0)
- Other: Yes (1), No (0)

9. How much feed does your baby take at each feed?
_______________________________ mls

10. How long does it take your baby to take this amount?
_______________________________ mins

Breast Feeding

11. Have you tried breast-feeding your baby?
Yes (1), No (0)
12. If you tried breast-feeding, how long did you try it for? ______________ days

13. If you have tried breast feeding are you continuing with it or are you planning to do so? Yes (1), No (0)

14. If you are not continuing breast feeding why not?
- baby is/ was unable to "latch onto breast" Yes (1), No (0)
- baby is/ was unable to generate or maintain adequate suck Yes (1), No (0)
- mother had inverted or painful nipples Yes (1), No (0)
- embarrassment Yes (1), No (0)
- unable to tell how much feed baby is taking Yes (1), No (0)
- baby has been unwell Yes (1), No (0)
- advised by CLAPC team that breast feeding is generally not successful with CLAP babies Yes (1), No (0)
- decided after speaking to other mothers of CLAP babies Yes (1), No (0)
- other ________________________________ Yes (1), No (0)

15. How long do you breast-feed your baby for at each feed? ______________ mins

16. How long has or did your baby have a feeding tube for? ______________ days

17. Is tube feeding ongoing? Yes (1), No (0)

18. How much feed does your baby have through the tube at each feed? ______________ mls

19. Have you noticed any feed in or coming from your baby's nose during feeds? Yes (1), No (0)

20. How often do you notice feed in the nose or coming from your baby's nose?
- occasionally Yes (1), No (0)
- once or twice a day Yes (1), No (0)
- at every feed Yes (1), No (0)
21. How much feed returns through the nose?
- □ trace amounts only
- □ about a thimble full
- □ about a dessertspoon full

22. Have you noticed your baby coughing, choking or gagging during feeds? Yes (1), No (0)

23. Have you noticed your baby's breathing becoming "gurglely" during feeds? Yes (1), No (0)

24. Does your baby ever show signs of discomfort during feeds? e.g. crying, arching, drawing legs to chest Yes (1), No (0)

25. Does your baby vomit after feeds? Yes (1), No (0)

26. How often does this occur?
- □ rarely
- □ once or twice a day
- □ during or after every feed

27. Does your baby have any medicine for this? Yes (1), No (0)

28. What medicine is your baby having for this?
- □ thickener added to feeds
- □ Gaviscon
- □ Ranitidine
- □ Domperidone
- □ Cisapride
- □ other _______________________________

29. Does your baby have a history of chest infections requiring antibiotic treatment? Yes (1), No (0)

30. Does anyone other than yourself feed your baby? Yes (1), No (0)

31. Who else feeds your baby?
- □ husband / partner
- □ other adult relative
- □ sibling
- □ carer
- □ friend
- □ nursing staff
- □ other _______________________________
Great Ormond Street Hospital for Children NHS Trust
and the Institute of Child Health
Speech and Language Therapy Department

"Use of PSO in feeding management of children with Cleft Lip and Palate" project

Data collection forms

**Anthropometric data (Data collection point 2, 3 and 5)**

<table>
<thead>
<tr>
<th>Baby's hospital number:</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Date of data collection (ddmmyyyy)</td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>1. Length: cm (SD: )</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>2. Weight: kg (SD: )</td>
<td></td>
</tr>
<tr>
<td>3. Head circumference: cm (SD: )</td>
<td></td>
</tr>
<tr>
<td>4. Mid arm circumference: cm (SD: )</td>
<td></td>
</tr>
</tbody>
</table>
Great Ormond Street Hospital for Children NHS Trust
and the Institute of Child Health
Speech and Language Therapy Department

"Use of PSO in feeding management of children with Cleft Lip and Palate" project

Data collection forms

Feeding Questionnaire (Data collection point 3)
(this information is collected by interview)

Baby’s hospital number: __________________________

Date of data collection (ddmmyyyy) __________________________

Milk feeds

1. What milk feed is your baby taking now?
   □ breast milk          Yes (1), No (0)
   □ expressed breast milk Yes (1), No (0)
   □ standard formula     Yes (1), No (0)
   □ “special” formula     Yes (1), No (0)
   □ other               Yes (1), No (0)

