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- 1 Title page
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# Cost-effectiveness of the Family Nurse Partnership (FNP) programme in England: evidence from the Building Blocks trial

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25 BC (Research Fellow, Health Economics) was responsible for conducting the economic evaluation and 26 produced first draft of the paper. Dr KB (Research Fellow, Health Economics) was the second Health 27 Economist and also conducted the economic evaluation. ES (Research Fellow, Health Economics), Dr GR 28 (Senior Research Fellow, Health Economics) and SR (Research Fellow, Health Economics) were also Health 29 Economists and contributed to the design and methodological approach for the health economics. JS (Senior 30 Clinical Researcher) was responsible for the delivery of the trial. Professor KH (Professor of Medical Statistics) 31 provided senior study design and statistical support to the trial. Professor MR (Director of Population Health 32 Trials) was the Chief Investigator of the trial and acts as overall guarantor for the study. Professor DT (Director 33 York Trials Unit) provided advice on the design and methodological approach for the trial as a whole and 34 oversaw the conduct of the economic analysis.

35

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- 48

- Cost-effectiveness of the Family Nurse Partnership (FNP) programme in England: evidence from the
  Building Blocks trial

52 Abstract

Rational, aims and objectives: The Family Nurse Partnership (FNP) is a licensed intensive home visiting
intervention developed in the USA. It has been provided in England by the Department of Health (DH) since
2006. The Building Blocks (BB) trial assessed the effectiveness and cost-effectiveness of FNP in England.

Methods: We performed a cost-utility analysis (NHS perspective) alongside the BB trial (over 2.5 years). The analysis was conducted in accordance with NICE (National Institute for Health and Clinical Excellence) reference case standards. Health-related quality of life was elicited from mothers using the EQ-5D-3L. Resource use data were collected from self-reported questionnaires, Hospital Episode Statistics (HES), general practitioner records and the central DH FNP database. Costs and quality-adjusted life years (QALYs) were discounted at 3.5%. The base case analysis used an intention to treat approach on the imputed dataset using multiple imputation (MI).

Results: The FNP intervention costs on average £1812 more per participant compared to usual care (95% CI £2700; £5744). Incremental adjusted mean QALYs are marginally higher for FNP (mean difference 0.0036,
95% CI -0.017; 0.025). The probability of FNP being cost-effective is less than 20% given the current NICE
willingness to pay threshold of £20 000 per additional QALY. The results were robust to sensitivity analyses.

Conclusion: Given the absence of significant benefits of FNP in terms of the primary outcomes of the trial and
 only marginal maternal QALY gains, FNP does not represent a cost-effective intervention when compared with
 existing services already offered to young pregnant women.

- 80 INTRODUCTION
- 81

The Family Nurse Partnership (FNP) programme is an intensive preventive home visiting service with positive results compared to usual services for mothers and babies both in the USA and the Netherlands [1-4]. It was introduced for first time young mothers into NHS England by the Department of Health in 2006 [5-7]. In October 2015 the FNP was transferred from NHS England to Local Authorities (LAs) and it is provided in approximately 125 different LAs in England. The FNP programme was introduced to be an integral part of the progressive universalism approach recommended in The Healthy Child Programme (HCP). The HCP is delivered by the Family Nurse rather than by health visitors for women who enrol onto the programme.

89 Given the lack of evidence on the benefits of the FNP programme in England, the Building Blocks (BB) trial 90 was commissioned to evaluate the effectiveness and cost-effectiveness of the FNP intervention when delivered 91 in a comprehensive publicly funded health care setting. The BB protocol has been published 11 and all 92 amendments were reviewed and approved by the Wales NHS Research Ethics Committee (09/MRE09/08). The 93 details of the trial design, outcomes and clinical effectiveness are reported elsewhere 12, 13. The results from 94 the effectiveness analysis showed no statistically or clinically significant difference associated with FNP for any 95 of the four primary outcomes: smoking cessation (adjusted OR 0.90, 97.5% CI 0.64-1.2), birth weight (adjusted mean difference 20.75 g, 97.5% CI -47.73 to 89.23), second pregnancies within two years (AOR 96 1.01, 0.77-1.33), or child A&E attendances and admissions to hospital (AOR 1.32, 97.5% CI 0.99-1.76, 97 98 p=0.03).

99 This paper reports on the cost-effectiveness analysis conducted alongside the Building Blocks (BB) trial. This 100 economic analysis also seeks to better understand a key example of an intervention that is routinely provided 101 without clear evidence on whether it represents good value for money for the health system compared to 102 comprehensive services, which in turn may help to guide disinvestment decisions that are unavoidable given the 103 financial constraints within a publicly funded health care system.