2. Are you adding anything to your baby’s milk feeds?
   Yes (1), No (0)

3. What are you adding?
   □ rice cereal          Yes (1), No (0)
   □ other solids         Yes (1), No (0)
   □ supplements          Yes (1), No (0)
   □ medication           Yes (1), No (0)
   □ flavours e.g. sugar   Yes (1), No (0)

4. How many milk feeds does your baby have each day?
   _______________ feeds

Appendix 23

- 409 -
**Bottle / cup feeding**

5. How is your baby taking milk / fluids now?
   - Mead Johnson Yes (1), No (0)
   - Soft Plas Yes (1), No (0)
   - Chicco Yes (1), No (0)
   - Standard Yes (1), No (0)
   - Rosti Yes (1), No (0)
   - Haberman feeder Yes (1), No (0)
   - training beaker Yes (1), No (0)
   - beaker or glass Yes (1), No (0)
   - other e.g. cup and spoon ______ Yes (1), No (0)

6. What teat are you currently using on your baby's bottle?
   - NUK orthodontic, vented (size 2) Yes (1), No (0)
   - NUK orthodontic, vented (size 1) Yes (1), No (0)
   - Standard term / newborn latex teat e.g. Cow and Gate Yes (1), No (0)
   - Standard term / newborn silicone teat Yes (1), No (0)
   - Premature teats Yes (1), No (0)
   - Extended length teat e.g. Lambs teat Yes (1), No (0)
   - NUK Cleft Palate teat Yes (1), No (0)
   - Cross cut teat Yes (1), No (0)
   - Other __________________________ Yes (1), No (0)

7. Have you made any adaptations to your baby's teat?
   - enlarged hole Yes (1), No (0)
   - cross cut teat Yes (1), No (0)
   - created a "new" hole on the side Yes (1), No (0)
   - use the teat upside down Yes (1), No (0)
   - other __________________________ Yes (1), No (0)

8. How many mls of milk does your baby have at each milk feed? __________________________ mls

9. How long does this take?
     ___________________________ mins

**Breast feeding**

10. Are you still breast feeding your baby? Yes (1), No (0)

11. How long do you breast-feed your baby for at each feed? ___________________________ mins

12. If you are not breast-feeding now, did you try breast-feeding your baby at any time? Yes (1), No (0)

13. If you tried breast-feeding, how long did you try it for?
14. Why did you stop breast-feeding?

☐ baby is/was unable to “latch onto breast”

☐ baby is/was unable to generate or maintain adequate suck

☐ mother had inverted or painful nipples

☐ embarrassment

☐ unable to tell how much feed baby is taking

☐ baby has been unwell

☐ advised by CLAPC team that breast feeding is generally not successful with CLAP babies

☐ decided after speaking to other mothers of CLAP babies

☐ other____________________________

15. If you are breast feeding your baby are you planning to continue with it? Yes (1), No (0)

Tube feeding

16. How long has or did your baby have a feeding tube for? _________________________________ days

17. Is tube feeding ongoing? Yes (1), No (0)

18. How much feed does your baby have through the tube at each feed? _____________________________ mls

Feeding observations (milk feeds)

19. Have you noticed any feed in or coming from your baby’s nose during milk feeds? Yes (1), No (0)

20. How often do you notice milk in the nose or coming from your baby’s nose?

☐ occasionally

☐ once or twice a day

☐ at every feed

21. How much milk feed returns through the nose?

- 411 -
square □ trace amounts only
square □ about a thimble full
square □ about a dessertspoon full

22. Have you noticed your baby coughing, choking or gagging
during milk feeds? Yes (1), No (0)

23. Have you noticed your baby's breathing becoming
"gurgley" during milk feeds? Yes (1), No (0)

Solid feeds

24. What solids is your baby taking now?

square □ runny smooth puree (like runny yoghurt)

square □ thick smooth puree (like set yoghurt)

square □ smooth puree with occasional "lumps" (like 7 month
cereals)

square □ lumpy puree (mashed banana consistency)

square □ bite and dissolve foods (e.g. baby rusks or Skips)

square □ other _______________________________________

25. How many solid food snacks / meals does your baby have
each day? ________________________________________ snacks / meals

26. How many mls approximately and on average does your
baby take at each snack or meal?

_______________________________________________ mls
(further details to be recorded in 3 day diary)

27. How long does it take your baby to eat this?

_______________________________________________ mins

28. Do you think your baby enjoys these solid feeds?

Yes (1), No (0)

Feeding observations (solid feeds)

29. Have you noticed any solid feed in or coming from your
baby's nose during feeds? Yes (1), No (0)

30. How often do you notice solids in the nose or coming

- 412 -
from your baby’s nose?

- Occasionally
- Once or twice a day
- At every feed

31. How much solid feed returns through the nose?
- Trace amounts only
- About a thimble full
- About a dessertspoon full

32. Have you noticed your baby coughing, choking or gagging during solid feeds?

33. Have you noticed your baby’s breathing becoming “gurgley” during solid feeds?

Feeding observations (fluid and solids)

34. Does your baby ever show signs of discomfort during feeds? e.g. crying, arching, drawing legs to chest

35. Does your baby vomit after feeds?

36. How often does this occur?
- Rarely
- Once or twice a day
- During or after every feed

37. Does your baby have any medicine for this?

38. What medicine is your baby having for this?
- Thickener added to feeds
- Gaviscon
- Ranitidine
- Domperidone
- Cisapride
- Other

39. Does your baby have a history of chest infections requiring antibiotic treatment?

40. Does anyone other than yourself feed your baby?

41. Who else feeds your baby?
- Husband / partner
☐ other adult relative     Yes (1), No (0)
☐ sibling                  Yes (1), No (0)
☐ carer                    Yes (1), No (0)
☐ friend                   Yes (1), No (0)
☐ nursing staff            Yes (1), No (0)
☐ other                    Yes (1), No (0)
"Use of PSO in feeding management of children with Cleft Lip and Palate" project

Data collection forms

Feeding Questionnaire (Data collection point 4)
(this information is collected by interview, either face to face or telephone dependent on clinic appointment schedule)

Baby's hospital number: ___________________________

Date of data collection (ddmmyyyy)  ____________________

Milk feeds

1. What milk feed is your baby taking now?
   - □ breast milk
   - □ expressed breast milk
   - □ standard formula
   - □ "special" formula
   - □ other
   
   Yes (1), No (0)

2. Are you adding anything to your baby's milk feeds?
   Yes (1), No (0)

3. What are you adding?
   - □ rice cereal
   - □ other solids
   - □ supplements
   - □ medication
   - □ flavours e.g. sugar
   
   Yes (1), No (0)

4. How many milk feeds does your baby have each day?

   _____________________________ feeds
Bottle / cup feeding

5. How is your baby taking milk / fluids now?
   □ Mead Johnson Yes (1), No (0)
   □ Soft Plas Yes (1), No (0)
   □ Chicco Yes (1), No (0)
   □ Standard Yes (1), No (0)
   □ Rosti Yes (1), No (0)
   □ Haberman feeder Yes (1), No (0)
   □ training beaker Yes (1), No (0)
   □ beaker or glass Yes (1), No (0)
   □ other e.g. cup and spoon ________ Yes (1), No (0)

6. What teat are you currently using on your baby’s bottle?
   □ NUK orthodontic, vented (size 2) Yes (1), No (0)
   □ NUK orthodontic, vented (size 1) Yes (1), No (0)
   □ Standard term / newborn latex teat e.g. Cow and Gate Yes (1), No (0)
   □ Standard term / newborn silicone teat Yes (1), No (0)
   □ Premature teats Yes (1), No (0)
   □ Extended length teat e.g. Lambs teat Yes (1), No (0)
   □ NUK Cleft Palate teat Yes (1), No (0)
   □ Cross cut teat Yes (1), No (0)
   □ Other ____________________________ Yes (1), No (0)

7. Have you made any adaptations to your baby’s teat?
   □ enlarged hole Yes (1), No (0)
   □ cross cut teat Yes (1), No (0)
   □ created a “new” hole on the side Yes (1), No (0)
   □ use the teat upside down Yes (1), No (0)
   □ other ____________________________ Yes (1), No (0)

8. How many mls of milk does your baby have at each milk feed? ____________________________ mls

9. How long does this take?
   ____________________________ mins

Breast feeding

10. Are you still breast feeding your baby? Yes (1), No (0)

11. How long do you breast-feed your baby for at each feed? ____________________________ mins
12. If you are not breast feeding now, did you try breast
your baby at any time?
Yes (1), No (0) □