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105 METHODS
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107 Overview
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Individual patient data from the BB trial were used to perform a cost-utility analysis measuring health-relatedquality of life (HRQoL) in terms of quality-adjusted life years (QALYs). The analysis was from the NHS and

personal social services (PSS) perspective and expressed in UK pounds sterling (2013 GBP). Costs and QALYs
were discounted at a rate of 3.5% [8]. We used a regression approach on an intention-to-treat (ITT) basis. The
base case analysis was conducted on the dataset generated by multiple imputation by chained equations [9].
Sensitivity analysis included complete case (CC) analysis to test the impact of excluding participants with
missing data on the final results. All analyses and modelling were conducted in Stata 13.1 (StataCorp 2011, TX,
USA).

116 The BB trial was a pragmatic, non-blinded, parallel-group, randomised controlled trial (RCT) which recruited 117 within a community midwifery setting at 18 partnerships between LAs and primary and secondary care organisations in England. The trial compared two arms; usual care (through primary-care public health and 118 119 social care services) plus FNP (FNP group) to usually provided health and social care alone (usual care group). 120 These groups were followed from early pregnancy (as soon as possible from the end of the first trimester) until 121 two years following childbirth. The trial recruited 1645 teenagers expecting their first baby, at less than 25 122 weeks gestation, between 16th June 2009 and 28th July 2010. The economic analysis is based on the 1618 123 participants that were assessed as eligible and did not withdraw consent for their data to be used. Mandatory 124 withdrawals (e.g. miscarriage, termination for fetal anomaly) were not included in the primary analysis 125 (FNP=26; usual care=24). Hence a total of 1568 (FNP=782; usual care=786) women were included in the base 126 case analysis.

127

#### 128 Health outcomes and quality-adjusted life-years

The primary outcome measure was QALYs, based on the EQ-5D-3LTM (EuroQoL Group Rotterdam, The Netherlands) reported by the women. The EQ-5D-3L has been used before in the UK setting within the context of a pregnant population, for example in the economic evaluation conducted alongside the Early Labour Support and Assessment (ELSA) trial [10]. The EQ-5D-3L health states were valued using a UK-based social tariff [11]. QALYs were calculated using the area under the curve method (AUC) [12] and were adjusted for baseline utility[13].

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#### 138 Resource use and costs

139 Costs and health outcome data were collected via self-reported questionnaires at various time-points throughout 140 the trial: baseline, late pregnancy (34-36 weeks gestation), and 6, 12, 18 and 24 months postpartum. Baseline 141 and 24 month data were collected by face-to-face interview by a locally based researcher. Follow-up self-142 reported data were collected via telephone by qualified telephone interviewers for the remainder of the time-143 points. Data related to the use of hospital services were obtained from maternity notes, the Health and Social 144 Care Information Centre (HSCIC, Hospital Episode Statistics (HES)); and general practitioner (GP) visits were 145 obtained from GP records. The cost of the FNP intervention was based on the centralised FNP Information 146 System (IS) database run by Connecting for Health in Exeter (e.g. number of nurse visits, duration of visit, 147 number of telephone encounters). The unit costs used to estimate the total cost per participant in the trial are 148 presented in Table 1, sourced from the Personal Social Services Research Unit [14] and NHS reference costs 149 [15].

150 Table 1

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152

### 153 Handling missing data

154 Complete case assessment excludes all participants with any missing or incomplete data. Excluding patients 155 with missing data leads to loss of power and biasing of the results due to a reduced sample size [16]. The 156 method we used to handle missing data was informed by the BB data. Incomplete data on costs and QALYs 157 were imputed using multiple imputation (MI) with chained equations and predictive mean matching; which 158 assumes that data are missing at random (MAR) [17-19]. The same set of covariates as in the clinical 159 effectiveness analysis was selected with stepwise regressions (e.g. site, smoking status, language and gestation). 160 Rubin's rules were used to combine point and variance estimates across imputed datasets, allowing the 161 estimation of difference in costs and QALYs between both groups [20].

162

#### 163 Base-case analysis

164 The base-case analysis was conducted on the multiple imputed dataset, and followed an intention-to-treat (ITT) 165 approach. For the base-case analysis total costs constituted the cost of the FNP programme (nurse time used for home based visits from the FNP nurse); GP and nurse visits (recorded in GP records), post-natal midwife and
health visitor visits (self-reported by mothers); and hospital activity (HES records for inpatient admission,
outpatient visits and A&E services).