13. If you tried breast-feeding, how long did you try it for?
____________________________________________ days

14. Why did you stop breast-feeding?

□ baby is/was unable to “latch onto breast” Yes (1), No (0) □
□ baby is/was unable to generate or maintain adequate
suck Yes (1), No (0) □
□ mother had inverted or painful nipples Yes (1), No (0) □
□ embarrassment Yes (1), No (0) □
□ unable to tell how much feed baby is taking Yes (1), No (0) □
□ baby has been unwell Yes (1), No (0) □
□ advised by CLAPC team that breast feeding is
generally not successful with CLAP babies Yes (1), No (0) □
□ decided after speaking to other mothers of CLAP
babies Yes (1), No (0) □
□ other_______________________________________ Yes (1), No (0)

15. If you are breast feeding your baby are you planning to
continue with it? Yes (1), No (0) □

Tube feeding

16. What type of feeding tube does your baby have?
□ nasogastric tube Yes (1), No (0) □
□ gastrostomy Yes (1), No (0) □

17. How long has or did your baby have a feeding tube for?
__________________________________________ days

18. Is tube feeding ongoing? Yes (1), No (0) □

19. How much feed does your baby have through the tube at
each feed? _________________________________ mls

20. Does your baby have overnight feeds?
Yes (1), No (0) □

21 How long do these overnight feed run for?
__________________________________________ hours

22. How much feed does your baby have over this time?
__________________________________________ mls
Feeding observations (milk feeds)

23. Have you noticed any feed in or coming from your baby's nose during milk feeds? Yes (1), No (0) [ ]

24. How often do you notice milk in the nose or coming from your baby's nose?

☐ occasionally Yes (1), No (0) [ ]
☐ once or twice a day Yes (1), No (0) [ ]
☐ at every feed Yes (1), No (0) [ ]

25. How much milk feed returns through the nose?

☐ trace amounts only Yes (1), No (0) [ ]
☐ about a thimble full Yes (1), No (0) [ ]
☐ about a dessertspoon full Yes (1), No (0) [ ]

26. Have you noticed your baby coughing, choking or gagging during milk feeds? Yes (1), No (0) [ ]

27. Have you noticed your baby's breathing becoming "gurgley" during milk feeds? Yes (1), No (0) [ ]

Solid feeds

28. What solids is your baby taking now?

☐ runny smooth puree (like runny yoghurt) Yes (1), No (0) [ ]
☐ thick smooth puree (like set yoghurt) Yes (1), No (0) [ ]
☐ smooth puree with occasional "lumps" (like 7 month cereals) Yes (1), No (0) [ ]
☐ lumpy puree (mashed banana consistency) Yes (1), No (0) [ ]
☐ bite and dissolve foods (e.g. baby rusks or Skips) Yes (1), No (0) [ ]
☐ mashed table foods Yes (1), No (0) [ ]
☐ "soft" / crumbly solids e.g. biscuits / bread Yes (1), No (0) [ ]
☐ other ____________________________________________ Yes (1), No (0) [ ]
29. How many solid food snacks / meals does your baby have each day?  ________________________________________snacks / meals

30. How many mls approximately and on average does your baby take at each snack or meal?  ________________________________________mls
   (further details to be recorded in 3 day diary)

31. How long does it take your baby to eat this?  ________________________________________mins

32. Do you think your baby enjoys these solid feeds?  Yes (1), No (0)

Feeding observations (solid feeds)

33. Have you noticed any solid feed in or coming from your baby's nose during feeds?  Yes (1), No (0)

34. How often do you notice solids in the nose or coming from your baby's nose?
   □ occasionally  Yes (1), No (0)
   □ once or twice a day  Yes (1), No (0)
   □ at every feed  Yes (1), No (0)

35. How much solid feed returns through the nose?
   □ trace amounts only  Yes (1), No (0)
   □ about a thimble full  Yes (1), No (0)
   □ about a dessertspoon full  Yes (1), No (0)

36. Have you notice your baby coughing, choking or gagging during solid feeds?  Yes (1), No (0)

37. Have you noticed your baby's breathing becoming "gurgley" during solid feeds?  Yes (1), No (0)

Feeding observations (fluid and solids)