169 The cost-effectiveness of the FNP programme was evaluated by comparing the mean differences in costs and 170 effects in the two groups, using conventional rules and estimating the incremental cost-effectiveness ratio 171 (ICER) as appropriate [21]. The mean differences in costs and QALYs were estimated using seemingly 172 unrelated regression (SUR) [22], and their 95% confidence intervals (CI) estimated using bias corrected and accelerated (BCA) bootstrap methods. Non parametric bootstrapping [23] was used to transform the uncertainty 173 around the trial estimates into the probability that the FNP intervention is cost-effective for thresholds used by 174 175 the National Institute for Health and Care Excellence (NICE) of £20 000 and £30 000 per QALY gained [8], 176 with cost-effectiveness acceptability curves (CEACs) used.

177

#### 178 Sensitivity analyses

Sensitivity analyses in the base case were conducted to test the robustness of the results using five scenarios: complete case (CC) analysis according to ITT; MI removing midwife visits reported by mothers allocated to FNP (i.e. to avoid double counting in case those visits were already included in the FNP IS dataset); MI including the limited data available for mothers that withdrew due to mandatory withdrawals (i.e if FNP were to be implemented, costs related to mandatory withdrawals would be covered by the NHS); MI using self-report data (i.e including resource use exclusively related to mothers, hence excluding resource use related to babies); and MI halving the cost for the FNP intervention.

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#### 187 RESULTS

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**189** The results from the effectiveness analysis showed no statistically or clinically significant difference associated

with FNP for any of the four primary outcomes: smoking cessation (adjusted OR 0.90, 97.5% CI 0.64-1.2),

birth weight (adjusted mean difference 20.75 g, 97.5% CI –47.73 to 89.23), second pregnancies within two

192 years (AOR 1.01, 0.77–1.33), or child A&E attendances and admissions to hospital (AOR 1.32, 97.5% CI

193 0.99-1.76, p=0.03).

#### 194 Health outcomes and quality-adjusted life-years

Table 2 presents the proportion of participants with complete EQ-5D data. A small number of trial participants 195 196 (n=10) allocated to the control arm were erroneously enrolled into FNP. Following the ITT principle they were 197 analysed in their allocated arm regardless. Data were considered missing or incomplete when women did not 198 complete the EQ-5D or provided a partially completed questionnaire. Two points would support the MAR 199 assumption used as the basis for the base-case analysis: (i) the missing data followed an intermittent pattern (e.g. 200 in both groups, more women were observed at 12 months than at six months, and the same pattern is observed at 201 18 and 24 months) hence complete case assessment would be, as a minimum, inefficient because it would 202 discard observed data from individuals with some missing outcomes; and (ii) the BB data showed that 203 participants with lower EQ-5D at baseline were more likely to have missing QALY data, which in turn suggests 204 that the data are unlikely to be MCAR.

Participants in the FNP group started from a lower baseline HRQoL was a mean (SD) of 0.90 (0.005) (FNP)
versus 0.91 (0.005) (usual care). The EQ-5D-3L scores did not differ significantly between groups at each
follow-up (Table 3). The difference in mean EQ-5D-3L scores at 2 years (FNP – usual care) when controlling
for baseline utility (for available cases: 320 FNP versus 265 usual care) was -0.008 (95% CI -0.023 to 0.008).

209 Table 2

210

211 Despite any difference seen in the EQ-5D-3L across the groups, this translates to very little difference in utilities
212 (Figure I) and QALYs between the FNP and usual care groups.

213 Figure 1

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### 215 Resource use and costs

There were no clear differences in resource use across the two groups (Table 3), though A&E attendances for mothers and babies were somewhat higher in the FNP group. Babies in the usual care group had on average longer inpatient length of stay in hospital than those babies whose mothers were randomised to the FNP intervention.

220 *Table 3* 

Cost differences were very small across groups (Table 4). Costs associated with the delivery of the FNP intervention and the inpatient stays in hospital for babies were the major cost drivers for the cost-effectiveness. Using the principle of ITT and assigning a cost of £0.00 to women who did not receive the intervention, the average cost of nurse time for FNP visits was £3845 (SD £77), and £33 (SD £2) for FNP phone calls; however, considering only the 719 women who received the intervention, the average cost of nurse time was £4,270 (SD £1,855) per woman. If we assume that on average women were recruited at 18 weeks gestation then, the annual cost of nurse time for the FNP home visits is £1,762 per women.