38. Does your baby ever show signs of discomfort during feeds? e.g. crying, arching, drawing legs to chest  Yes (1), No (0)

39. Does your baby vomit after feeds?  Yes (1), No (0)

40. How often does this occur?
   □ rarely  Yes (1), No (0)
   □ once or twice a day  Yes (1), No (0)
   □ during or after every feed  Yes (1), No (0)
41. Does your baby have any medicine for this? Yes (1), No (0) □

42. What medicine is your baby having for this?
□ thickener added to feeds Yes (1), No (0) □
□ Gaviscon Yes (1), No (0) □
□ Ranitidine Yes (1), No (0) □
□ Domperidone Yes (1), No (0) □
□ Cisapride Yes (1), No (0) □
□ other ____________________________ Yes (1), No (0) □

43. Does your baby have a history of chest infections requiring antibiotic treatment? Yes (1), No (0) □

44. Does anyone other than yourself feed your baby? Yes (1), No (0) □

45. Who else feeds your baby?
□ husband / partner Yes (1), No (0) □
□ other adult relative Yes (1), No (0) □
□ sibling Yes (1), No (0) □
□ carer Yes (1), No (0) □
□ friend Yes (1), No (0) □
□ nursing staff Yes (1), No (0) □
□ other ____________________________ Yes (1), No (0) □
Great Ormond Street Hospital for Children NHS Trust
and the Institute of Child Health
Speech and Language Therapy Department

"Use of PSO in feeding management of children with Cleft Lip and Palate" project

Data collection forms

Feeding Questionnaire (Data collection point 5)
(this information is collected by interview)

Baby's hospital number: ____________________________
Date of data collection (dd/mm/yyyy)

Milk feeds

1. What milk feed is your baby taking now?
- breast milk
- expressed breast milk
- standard formula
- "special" formula
- cows / soya milk
- other

2. Are you adding anything to your baby's milk feeds?

3. What are you adding?
- rice cereal
- other solids
- supplements
- medication
- flavours e.g. sugar

4. How many milk feeds does your baby have each day?

Appendix 25
Bottle / cup feeding

5. How is your baby taking milk / fluids now?
   □ Mead Johnson Yes (1), No (0)
   □ Soft Plas Yes (1), No (0)
   □ Chicco Yes (1), No (0)
   □ Standard Yes (1), No (0)
   □ Rosti Yes (1), No (0)
   □ Haberman feeder Yes (1), No (0)
   □ training beaker Yes (1), No (0)
   □ beaker or glass Yes (1), No (0)
   □ other e.g. cup and spoon Yes (1), No (0)

6. What teat are you currently using on your baby’s bottle?
   □ NUK orthodontic, vented (size 2) Yes (1), No (0)
   □ NUK orthodontic, vented (size 1) Yes (1), No (0)
   □ Standard term / newborn latex teat e.g. Cow and Gate Yes (1), No (0)
   □ Standard term / newborn silicone teat Yes (1), No (0)
   □ Premature teats Yes (1), No (0)
   □ Extended length teat e.g. Lambs teat Yes (1), No (0)
   □ NUK Cleft Palate teat Yes (1), No (0)
   □ Cross cut teat Yes (1), No (0)
   □ Other Yes (1), No (0)

7. Have you made any adaptations to your baby’s teat?
   □ enlarged hole Yes (1), No (0)
   □ cross cut teat Yes (1), No (0)
   □ created a “new” hole on the side Yes (1), No (0)
   □ use the teat upside down Yes (1), No (0)
   □ other Yes (1), No (0)

8. How many mls of fluid does your baby have at any one time? ____________________________ mls

9. How long does he / she take to drink this?
   ____________________________ mins

Breast feeding

10. Are you still breast feeding your baby?
    Yes (1), No (0)
11. How long do you breast-feed your baby for at each feed? ____________________________ mins

12. If you are not breast feeding now, did you try breast feeding your baby at any time? Yes (1), No (0)

13. If you tried breast-feeding, how long did you try it for? ____________________________ days

14. Why did you stop breast-feeding?

- baby is/was unable to "latch onto breast" Yes (1), No (0)
- baby is/was unable to generate or maintain adequate suck Yes (1), No (0)
- mother had inverted or painful nipples Yes (1), No (0)
- embarrassment Yes (1), No (0)
- unable to tell how much feed baby is taking Yes (1), No (0)
- baby has been unwell Yes (1), No (0)
- advised by CLAPC team that breast feeding is generally not successful with CLAP babies Yes (1), No (0)
- decided after speaking to other mothers of CLAP babies Yes (1), No (0)
- developmentally appropriate Yes (1), No (0)
- other_______________________________________ Yes (1), No (0)