229 Table 4

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#### 231 Cost-effectiveness analysis

232 The incremental analysis (Table 5), when adjusting for all covariates (baseline utility, site, smoking status, 233 language and gestation), showed the FNP intervention costs on average £1811 more per participant when 234 compared with usual care (95% CI -£2814; £5547). Incremental mean QALYs when adjusted for baseline utility 235 are marginally higher for FNP (mean difference 0.0036, 95% CI -0.017; 0.025). This difference is even lower when adjusted for the remaining covariates (mean difference 0.0030, 95% CI -0.017; 0.027). The Net Monetary 236 237 Benefit associated with the FNP intervention is negative (-£1750.57), indicating that the resources displaced would be greater than the benefit gained with the delivery of the FNP intervention. The probability of the FNP 238 239 intervention being cost-effective was less than 20% given the NICE currently accepted threshold of £20 000 to 240 £30 000 per additional QALY (Figure 2).

241 Table 5

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243 Figure 2

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The sensitivity analyses (Table 5) showed that the conclusions from the base case analysis were robust to all scenarios, FNP remained a non-cost-effective intervention, with the ICERs much higher than the thresholds that NICE normally consider for reimbursement decisions. Because it was felt that the cost of the intervention is the main cost driver for the analysis, the cost of the FNP intervention was halved to assess the effect on the costeffectiveness conclusions. The mean difference in costs per participant is reduced to £360 (95% CI £-3680 to
£4352); the gain in QALYs is still very marginal at 0.0047 (95% CI -0.013; 0.022). The results continue to be
uncertain and the probability of FNP being cost-effective less than 50%. The complete case analysis shows that
the FNP intervention cost, on average, £4549 more per woman when compared with usual care (95% CI £3175
to £5922). Participants allocated to the FNP intervention accrued fewer QALYs than those for usual Care (0.007, 95% CI -0.042 to 0.027). Therefore the results of the complete case analysis indicate FNP was dominated
by usual care.

256

#### 257 DISCUSSION

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This economic evaluation provides evidence that FNP is more costly than usual care and provides only a very small QALY gain. Similarly the analysis of uncertainty confirmed that it is unlikely that FNP represents an efficient intervention even if the cost was substantially reduced. The base case results indicate that the probability of FNP being cost-effective is 17%, with the results being robust to sensitivity analyses.

263 There is evidence of positive results of the FNP intervention in the US, a context where mothers are not able to 264 access many statutory supportive health and social services, and maternity community based services. In 265 contrast, pregnant women in the UK can access a wide provision of maternal care including community care family doctors, midwives and public health nurses, and as we observed in our trial sites, specialist teenage 266 267 pregnancy midwives as well. It is worth it to note that the differences between the US and UK health systems 268 can explain the lack of clinical or quality-of-life benefits for those women who receive the FNP programme as a public service offered in the UK. The Building Blocks trial is the first UK-based trial of FNP, therefore this 269 270 analysis represents the most up-to-date estimate of the cost-effectiveness of FNP when delivered in a universal, 271 publicly funded, health care setting.

There are two noteworthy limitations of this study. The first limitation relates to the level of missing data. Despite steps to minimise missing data (e.g. computer assisted telephone interviews), the number of EQ-5D questionnaires completed for each data collection wave decreased over time. This is a common problem in trialbased economic evaluations that is amplified where there are frequent assessments, as in here. It is worth noting that the use of HES data helped to minimise considerably the problem of incomplete data thus enabling more accurate estimates of hospital costs. The second limitation relates to the duration of the BB trial. In our trial we are able to assess programme cost-effectiveness in the short-term only and we recognise that for preventative

279 programmes benefits may be expected to accrue over a longer time period and in domains of child development. 280 Reported analyses of programme cost-effectiveness in the US have highlighted the advantage for high-risk 281 families in particular over longer but variable periods of time. The Social Research Unit at Dartington, and 282 Aldaba Limited conducted a cost-benefit analysis for FNP in the UK using a modelling approach which 283 describes the longer-term savings by the FNP programme, indicating that key savings being related to higher 284 earnings and higher attainment test scores [24] There have been some shortcomings identified with the 285 economics analyses previously reported [25], for example some double counting of non-independent outcomes. 286 In one meta-analytic review the cost savings for the programme were greatest for outcomes related to the mother (e.g. reduced crime, higher earnings, reduction in welfare) rather than the child [26]. Positive programme 287 288 outcomes identified the trial include maternally reported child language development. This is of potential 289 longer-term importance but requires further evaluation over the medium term to first verify with objective 290 ratings and second to determine whether any short-term advantage is continued to improvements in later 291 outcomes such as school readiness. These objectives are currently being assessed in a linked study.