15. If you are breast feeding your baby are you planning to continue with it? Yes (1), No (0)

Tube feeding

16. What type of feeding tube does your baby have?
- nasogastric tube Yes (1), No (0)
- gastrostomy Yes (1), No (0)

17. How long has or did your baby have a feeding tube for? ____________________________ days

18. Is tube feeding ongoing? Yes (1), No (0)

19. How much feed does your baby have through the tube at each feed? ____________________________ mls

20. Does your baby have overnight feeds? Yes (1), No (0)
21. How long do these overnight feeds run for? ___________________________ hours

22. How much feed does your baby have over this time? ___________________________ mls

Feeding observations (fluids)

23. Have you noticed any fluid in or coming from your baby's nose during milk feeds? Yes (1), No (0)

24. How often do you notice fluids in the nose or coming from your baby's nose?

☐ occasionally
☐ once or twice a day
☐ at every feed

25. How much fluid returns through the nose?

☐ trace amounts only
☐ about a thimble full
☐ about a dessertspoon full

26. Have you noticed your baby coughing, choking or gagging while drinking? Yes (1), No (0)

27. Have you noticed your baby's breathing becoming "gurgley" while drinking? Yes (1), No (0)

Solid feeds

28. What solids is your baby taking now?

☐ runny smooth puree (like runny yoghurt)
☐ thick smooth puree (like set yoghurt)
☐ smooth puree with occasional "lumps" (like 7 month cereals)
☐ lumpy puree (mashed banana consistency)
☐ bite and dissolve foods (e.g. baby rusks or Skips)
□ mashed table foods Yes (1), No (0)
□ "soft" solids e.g. biscuits / bread Yes (1), No (0)
□ chopped table foods Yes (1), No (0)
□ other ________________________________ Yes (1), No (0)

29. How many solid food snacks / meals does your baby have each day? ________________________________ snacks / meals

30. How many mls approximately and on average does your baby take at each snack or meal? (how many dessertspoons where each dessertspoon = approx 10 mls) ________________________________ mls
(further details to be recorded in 3 day diary)

31. How long does it take your baby to eat this? ________________________________ mins

32. Do you think your baby enjoys these solid feeds? Yes (1), No (0)

Feeding observations (solid feeds)

33. Have you noticed any solids in or coming from your baby's nose during feeds? Yes (1), No (0)

34. How often do you notice solids in the nose or coming from your baby's nose?
□ occasionally Yes (1), No (0)
□ once or twice a day Yes (1), No (0)
□ at every feed Yes (1), No (0)

35. How much solid feed returns through the nose?
□ trace amounts only Yes (1), No (0)
□ about a thimble full Yes (1), No (0)
□ about a dessertspoon full Yes (1), No (0)

36. Have you notice your baby coughing, choking or gagging with solids? Yes (1), No (0)

37. Have you noticed your baby's breathing becoming "gurgley" during solid feeds? Yes (1), No (0)

Feeding observations (fluid and solids)

38. Does your baby ever show signs of discomfort during feeds? e.g. crying, arching, drawing legs to chest Yes (1), No (0)
39. Does your baby vomit after feeds?  
- Yes (1), No (0)  

40. How often does this occur?  
- rarely  
- once or twice a day  
- during or after every feed  
- Yes (1), No (0)  

41. Does your baby have any medicine for this?  
- Yes (1), No (0)  

42. What medicine is your baby having for this?  
- thickener added to feeds  
- Gaviscon  
- Ranitidine  
- Domperidone  
- Cisapride  
- Other  
- Yes (1), No (0)  

43. Does your baby have a history of chest infections requiring antibiotic treatment?  
- Yes (1), No (0)  

44. Does anyone other than yourself feed your baby?  
- Yes (1), No (0)  

45. Who else feeds your baby?  
- husband / partner  
- other adult relative  
- sibling  
- carer  
- friend  
- nursing staff  
- Other  
- Yes (1), No (0)


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