292 Besides it could be argued that we only look at QoL for the women and not the children. The EuroQol Group 293 has developed a child-friendly EQ-5D version (EQ-5D-Y)[27], however the age of 8 is the lower age limit for 294 which the instrument is valid hence the EQ-5D-Y was not applicable for the children in the BB trial. Regarding 295 mothers, the EQ-5D-3L did detect differences in scores between stages of pregnancy (e.g. women reported more problems in pain/discomfort and mobility at late pregnancy than any other follow-up point); which shows that 296 297 this instrument can capture small yet important changes which are important to reflect the impact of the 298 intervention on health. Similarly it could be debated that the QALY instrument lacks sensitivity for measuring 299 the impact of FNP in health. Nonetheless within the UK the access to a new intervention has to be justified by 300 the health gain it provides compared with usual care, where the added benefit is typically expressed in QALYs. 301 In order to overcome the limitation of the QALY approach, this study was complemented with (i) a cost-302 consequence analysis (Policy Research Programme Project April 2015); and (ii) a discrete choice experiment 303 that examined the preferences of the general population for the outcomes of the trial.

There are two main points that this study adds. From a research perspective, this study emphasises the need to conduct trials and to evaluate the effectiveness and cost-effectiveness of policy interventions before they are implemented. From a policy making perspective this study can contribute to everyday decision making regarding which services to offer to young pregnant women. Our findings provide strong evidence to suggest that the delivery of FNP is not better for young mothers or their babies than comprehensive services in the short
term, while costing more. There is currently evidence supporting other cost-effective public interventions among
pregnant women aiming to address some of the primary outcomes of the BB trial, with a positive influence at a
relatively low cost, and maybe cost saving (e.g. smoking cessation) (Ruger et al. 2008; Tappin et al. 2015).

In England and Wales, there were 23 948 live births to women aged under 20 during 2015 (Statistics 2015)). If we assume that around 50% of these women were offered an FNP place then the annual cost saving in England from removing the FNP programme would be around £21 million.

315

#### 316 CONCLUSION

317 The Building Blocks trial is the first UK-based trial of FNP. As discussed, the FNP intervention did not deliver 318 significant benefits on any of the primary outcomes and only limited benefits on a small number of secondary 319 outcomes where the risk of a chance finding is greater. Thus, taken together with the effectiveness findings, the 320 results of this economic evaluation suggest that FNP does not represent a cost-effective intervention when 321 adding FNP to existing services already offered to young pregnant women in England. However, it is important 322 to note that these results are based on the two year trial only and cannot account for any longitudinal outcomes 323 that may emerge at a later stage. Hence, at this time, we cannot recommend the continuation of FNP delivery and it may be the case that displacing the resources currently used in the FNP and investing in alternative 324 325 interventions could potentially result in greater gains in child health, development and family economic stability 326 for this population.

327

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368	
369	References
370	<ol> <li>Kitzman H, Olds DL, Henderson CR, et al. Effect of prenatal and infancy home visitation by nurses</li></ol>
371	on pregnancy outcomes, childhood injuries, and repeated childbearing: a randomized
372	controlled trial. JAMA 1997; <b>278</b> (8):644-52
373	<ol> <li>Mejdoubi J, van den Heijkant SC, van Leerdam FJ, Crone M, Crijnen A, HiraSing RA. Effects of nurse</li></ol>
374	home visitation on cigarette smoking, pregnancy outcomes and breastfeeding: A
375	randomized controlled trial. Midwifery 2014;30(6):688-95
376	<ol> <li>Olds DL, Henderson CR, Tatelbaum R, Chamberlin R. Improving the delivery of prenatal care and</li></ol>
377	outcomes of pregnancy: a randomized trial of nurse home visitation. Pediatrics
378	1986;77(1):16-28
379	<ol> <li>Olds DL, Robinson J, O'Brien R, et al. Home visiting by paraprofessionals and by nurses: a</li></ol>
380	randomized, controlled trial. Pediatrics 2002;110(3):486-96
381	5. Barnes J. Nurse-family partnership programme: first year pilot site implementation in England:
382	pregnancy and the post-partum period. 2008
383	<ol><li>Barnes J. Nurse-family partnership programme second year pilot sites implementation in England</li></ol>
384	the infancy period. 2009
385	7. Barnes J, Ball M, Meadows P, et al. The Family-Nurse Partnership programme in England: Wave 1
386	implementation in toddlerhood and a comparison between Waves 1 and 2a of
387	implementation in pregnancy and infancy. 2011
388	8. National Institute for Health and Care Excellence. Guide to the methods of technology appraisal.
389	London: NICE, 2013.
390	<ol> <li>Faria R, Gomes M, Epstein D, White IR. A guide to handling missing data in cost-effectiveness</li></ol>
391	analysis conducted within randomised controlled trials. PharmacoEconomics
392	2014;32(12):1157-70
393	10. Spiby H. Improving care at the primary/secondary interface: a trial of community-based support
394	in early labour. The ELSA trial. Report for the National Co-ordinating Centre for NHS Service
395	Delivery and Organisation R&D (NCCSDO) 2008
396 397 398 399	<ol> <li>Dolan P, Gudex C, Kind P, Williams A. A social tariff for Euroqol: Results from a UK general population survey. York: Centre for Health Economics Discussion Paper. Sep; 138, 1995.</li> <li>Billingham L, Abrams KR, Jones DR. Methods for the analysis of quality-of-life and survival data in health technology assessment. Health technology assessment (Winchester, England)</li> </ol>
400	1998; <b>3</b> (10):1-152
401	13. Manca A, Hawkins N, Sculpher MJ. Estimating mean QALYs in trial-based cost-effectiveness
402	analysis: the importance of controlling for baseline utility. Health economics 2005; <b>14</b> (5):487-
403	96
404	14. Curtis L. Unit Costs of Health and Social Care 2013. University of Kent: Personal Social Services
405	Research Unit, 2013.
406	15. Department of Health. NHS Reference Costs 2012/13
407	https:// <u>www.gov.uk/government/publications/nhs-reference-costs-2012-to-2013</u> , 2013.
408	16. Little RJ, Rubin DB. The analysis of social science data with missing values. Sociological Methods
409	& Research 1989; <b>18</b> (2-3):292-326
410	17. Manca A, Palmer S. Handling missing data in patient-level cost-effectiveness analysis alongside
411	randomised clinical trials. Applied health economics and health policy 2005; <b>4</b> (2):65-75
412	18. White IR, Royston P, Wood AM. Multiple imputation using chained equations: issues and
413	guidance for practice. Stat. Med. 2011; <b>30</b> (4):377-99
414 415	19. Carlin JB, Galati JC, Royston P. A new framework for managing and analyzing multiply imputed data in Stata. Stata Journal 2008;8(1):49-67
415 416 417 418	<ul> <li>20. Rubin DB. <i>Multiple imputation for nonresponse in surveys</i>: John Wiley &amp; Sons, 2004.</li> <li>21. Johannesson M, Weinstein MC. On the decision rules of cost-effectiveness analysis. J. Health Econ. 1993;<b>12</b>(4):459-67</li> </ul>

419 420 421	22. Manca A, Hawkins N, Sculpher MJ. Estimating mean QALYs in trial-based cost-effectiveness analysis: the importance of controlling for baseline utility. Health Econ. 2005; <b>14</b> (5):487-96 doi: 10.1002/hec.944[published Online First: Epub Date]].
422	23. Efron B, Tibshirani RJ. <i>An introduction to the bootstrap</i> : CRC press, 1994.
423	24. Social Research Unit D. <u>http://investinginchildren.eu/interventions/family-nurse-partnership</u> .
424	25. Stamuli E, Richardson G, Duffy S, Robling M, Hood K. Systematic review of the economic
425	evidence on home visitation programmes for vulnerable pregnant women. Br. Med. Bull.
426	2015; <b>115</b> (1):19-44
427	26. Aos S, Lieb R, Mayfield J, Miller M, Pennucci A. Benefits and costs of prevention and early
428	intervention programs for youth: Olympia: Washington State Institute for Public Policy,
429	2004.
430	27. Wille N, Badia X, Bonsel G, et al. Development of the EQ-5D-Y: a child-friendly version of the EQ-
	5D. Qual. Life Res. 2010; <b>19</b> (6):875-86
431	5D. Qual. Life Res. 2010, <b>19</b> (0).875-80
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Item	Unit	Cost	Reference	Notes	
GP	Per Surgery consultation lasting 11.7 min	£45	Unit Costs of Health and Social Care 2013	Including direct care staff costs & qualifications	
	Per out of surgery (home visiting) lasting 23.4 min	£114			
GP Nurse	Per Surgery consultation lasting 15.5 minutes	£13.4	Unit Costs of Health and Social Care 2013	Assume same duration than GP home visit	
	Per home visiting lasting 23.4 min	£27.3			
Midwife	Antenatal visit (Community)	£51	NHS reference costs 2012/2013 (NHS trusts and NHS	Community Health Services – Health Visiting and Midwifery	
	Postnatal visit (Community)	£68	foundation trusts)		
	Home visit	£70	Unit Costs of Health and Social Care 2013		
	Midwife episode	£65		Total Outpatient attendances data	
Health visitor	Per hour	£49	Unit Costs of Health and Social Care 2010	Assume same duration than GP home visits	
Counsellor	Per hour of home visiting Surgery consultation	£71 £58	Unit Costs of Health and Social Care 2013		
Mental health	Per hour per team member	£36	Unit Costs of Health and Social Care 2013	Community mental health team for adults with mental health problems.	
Crisis Resolution team	Per hour per team member	£37	Unit Costs of Health and Social Care 2013		
Support worker	Per hour	£22	Unit Costs of Health and Social Care 2013		
Social worker	Per hour	£79	Unit Costs of Health and Social Care 2013		
Physiotherapist	Surgery session per hour	£34	Unit Costs of Health and Social Care 2013		
	Hospital session per hour	£36			
FNP Supervisor Nurse	Clinic or phone visit per minute	£1.34	Unit Costs of Health and Social Care 2013	Qualified nursing, midwifery & health visiting staff by Agenda for change band 8a, NHS England.	
	Home visit per minute	£1.62		Ratio of direct time on: Home visits (1:0.45) Patient work (1:0.20)	
FNP Nurse	Clinic or phone visit per minute	£1.17	Unit Costs of Health and Social Care 2013	Qualified nursing, midwifery & health visiting staff by Agenda for change band 7, NHS England.	
	Home visit per minute	£1.41		Home visits (1:0.45) Patient work (1:0.20)	

# *Table 1 Unit costs used for costing primary care and community services*

# *Table 2 Complete cases*

		Complet	e Cases	
		FNP	Usual Care	
		(n=808)	(n=810)	
	EQ-5D Baseline	808 (100%)	807 (99%)	
	EQ-5D 34-36 weeks	614 (76%)	616 (76%)	
	EQ-5D 6 months	507 (63%)	469 (58%)	
	EQ-5D 12 months	510 (63%)	480 (59%)	
	EQ-5D 12 months	499 (62%)	465 (57%)	
	EQ-5D 24 months	583 (72%)	537 (66%)	
	EQ-5D all assessments	349 (41%)	265 (34%)	
	GP records	480 (61%)	471 (60%)	
	Hospital data (HES records)	808 (100%)	810 (100%)	
	Complete-Case dataset ^	217 (28%)	186 (24%)	
458				l ll mothers whose all six EQ-5D-3L
	-		-	
459	assessments and all costs (GP rec	ords, nearth visito	r/midwife visits	and hospital attendances) were available.
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	FNP (n=782)			Usual Care (n=786)						
	n	Mean (SD)	Min, Max	Median	Missing	n	Mean (SD)	Min; Max	Median	Missing
GP surgery visits for mothers	471	9.55 (8.40)	0; 48	7	39.76%	480	8.49 (7.81)	0; 48	7	38.93%
GP home visits for mothers	471	0.22 (0.84)	0; 9	0	39.76%	480	0.21 (0.76)	0; 8	0	38.93%
Nurse surgery visits for mothers	471	2.14 (3.61)	0; 36	1	39.76%	480	2.22(3.01)	0; 20	1	38.93%
Community Midwife visits	459	10.40 (5.34)	0;41	10	41.30%	422	10.68 (5.25)	0; 41	10	46.31%
Community Health visitor visits	363	8.60 (13.74)	0; 68	0	53.58%	321	16.25 (12.15)	0;73	13	59.16%
Community Counsellor visits	612	0.29 (1.23)	0;12	0	21.73%	614	0.32 (1.64)	0;20	0	21.88%
Inpatient Length of Stay mothers	782	3.98 (6.35)	0; 99	3	0%	786	4.09 (6.39)	0;110	2	0%
Day case admissions for mothers	782	3.53 (5.19)	0; 60	2	0%	786	3.57 (5.48)	0;77	2	0%
Outpatient visits for mothers	782	8.61 (8.05)	0; 74	7	0%	786	8.55 (8.05)	0; 70	6.5	0%
A&E attendances for mothers	782	4.54 (2.43)	0; 36	1	0%	786	1.58 (2.55)	0; 29	1	0%
GP surgery visits for babies	471	8.17 (7.10)	0; 70	7	39.76%	480	7.60 (6.20)	0; 51	7	38.93%
GP home visits for babies	471	0.29 (1.25)	0; 17	0	39.76%	480	0.29 (1.61)	0; 20	0	38.93%
Nurse surgery visits for babies	471	0.88 (2.17)	0; 22	0	39.76%	480	0.90 (2.05)	0; 18	0	38.93%
Inpatient length of stay for babies	724	2.82 (21.32)	445 ;0	0	0%	757	3.10 (25.29)	0; 466	0	0%
Day case admissions for babies	724	1.74 (3.42)	0;34	0	0%	757	1.79 (3.31)	0; 32	0	0%
Outpatient visits for babies	724	1.82 (5.29)	0;69	0	0%	757	2.08 (7.03)	0; 135	0	0%
A&E attendances for babies	724	2.58 (3.24)	0; 30	2	0%	757	2.21 (2.53)	0; 15	1	0%
FNP visits/encounters	709	39.28 (15.19)	1; 88	41	10%	10	0.45 (4.26)	0; 53	0	NA
FNP phone calls	709	6.29 (5.34)	2; 31	4	10%	10	0	0	0	NA

# Table 3 Mothers and babies average resource use per arm of the trial from baseline up to two years following child birth

SD, standard deviation

	Mean cos	st £ (SD)	Difference (FNP - Usual Caro) (95% CI)		
	FNP	Usual Care	Care) (95% CI)		
GP surgery visits for mothers	429.95 (17.49)	382.35 (16.20)	47.60 (0.82; 94.37)		
GP home visits for mothers	25.82 (4.42)	24.92 (4.18)	0.89 (-11.96; 12.84)		
Nurse surgery visits for mothers	21.17 (1.69)	22.63 (1.41)	-0.83 (-5.17; 3.50)		
Community Midwife visits	15.51 (332.38)	15.29 (314.19)	-20.96 (-63.82; 21.89)		
Community Health visitor visits	135.67 (11.34)	217.78 (10.25)	-82.10 (-112.42; -51.78)		
Community Counsellor visits	16.86 (71.56)	19.08 (94.79)	-11.62; 7.20)		
Inpatient length of stay mothers	6354.57 (8460.72)	6661.17 (9679.04)	-306.59 (-1193.20; 580.00)		
Day case admissions for mothers	775.22 (1041.62)	781.72 (1216.929)	-6.50 (-116.98; 103.96)		
Outpatient visits for mothers	889.49 (903.30)	875.63 (918.99)	13.85 (-75.01; 102.71)		
A&E attendances for mothers	167.06 (277.82)	172.79 (289.34)	-5.72 (-33.39; 21.93)		
GP surgery visits for babies	367.74 (14.92)	342.37 (12.80)	25.36 (-13.13; 63.86)		
GP home visits for babies	33.13 (6.64)	32.64 (8.38)	0.49 (-20.57; 21.56)		
Nurse surgery visits for babies	8.96 (1.03)	9.21 (0.95)	-0.25 (-3.00; 2.50)		
Inpatient length of stay for babies	3773.35 (25939.83)	4882.99 (50019.88)	-1109.634 (-5198.32979.6)		
Day case admissions for babies	142.02 (702.62)	145.17 (615.10)	-3.15 (-70.38; 64.08)		
Outpatient visits for babies	290.96 (983.72)	272.92 (842.08)	-18.03 (-111.56; 75.49)		
A&E attendances for babies	293.12 (370.60)	254.93 (298.36)	38.16 (3.94; 72.38)		
FNP visit/encounters	3845.32 (76.69)	47.27 (16.08)	3798.05 (3644.70; 3951.4)		
FNP phone calls	33.27 (2.84)	0 (0)	33.27 (27.70; 38.83)		

Table 4 Costs associated with all available cases: mean and standard deviation (in brackets). Costs discounted from year 2 at 3.5% accordingto ITT. Mean incremental costs and 95% CI estimated using OLS regression

SD, standard deviation

Analysis	Difference in costs <sup>*</sup>	Difference in QALYs*	ICER for FNP intervention (£ per QALY)	Probability FNP Cost-effective† £20 000/QALY
Base case (MI)	1812 (-2814; 5447)	0.0030 (-0.01; 0.02)	Above £100,000 per QALY	17%
Sensitivity i (CC)	4549 (3175; 5922)	-0.007 (-0.042; 0.027)	FNP dominated	0%
Sensitivity ii	1933 (-2641;5654)	0.005 (-0.017; 0.027)	Above £100,000 per QALY	16%
Sensitivity iii	2061 (-1949; 6072)	0.005 (-0.014; 0.025)	Above £100,000 per QALY	17%
Sensitivity iv	3272 (2288; 4295)	0.004 (-0.018; 0.025)	Above £100,000 per QALY	0%
Sensitivity v	360 (-3680; 4352)	0.005 (-0.013; 0.022)	£73,924 per QALY	45%

Table 5 Summary of incremental analysis (ITT), cost-effectiveness results and uncertainty for the base case (highlighted) and sensitivity analyses